

**Making the “Miracles” Happen – Parenting Stress and Experiences
among Parents of Extremely Preterm Young Adolescents in
England: A Convergent Mixed Methods Study**

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Declaration

I, Emmi Maria Suonperä, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Signature:.....  7 November 2021

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Abstract

Caring for a child born at an extremely low gestational age places stresses on parents in the context of worries about the child's mental and physical development. This convergent multistrand mixed methods inquiry aimed to improve our understanding of experiences of parents of children born extremely preterm (EP) entering adolescence. In the systematic narrative review of literature, I collated findings from current publications assessing long-term parent outcomes following preterm birth. The empirical data comprised qualitative semi-structured telephone interviews with parents of EP children and cross-sectional survey data of parenting stress among parents of EP and full-term born children as they transitioned into adolescence at 11 years of age, collected as part of a longitudinal national birth cohort study of extreme prematurity in England (EPICure 2). The findings were reflected against the modern Western understanding of parenting, namely the framework of parental determinism, to place them in a wider sociocultural context. Three hundred parents (175 EP parents and 125 full-term parents) completed a postal Parent Questionnaire and 22 parents of EP children participated in an interview. The data were analysed statistically and thematically as appropriate and the outcomes were narratively integrated. The systematic review results indicated that the research on long-term parent outcomes following EP/VP birth had methodological limitations and the findings were inconclusive. Yet publications reported a trend towards increased family impact among families with adolescents born preterm. EP parents reported higher levels of parenting stress in comparison with the parents of full-term born children. High parenting stress among EP parents was associated with younger child age, child attendance at a special educational needs school or unit, and higher parent educational level. Parental ambitions and the parents' perceptions of their child's ability to reach adult independence guided how they described their parenting behaviour. Parents who anticipated their children to have challenges with future independence described varying parental behavioural responses to support their child's development in accord with parental determinism. Findings from this study suggest that wider social factors influence parenting stress and experiences, and therefore affect the way in which parents approach their roles. This study has further directed attention to the health and well-being of parents who care for children with long-term morbidities.

Keywords: Extremely preterm; Long-term outcome; Adolescent; Parenting stress; Parenting Culture; Mixed methods research

Impact Statement

This mixed methods study aimed to improve our understanding of parenting a child entering adolescence among groups of parents with or without the experience of extremely premature birth. Previous research has established that parents of EP children experience higher levels of parenting stress when their children are young in comparison with parents of full-term born children. Parenting stress is linked to family health and child development. Few studies have explored parent outcomes when the preterm born children reach adolescence, encounter new developmental expectations, and strive for greater independence. Parents have a pivotal role in supporting their children at this time of transition, which may provoke parenting stress. Few studies have explored these topics from the parents' perspectives. This study will directly address these gaps in knowledge. Its intended impact is to improve our understanding of parenting an EP young adolescent, in order to be able to support parents more effectively, improve their parenting abilities and wellbeing, family functioning, and children's outcomes.

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Chapter 1 Introduction and Background

1.1 Study Summary

“Parents and the home environment they create are the single most important factor in shaping their children's well-being, achievements and prospect” (Department for Education and Skills, 2007, p. 1)

From the 20th century onwards, in scientific literature, parenting behaviour has been regarded as a key factor promoting a child's cognitive and social development (Apple, 1995; Maccoby, 1992). Influences of individual parent or child factors and family or environmental factors on parenting behaviour and the parent-child relationship have been investigated (Kuppens & Ceulemans, 2019). In this context, parenting research among prematurity has focused on parenting behaviours that may promote or inhibit optimal child development following the risk exposure of preterm birth. Alongside this, in the modern Western sociocultural context, the increased research attention towards parenting behaviour has been paralleled with a wider social interest towards ‘parenting’ and ‘parenthood’ (Lee, Bristow, Faircloth, & Macvarish, 2014, p. 1-9; Macvarish & Lee, 2019; Smith, 2010). As opposed to a family-based relatively private responsibility to ‘guide’ the offspring from infancy to adulthood, in a Western contemporary dominant discursive understanding of parenthood, modern parenting has become more demanding and time consuming than before (Lee et al., 2014, p. 51-101). Scholars have, for example, noted that parents are increasingly intensively involved in their adolescent children's lives (Schiffrin et al., 2013). In this thesis, I will argue that the modern discursive framing of parenting has influenced the way in which parenting behaviour has been investigated in the research field of prematurity. I will showcase that this social understanding is also evident in parental experiences of parents of extremely preterm (EP) born young adolescents (YA). In the context of parenting research, I will propose that to focus primarily on the impact that parenting behaviour may have on child outcomes may divert research attention away from factors that may impact parental wellbeing.

In this convergent multistrand mixed methods inquiry (Teddlie & Tashakkori, 2003, p. 688; 2006, p. 21), the empirical data comprised qualitative semi-structured telephone interviews with 22 parents of EP YA, and cross-sectional survey data of parenting stress, sociodemographic, child and parent health factors among parents of EP and full-term (FT) born YA, collected as part of a longitudinal national birth cohort study of extreme prematurity in England. The present study not only examined the parent outcomes in this population, but also conveyed perspectives of parents to situate the outcomes in lived experiences. Thus, it

aimed to improve both the “depth and breadth” (Bazeley, 2018, p.12) of current understanding of parenting. By exploring these issues, this unique study contributed to identifying challenges in bringing up an EP child, and key issues and anxieties facing families of EP children as they transitioned to adolescence.

From a philosophical viewpoint, this study referred to pragmatism in that for this research project the most important consideration in understanding parental experiences was ‘workability’ (Morgan, 2007); how parents explained or understood the social realities of their ‘parenting’. Thus, in a theoretical sense, the present study was most interested in the social construction of ‘parenting’ that occurred in the research participants’ accounts of their experiences when they discussed their parenting roles, actions, and decisions.

In this introductory chapter, I will discuss the general understanding of parent-child relationship and the concept of ‘parenting’. I will explain how this parent-child relationship may be altered as a consequence of EP birth. I will critically evaluate the potential contribution of the theory of parental determinism in understanding the experiences of parents of EP YA. Differences between concepts of ‘parenting’ and ‘parent outcomes’ will be discussed and parent outcomes over time among parents of children born preterm will be presented. The EPICure studies will be presented, as these were the research context in which this mixed methods study was situated. General outcomes of interest among EP children and social representations of prematurity will be explored. This introductory chapter will culminate in research aims and questions.

1.2 Parent-child Relationship and Parenting Behaviour

Parent-child relationship can be understood as a bidirectional interaction process that fosters development of the child’s cognitive and socio-emotional abilities and self-regulation (Feldman, 2007; Kotchick & Forehand, 2002; Maccoby, 1992). Parenting behaviour in the parent-child dyad, such as parental sensitivity or responsiveness (e.g., warm affective physical and visual contact and consistent appropriate respond to child’s communicational signals (Feldman, 2007)), a level of parental protection (e.g., facilitation/promotion of age-appropriate child independence (Thomasgard & Metz, 1997)) and/or behavioural control (e.g., control of child behaviour with rules, supervision and/or reward (Kuppens & Ceulemans, 2019)), impacts child development and social behaviour through to adulthood (Kuppens & Ceulemans, 2019). As the nature of the parent-child relationship is reciprocal, both parent and child factors are influential (Maccoby, 1992). Parental reactions to child characteristics (actual or perceived) may generate a feedback loop (Harris, 1995); for example, parental perception

of child vulnerability (PPCV) may provoke protective parenting behaviour (Tallandini, Morsan, Gronchi, & Macagno, 2015). High level of parental protection may in return promote child dependence on the parent (Thomasgard & Metz, 1997). A comparability between parent and child characteristics, such as matching temperaments, may impact the relationship by altering the reciprocal system/responsiveness within the dyad (Harris, 1995). Parental perception of difficult infant temperament has been associated with reduced personal growth in transition to parenthood among first-time mothers of 1-month-old preterm born infants (mean gestational age (GA) 33.7 weeks) (Spielman & Taubman-Ben-Ari, 2009). Parental factors, such as psychological and/or physical illness may also impact the dyadic interaction by reducing the reciprocity between the parent and the child. A parent with depressive symptoms may perceive the parenting role as demanding and respond less frequently and slower to child's communicative cues (Feldman, 2007). Positive parental mood may promote child compliance (Maccoby, 1992). Thus, the process is bidirectional.

Factors outside the parent-child dyad may also impact the interaction. In the family, characteristics of one child may influence the way in which the parent approaches the sibling/s (Maccoby, 1992); the child perceived as less vulnerable may be pushed to behave more independently earlier. Interactive sensitivity of one parent may support the parent-child interaction with the other parent by promoting the child's social responsiveness (Feldman, 2007). Parenting behaviour is also impacted by parents' peer groups (Harris, 1995) and contextual factors such as cultural values and beliefs (Kotchick & Forehand, 2002). For example, at what age parents let their child walk to school alone, may be influenced by other parents' decisions, cultural practices (e.g., start of secondary school may be regarded as the 'correct' time) and/or characteristics of the neighbourhood such as safety (Kotchick & Forehand, 2002).

Achievement of individual goals in the parent-child relationship, such as increased child autonomy or parent role satisfaction, are dependent on synergy between the parties (Maccoby, 1992). Yet parents have greater interaction skills and therefore greater ability to adapt their parenting behaviour to respond to their child's characteristics (Harris, 1995). The parent-child relationship develops over time with cumulative dyadic experience (Thomasgard & Metz, 1997) but also from parental experience of interaction with possible siblings (Harris, 1995). Parental authority, such as protection or/and control, generally declines over time along with increasing child independence, reducing the power asymmetry in the relationship. Thus, parental experiences in the parent-child relationship are co-constructed together with the child and reconstructed continuously through child development (Harris, 1995; Maccoby, 1992).

The bidirectional interlinkages between varying factors, parent-child relationship, and parenting behaviour are illustrated in Figure 1-1 below.

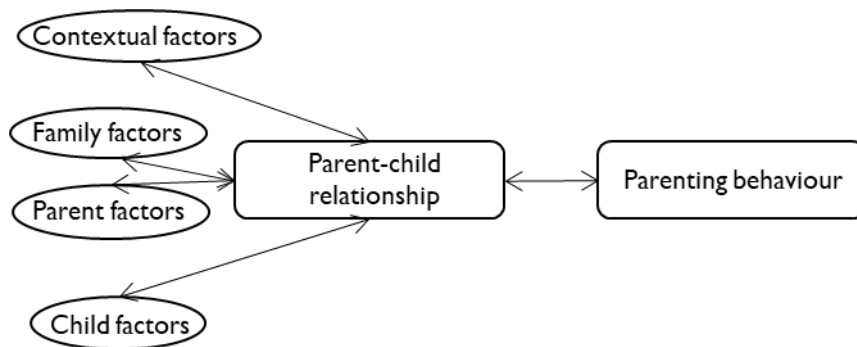


Figure 1-1 Interlinkages between factors, parent-child relationship, and parenting behaviour

Arrows linking varying factors, parent-child relationship, and parenting behaviour represent bidirectional impact between them

Parenting behaviour has received extensive research attention since the early 20th century. A few overarching theories on child socialisation have emerged during this time (Maccoby, 1992). In psychoanalytic theory, children were perceived to have “primitive impulses” (Maccoby, 1992, p. 1008) that parents brought under control by teaching children to behave in a socially acceptable manner. The effects of early childhood experiences were regarded as permanent, although their manifestations could be altered in adulthood. Later, attachment theory emphasised the reciprocal behaviours between the primary caregiver and the child. The primary caregiver instinctively responded to the infant’s cues, who in return responded to the parent’s behaviour. A secure and trusting attachment relationship in infancy was seen to support subsequent child development. Children internalised the quality of the attachment relationship, which was then recreated in future adult relationships (Harris, 1995; Maccoby, 1992). Both the psychoanalytic framework and the attachment theory are influential today, and have, for example, stimulated a research interest towards the association between early parent-child relationship and long-term child outcomes (Macvarish & Lee, 2019).

More recently, studies have focused on particular parenting behaviours and variations in parenting practices in varying socio-cultural contexts such as ethnic groups or social classes (Gillies, 2008; Kotchick & Forehand, 2002; Lareau, 2002). Distinct, although related concepts such as parenting practices (observable parenting behaviours), parenting dimensions (related parental behavioural variables such as ‘responsiveness’) and parenting styles (combinations of parenting practices) have been examined both theoretically and empirically (Kuppens &

Ceulemans, 2019). For example, Baumrind's (1973) parenting styles, further developed by Maccoby and Martin (1983), as neglectful (low control and warmth), authoritarian (more control than warmth), permissive (more warmth than control) and authoritative (control and warmth) parenting behaviour have become well accepted constructs (Kuppens & Ceulemans, 2019). Authoritative parenting style has been shown to correlate positively with adolescent outcomes, such as social competence and academic achievements, among general population samples (Steinberg, Lamborn, Darling, Mounts, & Dornbusch, 1994). Although, in these earlier studies adolescent outcomes were generally examined after placing parents in predefined categories of parenting behaviour rather than allowing varying parenting styles to emerge inductively from data (Kuppens & Ceulemans, 2019).

Further, concepts such as 'scaffolding' emerged in parenting research. As an alternative to a framework in which parental reward or punishment promoted child learning, the parent's role was to provide an ideal surrounding, i.e., scaffolding, for child development and to 'guide' child learning (Mermelshine, 2017). In all these conceptualisations, 'effective' and 'ineffective' parenting behaviour were defined based on the level of child socialisation (Maccoby, 1992). Indeed, developmental psychology has been a prominent framework to understand child development and its influence has been visible in the conceptualisation of 'appropriate' parenting behaviour (Gillies, 2008; Ribbens McCarthy & Gillies, 2017). Appropriateness of parenting behaviour depended on the child's developmental status; a low level of parental control was regarded optimal in adolescence to promote age-appropriate independence but would have yielded different outcomes in early childhood. A unifying feature of varying framings of optimal parenting behaviour was parental commitment to the parent role. Parenting was understood as an altruistic task. In her early review of history of parenting research, Maccoby (1992) concluded that parenting called for "considerable effort and skill" which implied that parents "accepted almost unlimited, long-term commitment to promoting the child's welfare" (Maccoby, 1992, p. 1015).

1.3 Cultural Determinants of Parenting Behaviour

When the purpose of being a parent is regarded as child socialisation (Maccoby, 1992) then the task of parenting could be regarded as an inherently goal-oriented set of actions. Thus framed as 'doing' rather than 'being' (Lee et al., 2014, p. 7-8; Smith, 2010). In fact, since the 1970s, in public Western discourse, 'being a parent' has been increasingly referred to as actions that parents do for their child (e.g., the emergence of a verb 'parenting' opposed to 'raising a child' or 'childrearing') rather than as an identity or an interpersonal relationship with the child (Gillies, 2008; Ribbens McCarthy & Gillies, 2017; Smith, 2010). Within this

framework many authors have examined the determinants of parenting behaviour (Abidin, 1992; Deater-Deckard, 1998; Miles & Holditch-Davis, 1997). I will discuss these models later in this introductory chapter. In addition to child, parent, and family factors sociocultural context may also impact parental values and beliefs about parenthood and therefore, impact parenting behaviour (Kotchick & Forehand, 2002; Ogbu, 1981).

In his influential essay, anthropologist Ogbu, J. U. (1981) discussed the development of child competence defined as an ability to perform culturally valued tasks. An ability referred to a set of functional or instrumental skills needed to perform the valued tasks. According to him, cultural schemas or “folk theories” (p. 420) consist of shared values and beliefs that provoke a group of individuals to act towards shared goals. Thus, child competence, for example, may not be regarded as a stable set of physical, emotional, and social skills but as a cultural construct of characteristics that are needed to fulfil an adult role in a particular context (i.e., to “make it” in the society (p. 410)). The cultural model of an adult role can be a determinant of parenting behaviour. Ogbu (1981) argued that parenting behaviour is an ever-evolving culturally learned system of techniques that are effective in “transmitting” (p. 415) adult competencies to the next generation. Thus, parenting behaviour is likely to focus on skills that are perceived as useful to ‘function’ in a particular context; for example, parents may promote early independence in an unsafe neighbourhood (Kotchick & Forehand, 2002) or value their adolescent children’s confidence building among working-class families (Gillies, 2008). Thus, parenting behaviour may not be merely influenced by factors such as socio-economic status (SES), parent psychological health, child disability, and/or previous life events. Factors such as SES and related parent educational level, in addition to being proximal parent factors, may also reflect the sociocultural context of parenting. Studies have suggested that improved parent outcomes such as lower levels of parenting stress among well-educated parents may indicate parents’ improved ability to respond to their children’s behaviours (Almogbel, Goyal, & Sansgiry, 2017). Yet these families may also live in an environment in which worrying about factors such as child behaviour and neighbourhood safety are less relevant, or on the contrary, the cultural context may be less accommodating of even mildly differing child behaviour due to a highly competitive cultural context.

Ogbu (1981) also introduced a concept of ‘alternative competencies’. In their role, parents may value promoting alternative child competencies in relation to a dominant majority culture. Ogbu himself explored this concept in relation to what child competencies black ethnic minority parents valued in their parenting behaviour as a response to what was valued in the surrounding dominantly white American culture. Alternative competencies may also appear among working-class versus middle-class parents (Gillies, 2008). Varying ‘groupings’ such

as these, may appear within and between cultures (Kotchick & Forehand, 2002; Suizzo, 2016). For example, parents of children with chronic conditions may also reflect their childrearing goals and perceptions of child competence against what is valued among parents of healthy children (Landsman, 1998) and perhaps construct ‘alternative competencies’.

1.4 Preterm Birth and Altered Parent-child Relationship

Evidence indicates that children with chronic conditions may interact differently with their parents in comparison with healthy children and their parents (Holmbeck et al., 2002). It is generally understood that this altered parent-child interaction may affect child development and social-emotional behaviour long term (Deater-Deckard & Bulkley, 2000). Increased medical risk factors associated with EP birth may challenge early parent-child interaction. For example, preterm born infants may be less responsive, have reduced alertness, weaker cries and look at their parents less frequently (Deater-Deckard & Bulkley, 2000; Feldman, 2007; Montirosso, Borgatti, Trojan, Zanini, & Tronick, 2010). Parents may respond to this with reduced physical and visual contact and smile less frequently at their child (Deater-Deckard & Bulkley, 2000; Forcada-Guex, Borghini, Pierrehumbert, Ansermet, & Muller-Nix, 2011). Child non-engagement may promote intrusive parenting behaviours such as increased verbal control, toy stimuli, and/or physical management of the child to promote play (Feldman, 2007). The parent-child interaction may be further challenged by parental distress following preterm birth (Feldman, 2007; Forcada-Guex et al., 2011). Feldman (2007) examined the impacts of infant risk (prematurity or intrauterine growth restriction) and maternal risk (depression and/or anxiety) on parent-infant interaction (reciprocity and intrusiveness) among 145 couples and their 4-month-old children. Feldman (2007) concluded that the infant risk had highest negative impact on parent-child interaction. Negative infant emotionality promoted parent intrusiveness and decreased parental sensitivity. Yet, the lower impact of maternal depression and/or anxiety on parent-child interaction in this study may have been influenced by highly controlled contextual and family risk factors (SES, social support level, paternal mental health, single parenthood) in the study design (Feldman, 2007).

Altered parenting behaviour due to child chronic condition/s may persist from infancy into childhood (Deater-Deckard & Bulkley, 2000) and carry over to early adolescence (Holmbeck et al., 2002). Among EP populations, increased neurodevelopmental disability and behavioural problems such as attention deficit, hyperactivity disorder symptoms, and limitations in social competence (Johnson & Marlow, 2017) may place further demands on the parent-child relationship long term. If parents perceive their children’s behaviour as demanding they may utilise increased parental psychological control, such as criticisms, guilt induction, and/or

love-withdrawal to promote child compliance (Holmbeck et al., 2002). Parents may use 'scaffolding' to attempt to sustain their child's attention by increasing verbal control and direction (Jaekel, Wolke, & Chernova, 2012). Altered parenting behaviour may in return impact outcomes such as stress and/or anxiety among the parents. In Chapter 2 of this thesis, I will examine long-term parent outcomes following extremely or very preterm (VP) birth reported in literature.

In an early prospective longitudinal cohort study of prematurity, Beckwith et al. (1992) examined the continuity of maternal responsiveness from infancy to early adolescence and child outcomes among 44 mothers and their preterm born children (mean GA 33.2 weeks). The study protocol included parent, child, and teacher self-report questionnaires, as well as observations of mother-child interaction. A responsive parent was defined by comparing parent behaviour against predefined characteristics of parental responsive behaviours varying from less responsive behaviours (hostility, criticism) to more responsive behaviours (clarity, consistency, support of child autonomy). Child sex or number of previous children did not impact maternal responsiveness. Mothers with a higher education level tended to be more responsive in comparison with mothers with lower educational attainment. At the age of 12 years, preterm born children of responsive mothers had higher IQs, reported less family conflict, perceived themselves as more intelligent, but not socially more competent than children with less responsive mothers. Also, their teachers rated them as having fewer behavioural issues. Yet children whose mothers became more responsive over time scored similarly in cognitive assessments at the age of 12 years in comparison with children whose mothers were defined as responsive since infancy (Beckwith et al., 1992).

In addition to clinically defined child morbidity, parents of preterm born children may perceive their children as vulnerable following the high-risk birth and uncertainty around infant survival (Miles, Holditch-Davis, & Shepherd, 1998). Perceived child vulnerability may promote increased parental protection (Thomasgard & Metz, 1997). Parents of 3-year-old children born preterm (mean GA 28.2 weeks) described challenges in limiting their children's behaviour and granted them with 'special treatment' due to the experience of preterm birth (Miles et al., 1998). The concept of 'vulnerable child syndrome' was first described by Green and Solnit in (1964) as an unwarranted parental perception of high child medical risk provoked by previous child illness or injury that had since resolved. Thomasgard and Metz (1997), when developing their widely used survey to assess parental overprotection, examined parent perceived child vulnerability and parental protection among 290 parents of five to 10-year-old children recruited from primary care settings. Parent reported child prematurity, child medical condition, pregnancy complications, or previous child illness or life-threatening injury were

not associated with perceived child vulnerability. In a similar study of children aged two to five years, the same authors found these factors to be associated with parent reported child vulnerability. The authors suggested that the potential shift in parental perceptions over time might have been impacted by continuous support from healthcare professionals over the years, cumulative dyadic experience in the parent-child relationship, and increased child ability to express concerns at older ages (Thomasgard & Metz, 1997). A more recent meta-analysis of factors impacting parental perception of child vulnerability among parents of preterm born children concluded that maternal psychological health was more impactful in promoting perceptions of child vulnerability compared to child physiological health (Tallandini et al., 2015).

Parental perception of child vulnerability may be associated with overprotective parenting behaviour (Thomasgard & Metz, 1997). Overprotective parenting behaviour has been defined as excessive parental protection in relation to the child's developmental stage and abilities, which may delay child independence (Holmbeck et al., 2002; Thomasgard & Metz, 1997). Parental overprotection may be adaptive to the child's chronic condition/s. For example, both mothers and fathers of eight to 9-year-old children with spina bifida reported higher rates of parental overprotection in comparison with parents of healthy children. Parental overprotection correlated with increased child physical disability and lower child IQ (Holmbeck et al., 2002). Parental protection or monitoring in childhood may change the trajectory of development of independence and thus, alter adolescent behaviour. For example, studies among EP populations have suggested a link between perceived parental overprotection and increased anxiety and substance use disorders in adolescence/adulthood (Day, Dobson, et al., 2018; Day, Schmidt, et al., 2018). Yet these studies have largely relied on retrospective adolescent/adult self-reported assessment of parenting behaviour.

1.5 Parental Determinism in Research among Prematurity

As survival of EP children has continued to improve (Torchin, Morgan, & Ancel, 2020), the outcomes for EP children and the wider factors influencing these outcomes have become a major point of interest (Treyvaud, 2014). Many studies have moved their focus from the neonatal period to assessing the effects of factors such as parent-child interaction (Jaekel et al., 2012) or family environment (Greenley, Taylor, Drotar, & Minich, 2007) on long-term child outcomes. The increased research attention towards parenting behaviour is paralleled with a wider social interest towards 'parenting' and 'parenthood' (Lee et al., 2014, p. 1-9). Yet general process models of parenting behaviour have focused more on proximal child, parent, and family factors' influences on parenting behaviour, and less commonly examined the

effects that the wider social and cultural context such as neighbourhood safety (Kotchick & Forehand, 2002) or ethnic minority status (Ogbu, 1981) may have on individual parenting behaviour, or/and how discursive framings of parenthood may appear in parents' perceptions of their roles and thus, impact parenting behaviour (Eerola, Närvi, Terävä, & Repo, 2021; Sevón, 2011; Widding & Farooqi, 2016)

As opposed to a family-based relatively private responsibility to 'guide' the offspring from infancy to adulthood, in a Western contemporary dominant discursive understanding of parenthood, modern parenting has become a subject of expert knowledge and extensive public debate (Lee et al., 2014, p. 51-101; Macvarish & Lee, 2019; Smith, 2010). To support the child's optimal physical, mental, and emotional development parents are required to have more knowledge and resources than before. Instead of 'being', parenting is framed as 'doing'; it is a skillset that must be learnt (Apple, 1995; Lee et al., 2014, p. 7-8; Smith, 2010). Similarly to other skills in life (e.g., driving a car), to acquire the 'parenting skillset' parents must "work out how, look up the solution, consult the manual, go to classes, ask an expert" (Smith, 2010, p. 362). A key aspect of this dominant discursive understanding of parenting has been parental determinism; the concept that parental behaviour has direct causal impact on future success for the child. Parenting has been framed as the single most important set of actions ensuring the child's future competence as an adult. Thus, parental determinism can be regarded to dominate the wider social context in which contemporary Western parents act (Dermott, 2012; Füredi, 2008, p. 55-61; Lee et al., 2014, p. 6-7). In the present study, I will explore parental experiences of parents of YA born EP within the framework of parental determinism (PD).

The framework of PD (please see Figure 1-2 below) suggests that parenting behaviour can either promote or challenge the parent-child relationship, that then affects child outcomes long term (Kanieski, 2010). Thus, parental determinism, as a contextual factor, a sociocultural framing of 'parenting', is linked to parenting behaviour. In the reciprocal system, the child is perceived as vulnerable; at risk of unknown external threats, such as accidents or unfamiliar adults, potentially endangering the immediate parent-child relationship (Füredi, 2008, p. 26-27; Lee et al., 2014, p. 10-16). Simultaneously, the child passively faces risks of parenting behaviour that does not support (or hinders) optimal child development, such as overprotective parenting (Wall, 2021). Thus, parenting behaviour can at the same time be a protective factor as well as a risk (Kanieski, 2010). Parenting behaviour can be utilised to mitigate other risks in child development (Macvarish, Lee, & Lowe, 2014), such as EP birth may present. Parental determinism and the need for risk management together promote intensive parenting practices as parents are required to do more to support their child's optimal health and development, ensure their social-emotional competence later in life, and to avoid 'risks' (Clarke, 2013; Lee

et al., 2014, p. 25-31). Thus, intensive parenting as “child-centred, emotionally absorbing, labour intensive, and financially expensive” (Hays, 1996, p. 8) parenting practices relies on expert knowledge on what kind of parenting behaviour is ‘effective’ in promoting optimal child outcomes (Apple, 1995; Clarke, 2013; Hoffman, 2010). The aim to optimise long-term child outcomes rationalises/justifies early parent-child intervention programmes (Kanieski, 2010; Macvarish et al., 2014) and magnifies parental responsibility for child outcomes over considerations of other potentially impactful factors such as child genetic factors or/and chronic condition/s, the community, the school or the surrounding social and economic structure (Clarke, 2013; Macvarish & Lee, 2019; Taylor, Spencer, & Baldwin, 2000) such as the healthcare system (Saigal, Pinelli, Streiner, Boyle, & Stoskopf, 2010). In what follows, I will further explore the framework of PD as presented in Figure 1-2 below using relevant research examples from the research field of prematurity.

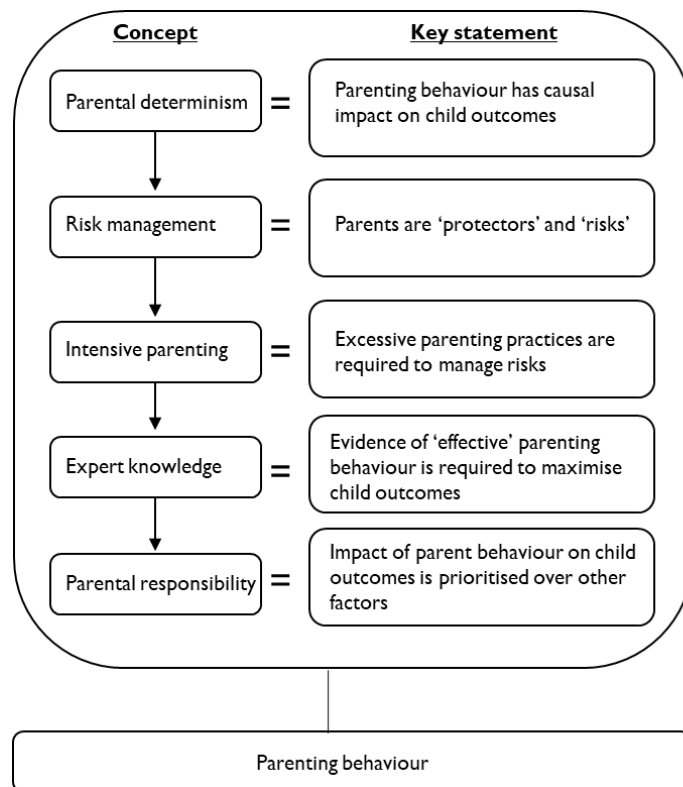


Figure 1-2 Conceptual framework of parental determinism and its link to parenting behaviour

Arrows linking varying concepts represent the line of thought between them

1.5.1 Parental determinism in attachment and early childhood discourses

A basis for a long-lasting impact on parent and child outcomes from an adverse birth experience and neonatal illness, and the importance of the early childhood period, emerge from

attachment theory and the psychoanalytic framework of the impact of the child's early experiences on later life (Maccoby, 1992; Ogbu, 1981; Thornton, 2011). The concept of parental determinism is central to discourses on attachment and early childhood as in them parent behaviour, independently of child characteristics, can either promote or challenge secure attachment and the parent-child relationship. Thus, it can impact child outcomes (Kanieski, 2010).

As discussed earlier, attachment theory, originally developed by Bowlby and Ainsworth in the 1950s and 1960s, is based on the critical importance of the attachment of a child to their primary caregiver. Attachment is viewed as a biological phenomenon based on cross-species evolutionary logic. Secure attachment requires both an emotional bond as well as physical proximity such as touching, hugging, or cuddling. Thus, attachment is impacted both by the child and parent factors (Bowlby, 2005, p. 5-10; Macvarish et al., 2014). Within this framework, the parent-child attachment experience itself may form a risk (insecure attachment may lead to child problem behaviour and hindered development) as well as be a protective factor (securely attached children may eventually be successful in their social and academic lives) (Kanieski, 2010). Parents have causal impact on their children's outcomes as they may prevent, with appropriate parenting behaviour, other risks for insecure attachment (Yerkes, Hopman, Stok, & De Wit, 2019) such as EP birth and prolonged neonatal intensive care unit (NICU) stay may present. For example, in the NICU context, increased efforts have been made to support parent-infant attachment, such as the promotion of skin-to-skin care (Charpak et al., 2017) and parents' involvement in their infant's care (Shaw et al., 2019). However, as secure attachment also requires the emotional bond, parental (commonly maternal) emotions, such as stress or anxiety, may risk attachment and child outcomes (Hoffman, 2010; Macvarish & Lee, 2019). Yet studies that have assessed parent and child attachment experiences among preterm born populations have found modest, if any, evidence of increased insecure attachment organisations in comparison with full-term born children and their parents, unless other parent or child risk factors, such as mental health disorders or bronchopulmonary dysplasia (BPD), have been present (Charpak et al., 2017; Hallin, Bengtsson, Frostell, & Stjernqvist, 2012; Korja, Latva, & Lehtonen, 2012).

Hallin et al. (2012) examined whether children born EP differed from those born full-term in their attachment organisation in late adolescence (mean age of 18 years) using the Adult Attachment Interview (George, Kaplan, & Main, 1996). The authors established the framework of risk of insecure attachment due to adverse birth experience and challenged parent-child interaction during early childhood. Overall, they found EP young adults to experience less secure attachment, that was not impacted by neonatal variables. The authors

noted that traumatic birth and deficits in child development may challenge attachment, but also that secure attachment may promote better child outcomes. They presented a balanced discussion acknowledging the lack of data on parent mental health outcomes or parent-child attachment at any other time point. Nevertheless, the authors recommended future research to focus more on parent-child interaction in early childhood following EP birth (Hallin et al., 2012).

Within the discourses of attachment and importance of early childhood, recent advances in neuroscience have linked parental determinism to child brain research (Macvarish et al., 2014); another particularly relevant matter in extreme prematurity. The time from late pregnancy till early years of childhood is characterised by rapid brain development. Among preterm born infants, the normal maturational processes in late pregnancy take place outside the womb, subjecting the infant's brain to external disturbances (de Kieviet, Zoetebier, van Elburg, Vermeulen, & Oosterlaan, 2012). A general reasoning states that parental (mostly maternal) emotions and actions (love/care) can reduce the infant's state of stress that, if continuous or/and severe, through physiological responses in the body would permanently alter infant brain development, and therefore, potentially challenge the child's emotional and cognitive abilities (Macvarish et al., 2014). When viewed like this, parent behaviour is not only causal, but also medicalised, as it is written into the child's brain, making parental 'love' physically measurable (Kanieski, 2010; Macvarish et al., 2014).

As an example, Treyvaud et al. (2020) examined the effects of "sensitive and intrusive parenting behaviours" (Treyvaud et al., 2020, p. 3) (a 10-minute observed mother-child interaction session assessing five parenting dimensions of positive and negative affect, sensitivity, facilitation, intrusiveness/over-control) at the child's age of two years on the child's brain volume and white matter microstructure at the age of seven years, and on overall brain growth from term equivalent age to seven years among VP born (<30 weeks' gestation) Australian children. The authors acknowledged the modest effect sizes but found a positive independent association between brain growth of VP born 7-year-olds and 'positive parenting' in early childhood. The authors noted that a low inter-rater reliability of negative parenting behaviour, potentially inconsistent parenting behaviour over time, and a potentially bidirectional relationship between parenting and child brain development (controlling parent behaviour may be a beneficial response to already altered brain development) may have reduced the validity of their findings. Nevertheless, they concluded on an aim of developing effective early intervention programmes and for "future studies to help better understand how parenting shapes the developing brain for at-risk children" (Treyvaud et al., 2020, p. 13).

As the research examples of Hallin et al. (2012) and Treyvaud et al. (2020) have illustrated, understanding parenting behaviour within the framework of parental determinism also appears in the research field of prematurity. In fact, I postulated that perceiving parenting behaviour as causal for child outcomes may be a particularly appealing framing in the context of extreme prematurity in which the parenting task (at least in relation to this particular child) begins with the risk exposure of adverse birth and prolonged hospitalisation. However, it is important to note that the parents' relationship with their child begins to develop before the birth (MacKinnon, 2006). Indeed, it has been argued that in some research fields such as epigenetics, 'parental responsibility', for example in relation to healthy maternal life choices, is framed as beginning prior to conception (Pickersgill, 2021; Valdez, 2018). Thus, the parenting task may be regarded to precede the birth. Nevertheless, the perception of early risk among EP parents may further emphasise the parents' 'protector' and 'mitigator' roles in their child's development from EP infant to competent adult. When parents are aware of their child's 'at-risk' status they may be perceived as morally responsible to act to reduce the risks (Kanieski, 2010). Following from this framing, parents may feel accountable to explain how they have mitigated risks in their children's lives (Faircloth, 2010). As a dominant discursive understanding of 'parenting', deterministic thinking may influence both parents' as well as professionals' perceptions of parental roles. I will explore these ideas further when discussing the findings from the interviews with parents of EP YA later in this thesis.

1.5.2 Parents as 'protectors' or 'overprotective' parents

Risk management as the key function and purpose of parenting (Lee, Macvarish, & Bristow, 2010) can be placed within a wider context of modern life as risk focused (Giddens, 2013, p. 119-128; Hoffman, 2010). The way risk is conceptualised has changed; risk is no longer a probability, odds of having a 'good' or a 'poor' outcome, but rather synonymous with 'danger' and 'bad'. Parenting behaviour can be regarded as a response to, or a way of mitigating the risks surrounding us, for example by increasing parental control and monitoring in children's lives to prevent accidents or injuries. Thus, parenting as risk management is an outcome of the overwhelmingly risk focused apocalypse threatened society we live in (Giddens, 2013, p. 9 & 119-128; Lee et al., 2014, p. 10-15). Evidence of understanding both parenting and risks in this manner can be found in research among prematurity; the increased interest towards parent behaviour can be regarded as a response to the risk consciousness as the research efforts are aimed at preventing/mitigating possible adverse outcomes. Following from this framing, effectiveness of parenting behaviour (i.e., level of child socialisation (Maccoby, 1992)) can be retrospectively measured based on whether 'risks' (i.e., poor outcomes such as anxiety disorders) have manifested (Day, Dobson, et al., 2018; Day, Schmidt, et al., 2018).

In two separate manuscripts, Day et al. (2018, 2018) examined retrospectively 1) whether overprotective parenting recalled at the age of 22-26 years mediated the presence of substance misuse and/or anxiety disorders at the age of 29-36 years among 81 Canadian adults born with extremely low birth weights (ELBW), and 2) whether the experience of overprotective parenting mediated association between the experience of peer victimisation and anxiety and/or substance use disorders among the same cohort of participants. The authors established the familiar framework of altered parenting behaviour due to adverse birth experience and parental perception of child vulnerability potentially altering long-term child outcomes. Limitations of both investigations included statistically modest associations between recall of parenting behaviour and later life outcomes among preterm born adults, significant attrition (>50%) of the study sample, and the potential for reverse causality between recall of parenting behaviour and anxiety/substance use or experience of peer victimisation. However, the authors concluded that their results aligned with previous studies showing that even though parents of preterm born children may feel they are protecting their children from harm, their actions may be “detrimental” (Day, Schmidt, et al., 2018, p. 913) to these children long term. They called for future research to “create and implement interventions that focus on parents of ELBW survivors” (Day, Schmidt, et al., 2018, p. 913).

Research narratives such as Hallin et al. (2012), Treyvaud et al. (2020), and Day et al. (2018; 2018) demonstrate the appeal of parental determinism over considerations of other potentially impactful factors such as developmentally supportive relationships that the child may have with other adults in the family or community context (Kotchick & Forehand, 2002; Macvarish & Lee, 2019). Research on parenting behaviour in general has framed child health as a principally parental responsibility, with less focus on other factors such as community, school, or the socio-economic surroundings as possible avenues for improving child outcomes (Clarke, 2013; Taylor et al., 2000). Thus, interventions have focused on the parent-child relationship (Gillies, 2008). Parents equipped with the appropriate ‘skillset’ are framed as ‘able’ to face the primary responsibility (Smith, 2010). Early parent-child intervention programmes have attempted to prevent adverse child outcomes by utilising individual parent behaviour as the medium (Macvarish & Lee, 2019). The concept of parental responsibility may be particularly relevant for parents with children with long-term health concerns, such as those born EP, as the parental role to support the child is further heightened (Holmbeck et al., 2002). In the present study, I will explore how parents of EP YA experienced parental responsibility.

1.5.3 The culture of intensive parenting

Parental determinism and the need for risk management have together promoted an intensive parenting culture (Smith, 2010) by following a rationale that earlier and greater investments towards a child's future are likely to yield greater returns (Gillies, 2020). Intensive parenting can be understood as a parenting culture that presumes that intensive parental involvement in children's lives is necessary to maximise the child's physical, cognitive, and social development (Hays, 1996, p. 9; Lee et al., 2014, p. 26-27; Schiffrin, Godfrey, Liss, & Erchull, 2014). Thus, it may impact parenting behaviour. The required intensive parenting practices can vary from optimal child diet and/or toys, books or games to stimulate 'brain growth' (Thornton, 2011) to adult organised and monitored extracurricular or social activities and tutoring to maximise academic success (Lareau, 2002; Schiffrin et al., 2014). Intensive parenting culture does not only entail increased parental involvement in children's lives but also includes the rationale of parental determinism. Thus, it is more than just intensive parenting practices, as it entails the belief that these actions are 'essential' to maximise child outcomes (Yerkes et al., 2019). Intensive parenting culture relies on five key assumptions that 1) in the parent-child dyad children are vulnerable and precious and that 2) generally mothers, not fathers, form the parent part of the dyad, 3) parenting is time consuming and 4) financially expensive, and that 5) parents should rely on expert knowledge of effective practices (Yerkes et al., 2019). If parental responsibility and support are heightened factors among parents of EP children due to the frequent developmental and chronic health concerns among preterm born children or due to parental perception of child vulnerability, parents may rely on intensive parenting practices to promote or compensate for their children's outcomes (Kantrowitz-Gordon, Altman, & Vandermause, 2016), for example to increase cognitive stimuli and parental control (verbal or non-verbal) (Wolke, Jaekel, Hall, & Baumann, 2013). Increased intensive parenting efforts may negatively impact parents' wellbeing (Wall, 2010).

Although empirical studies of the effects of intensive parenting practices on child outcomes are scarce, a general hypothesis has suggested that intensive parental involvement is adaptive during early childhood to promote child development, but may become less optimal among older children as it may hinder adolescent independence (Wall, 2021; Widding, 2014; Yerkes et al., 2019). For example, in a study of the impacts of 'helicopter parenting', 297 American adolescents in college who perceived their parents as 'over-controlling' reported more depressive symptoms and less life-satisfaction in comparison with adolescents who perceived their parents as less involved (Schiffrin et al., 2013) Yet, in studies like these, the direction of causality between parenting behaviour and adolescent outcomes could be reverse. In addition, intensive parenting culture presumes a so-called dose-response relationship between parenting

behaviour and child outcomes (i.e., more parental involvement will correlate with improved child outcomes) but this has not been demonstrated among general population samples (Widding, 2014; Yerkes et al., 2019). In contrast, in a prospective longitudinal cohort study of British families, 14-year-old adolescents rated their psychological wellbeing similarly regardless of whether their parents reported intensive parenting or ‘intermediate’ parenting practices (Yerkes et al., 2019).

Among a VP born population, Wolke et al. (2013) assessed the effects of sensitive parenting behaviour (observed parental verbal and non-verbal control, criticism, and parent-child harmony during a parent-child interaction session) and cognitively stimulating parenting behaviour (a questionnaire assessing parental teaching efforts, provision of teaching material, and the quality of literacy and leisure activities with the child) among VLBW/VP and FT 6-year-old children and their academic competence (type of school, repeated classes and knowledge in mathematics and German language) at the age of 13 years. The authors concluded that sensitive parenting behaviour (categorised as high, medium, low) correlated with adolescent academic success among the preterm born group but not among their FT born peers. Cognitively stimulating parenting behaviour, however, was beneficial for all adolescents’ academic competence regardless of birth status. The authors suggested that increased sensitive parenting behaviour among preterm populations may be more relevant during early school years than in infancy due to increased child responsiveness (Wolke, Jaekel, et al., 2013).

Intensive parenting practices may alter the parent-child relationship and impact parent outcomes. In an ethnographic study in the US of parenting practices among families with eight to 10-year-old children from varying socio-economic and ethnic backgrounds, intensive parenting behaviour reduced child contact with the extended family and the community (Lareau, 2002). Wall (2010) examined the extent to which Canadian mothers of five-year-old children and younger accepted the assumptions of intensive parenting culture in relation to their children’s ‘brain development’. The mothers in her study had internalised responsibility for their children’s cognitive development and future accomplishments by investing more time and resources on activities to stimulate their children’s brain growth. A similar construct to Wolke et al. (2013) ‘cognitively stimulating parenting’; an index they had developed for the study among parents of VP born children discussed above (Wolke, Jaekel, et al., 2013, p. 644). Wall (2010) found, that the internalised parental responsibility of the child’s cognitive development had generated stress and anxiety among the mothers, as they had limited their personal time to spend more time with their children. Generally, it appears that the intensive parenting culture has emphasised parental factors in the parent-child relationship over the

influence of child factors; for example, to focus on the parent's efforts in promoting the child's intellectual development may overlook the child factors' impact on cognition, such as genetic factors or possible impairments. In addition, in the parent-child dyad the parents also learn from their children (Smith, 2010), particularly when the children have special needs that may develop or change over time (Woodgate, Edwards, Ripat, Borton, & Rempel, 2015). Thus, the parent, although having a position of authority, is not the only driving force in the parent-child relationship (Harris, 1995).

To the best of my knowledge no previous studies have explored current parental experiences and parenting stress among English parents of EP YA in the context of intensive parenting culture and the framework of parental determinism. Adolescent behaviour among EP populations may differ from those born full-term; for example EP adolescents have reported lower rates of smoking and alcohol consumption, on average they have left their parental homes later, and reported having their first sexual intercourse at later ages than their full-term born peers (Kajantie et al., 2008; Saigal, Morrison, & Schmidt, 2020). These findings are influenced by EP adolescents' characteristics, such as behavioural problems and/or reduced social competence (Johnson & Marlow, 2017), and parenting behaviour such as 'scaffolding' to support child independence (Jaekel et al., 2012), but whether the intensive parenting culture and the framework of parental determinism are relevant constructs for parental experiences of parents of EP YA will be explored in this study.

I have reflected upon selected studies from the research field of prematurity on attachment, brain development, and overprotective parenting behaviour, situating the discussion in a critical sociology of discussion on parental determinism, risk management, and the culture of intensive parenting. Examples like the above, demonstrate that the research agenda is embedded in a wider sociocultural context (Macvarish & Lee, 2019; Macvarish et al., 2014). The movement may be bidirectional in which research findings fuel wider social discussion, which in turn triggers research interests. As my discussion on the intensive parenting culture has demonstrated, in addition to research among prematurity, parents themselves may view entities such as 'parenting' within the framework of parental determinism, which may impact parental experiences and parent outcomes such as anxiety and stress (Wall, 2010; Widding, 2014).

Framing parenting behaviour as deterministic may be harming for at least two reasons. Firstly, at the level of society, internalising parental determinism may create a culture where those with expert knowledge of parenting can be seen to 'police' the behaviour of 'lay' parents who 'do not know' (Thornton, 2011). Secondly, the logic following from parental determinism may

create a blame culture by implying that parenting behaviour of those parents whose children are facing challenges presently, such as behavioural difficulties or mental health disorders, has not been optimal (Gillies, 2008; Lee et al., 2014, p. 92-93). I considered the research examples of recall of overprotective parenting among EP adults to imply this rationale. A research culture in which those parents who have done it ‘right’ are compared to those who have done it ‘wrong’ based on their children’s long-term outcomes would not seem ideal or helpful for parents who experience difficulties today (Macvarish & Lee, 2019).

As the preceding discussion has shown, the concept of parental determinism is dominant in the understanding of parenthood in modern Western societies. I have also demonstrated that within the context of extreme prematurity, the parent-child relationship and parenting behaviour may be regarded as particularly important in supporting child development due to the potential sequela following from EP birth. This argument will be further discussed in the following sections of this introductory chapter. Studies exploring parental determinism, parenting as risk management, and/or intensive parenting practices, have examined the emergence of these constructs in policy documents (Gillies, 2008; Macvarish & Lee, 2019), in women’s magazines (Clarke, 2013), among parents from varying ethnic backgrounds (Lareau, 2002), among parents of healthy children (Schiffrin et al., 2013), children with learning disabilities (Valle, 2017) and among parents of children with chronic health conditions (Webster, 2018). To the best of my knowledge, current parental experiences of English parents of EP YA have not been explored using the framework of parental determinism. Nevertheless, as discussed above, parental determinism may be particularly relevant in the context of later life outcomes following EP birth in which the parental role in supporting child development may be further emphasised. The culture of intensive parenting may seem appealing to an individual parent if increased parenting practices are presumed to correlate with improved child outcomes (Shirani, Henwood, & Coltart, 2011). The linkage between the parent-child relationship, parenting behaviour, and parental determinism will be further illustrated and the roles that these concepts play in the present study will be summarised at the end of this chapter. First, I will present a closer examination of parent outcomes among parents of preterm born children over time.

1.6 Parenting Stress and Experiences among Parents of Preterm Born Children Over Time

1.6.1 Theoretical understanding of parenting stress

According to Deater-Deckard (1998) theoretically parenting stress is linked to the general understanding of stress. Stress is a reaction to an external or internal event whose manifestation

is modified, following appraisal, by individual's coping mechanisms. Parenting stress is the response to a trigger originating in the child/children and the parenting role. Thus, a level of stress in being a parent is typical when events/triggers occur in the parent-child relationship. An individual's response to the stress is influenced by child and parent factors, contextual factors such as societal expectations of the parenting role as well as parents' own perceptions of the appropriateness of child and parent behaviour. Individual coping mechanisms can mitigate stress reactions. A balance between parental perceptions of how demanding the child is to parent and the available resources to support the parenting task is important; perceptions of low levels of resources and challenging child behaviour and/or life situation may provoke parenting stress (Deater-Deckard, 1998).

Generally, increased parenting stress is linked to parenting behaviour that is not optimal for child development, such as less responsive and/or more controlling, or even abusive parenting behaviour (Abidin, 1992; Deater-Deckard, 1998; Maccoby, 1992). Parenting behaviour may create not only a more adverse context for child development, but also increase distress in the parent and reduce family functioning. Thus, the association between parenting stress and child outcomes is mediated by parenting behaviour (Figure 1-3). Low levels of parenting stress are viewed as important for optimal child outcomes and parent psychological wellbeing (Abidin, 1992; Deater-Deckard, 1998).

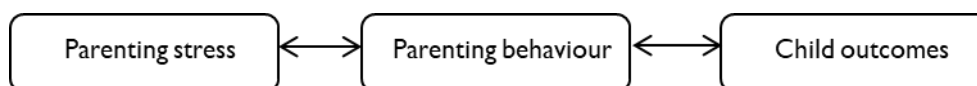


Figure 1-3 Association between parenting stress and child outcomes

Arrows linking varying factors represent the bidirectional impact between them

The relationship between parenting behaviour and stress, however, is not precise; for example, not all parents who are 'over' protective or controlling (as discussed above) experience high levels of parenting stress. Parenting stress is also impacted by the parent's motivation, thoughts, and beliefs. Abidin (1992) has focused his parenting stress theory on understanding the factors that determine parenting behaviour; how individuals act in their roles as parents. According to him, parenting behaviour is influenced by not only sociological, environmental, and behavioural factors, but also by parental perceptions, belief, and motivational systems. Parenting stress may appear when parents assess the benefits and harms in their parenting role through their internal understandings of themselves as parents. This leads to the concept of "self-as-parent" which is formed of "personal attachment history", "individual goals", and

“internalised expectations of others” (Abidin, 1992, p. 410). Abidin considered that parenting stress is the result of the summation of “a series of appraisals made by each parent in the context of his or her level of commitment to the parenting role” (Abidin, 1992, p. 410). Thus, highly protective, controlling or even abusive parents, even though displaying behaviours that may not be optimal for child development, may feel content with themselves as parents.

1.6.2 Parental distress in the Neonatal Intensive Care Unit

Arrival of a newborn infant may provoke intense emotions in parents, both excitement as well as uneasiness (Deater-Deckard & Bulkley, 2000). Generally, the prevalence of depressive symptoms and anxiety among all mothers after giving birth have been estimated to vary from approximately 10 to 20 percent (Carson, Redshaw, Gray, & Quigley, 2015; O'Hara & McCabe, 2013), although more varied figures have been reported. For example, a lack of support available for the new parents from their social networks may impact the level of experienced postnatal depressive symptoms (Myers & Emmott, 2021). Maternal mental illness prior to and during pregnancy may also impact symptomology postpartum (Kee et al., 2021). A wealth of research has found elevated levels of parental stress among mothers and fathers whose infants have been admitted to NICU following preterm birth (Caporali et al., 2020). For these families, the preterm birth may have followed from a stressful high-risk pregnancy and/or required an unexpected transfer to an unfamiliar tertiary level hospital (Kantrowitz-Gordon, 2013; Lasiuk, Comeau, & Newburn-Cook, 2013). Parental distress during infant hospitalisation has been reported to relate to altered parental role (Caporali et al., 2020), such as reduced involvement in the infant's care (Black, Holditch-Davis, & Miles, 2009), lack of information (Baia et al., 2016), frightening infant appearance (Caporali et al., 2020; Holditch-Davis & Miles, 2000; Widding & Farooqi, 2016), unfamiliarity with the required technological care interventions (Black et al., 2009) and concerns about future outcomes (Holditch-Davis & Miles, 2000). Parent gender, age, and high-risk obstetric variables, such as multiple birth or bleeding in pregnancy, may impact reported levels of stress and mental health outcomes such as depressive symptoms, anxiety or/and trauma reactions during infant hospitalisation (Baia et al., 2016; Misund, Nerdrum, Bråten, Pripp, & Diseth, 2013).

1.6.3 Parents' psychological health and stress in early childhood

The infant's discharge from hospital may be another event provoking heightened emotions among all parents. Retrospectively parents of children born preterm have reported feeling simultaneously eagerness to take their child home, as well as fear and anxiety about managing with less support (Granero-Molina et al., 2019; Schuetz Haemmerli, Lemola, Holditch-Davis, & Cignacco, 2020; Widding, Hägglöf, Adamsson, & Farooqi, 2020). In a study about

experiences of parents of preterm (≤ 34 weeks' gestation) born children over time (until child's age of 1.5 years) among seven sets of Swedish mothers and fathers, at discharge from NICU, the parents felt unprepared, as if they were "borrowing the baby from the staff" (Jackson, Ternstedt, & Schollin, 2003, p. 124). Six months later, the interviewees, particularly mothers, felt more adjusted to their roles as parents (Jackson et al., 2003).

Using data from the UK Millennium Cohort study, a nationally representative prospective cohort, Carson et al. (2015) assessed whether parents of children born preterm were in excess risk of psychological distress beyond the neonatal period and whether the degree of prematurity had an effect on the level of distress. Overall, 12,100 families reported on symptoms of parental distress (Rutter Malaise Inventory (RMI)) nine months after the birth. One hundred and nineteen families had children born VP (< 32 weeks' gestation). The prevalence of parental distress (PD; in the study defined as RMI score > 4), as well as self-reported current treatment for depression were highest among mothers of VP born children (PD: 21.7% and treatment for depression: 14.6% in comparison with all mothers PD: 12.1% and treatment for depression: 8.0%, respectively). Fathers met criteria for PD less frequently than mothers, although there was evidence of comorbidity in fathers when PD was present in mothers. Thus, family factors (comorbidity), parent gender (parent factors) and child's degree of prematurity (child factors) may all impact the levels of experienced parental distress. In their study, Carson et al. (2015) included married couples in which both parents were available to participate and excluded lone parents. Single parenthood has been associated with higher levels of parenting stress (Anderson, 2008). Consequently, the prevalence of PD reported in their study might have been higher if parents from wider backgrounds had been included (Carson et al., 2015).

Analyses like this highlight the issue of temporality; among mothers of preterm born infants a history of psychological illness and presence of risk factors for preterm birth, such as preeclampsia, have been found to be associated with parental distress post parturition (Misund et al., 2013). In the UK Millennium cohort study 23.2% of all mothers reported ever being diagnosed with depression, whereas 41.2% of mothers of VP born children had had a previous diagnosis of depression (Carson et al., 2015). Mothers' psychological illness may carry through from preconception, into pregnancy, through to the postnatal period (Deater-Deckard & Bulkley, 2000; Kee et al., 2021; Misund et al., 2013). In the context of prematurity, the stressful experience of infant hospitalisation in the NICU may be followed by adjustment to life at home while parents (particularly mothers) may still be experiencing psychological distress (Misund et al., 2013).

Other investigations have also considered what impact severity of infant's medical condition may have on parents' early experiences. In the NICU, the infants' neonatal risk level does not directly correlate with parental stress scores (Caporali et al., 2020). This is aligned with post-traumatic stress and growth literature among severe paediatric illness showing that personal parental appraisal of the situation may impact the degree of distress more than objective severity of child illness (Picoraro, Womer, Kazak, & Feudtner, 2014). Yet parents of the smallest infants (who have the highest medical risk) have reported experiencing insecurity and overprotectiveness in their roles as parents for longer times than parents of more mature preterm infants (Jackson et al., 2003). At one month after birth, mothers of very low birth weight (VLBW) high medical risk infants reported higher levels of parenting stress compared with mothers of VLBW low-risk infants (Singer et al., 1999). At 9 months after birth, parents of VP born infants reported higher levels of parental distress than parents of late preterm born children (Carson et al., 2015). However, 24 months after birth, greater infant neonatal risk was associated with less reported parenting stress, potentially reflecting a sense of relief among parents of initially high-risk infants, parental adjustment to the expected challenges, or/and the impact of continued support received through follow-up care in comparison with families with children born with low medical risk, who perhaps have less support from follow-up services (Gerstein & Poehlmann-Tynan, 2015).

1.6.4 Parenting stress in later childhood

Studies of parenting stress among parents of VP born children have suggested that the initially heightened levels of stress may decrease during early childhood (Treyvaud, 2014), but increase towards middle childhood (Treyvaud, Lee, Doyle, & Anderson, 2014). Yet, stable levels of average parenting stress scores (the Parenting Daily Hassles Scale) from the child age of two to six years among mothers of preterm born (≤ 36 weeks' gestation) children from the US have been reported (Gerstein & Poehlmann-Tynan, 2015). A few studies have examined stress levels of parents of early adolescents born preterm (Treyvaud, 2014). Parents of Australian children born either VP or FT in early 2000s reported comparable levels of parenting stress (using the Parenting Stress Index (PSI)) when their children were two years old (Treyvaud et al., 2011). When re-assessed at the children's age of seven years, parents of children born VP reported higher average parenting stress scores than parents of FT born children, particularly child-related stress, suggesting that parenting stress fluctuated over time. Yet, among both groups of parents, high parenting stress scores at the child's age of two years predicted high scores at the age of seven, supporting continuity of experiences among parents reporting high stress. The differences in stress scores between the groups of parents persisted after adjusting for social risk level and child neurodevelopmental disability (Treyvaud et al., 2014). Similarly,

in a study of stress among parents of VP born 7-year-old Canadian children and among parents of their FT born peers, VP mothers reported higher levels of parenting stress that was associated with child male gender, escape/avoidant maternal coping style, and maternal perception of child externalising behaviour (Linden, Cepeda, Synnes, & Grunau, 2015).

The potential changes in parenting stress scores over time among parents of preterm born children may be influenced by changes in causation (Deater-Deckard, 1998). When the initial parental distress may relate to the challenging birth and NICU experience, later scores may be impacted more by child developmental and behavioural factors (Brummelte, Grunau, Synnes, Whitfield, & Petrie-Thomas, 2011). Indeed, when utilising the PSI which has distinct child and parental stress domains, several studies (such as Treyvaud et al. (2011; 2014) above) have found that parents reported the most heightened levels of child-related parenting stress in comparison with parental distress (Schappin, Wijnroks, Uniken Venema, & Jongmans, 2013). Linden et al. (2015) concluded that among parents of VP and FT born seven year old Canadian children without major neurodevelopmental impairments, child externalising behaviour was positively associated with increased parenting stress scores. Although parent factors such as coping strategies (Linden et al., 2015; Singer et al., 2007), level of education (Singer et al., 2010) and psychological health (Linden et al., 2015) have been found to be associated with parenting stress, it is important to note that scores reported during pre- and early schoollyears have generally shown only modest elevation in comparison with the scores of parents of FT born children and the general population (Brummelte et al., 2011; Landsem, Handegard, Tunby, Ulvund, & Ronning, 2014; Linden et al., 2015; Schappin et al., 2013; Treyvaud et al., 2014).

Higher child-related parenting stress scores suggest that the parent perceives the child to have characteristics that make them challenging to 'parent' (Abidin, 2012). Among EP children this may relate to the high prevalence of impaired neurodevelopment (Johnson et al., 2009; Marlow et al., 2021) and/or increased behavioural problems in childhood and early adolescence in comparison with children born FT (Dahl et al., 2006; Taylor, Margevicius, Schluchter, Andreias, & Hack, 2015). Researchers have acknowledged potential transactional processes in the experience of parenting a preterm born child. Initially heightened parenting stress may provoke child behavioural problems and child non-compliance that in turn increases parenting stress (Gerstein & Poehlmann-Tynan, 2015; Sameroff & Mackenzie, 2003). After assessing parent-child interaction and perceived parenting stress among mothers and their preterm born children from the neonatal period to the ages of 24 and 36 months up until six years of age, Gerstein et al. (2015) found that higher early parenting stress scores were associated with increased maternal insensitivity ratings at the age of 36 months. Both parenting behaviour and

stress were then associated with child behavioural problems at the age of six years. The authors found less evidence of observed child non-compliance at the ages of 24- and 36-months to predict parenting stress scores at the age of six years. The authors concluded that among their study population parenting stress was stable over time and associated with altered parent and child behaviour. They called for long-term support for all families of preterm born children, instead of only targeted follow-up care for families with the most medically at-risk children (Gerstein & Poehlmann-Tynan, 2015).

Interventions targeting parenting behaviour, such as those supporting parental psychological adjustment and caregiving practises, can reduce parenting stress (Deater-Deckard, 1998; Deater-Deckard & Bulkley, 2000). At first, child developmental interventions among preterm born populations were developed to offset the developmental disadvantage of preterm birth. Later, the transactional system models (i.e., parent-child interaction) recognised the parent as an active interventional agent and concentrated also on supporting the parent (Sameroff & Mackenzie, 2003). The interventions have varied largely, but have generally included multidisciplinary services for parents promoting child development, minimising and/or preventing developmental delays, and supporting parenting and family functioning from birth to the age of five years (Spittle, Treyvaud, Lee, Anderson, & Doyle, 2018). The impact of such interventions have been assessed in randomised controlled trials (RCT) (Landsem et al., 2014; Spittle et al., 2018).

Although the evidence to support a sustained impact of early interventions on child or parent outcomes has varied (Benzies, Magill-Evans, Hayden, & Ballantyne, 2013), some RCT have reported improved parent outcomes in later childhood (Landsem et al., 2014; Spittle et al., 2018). Following a mother-infant interaction intervention at the time of infant's discharge from hospital among 146 preterm born Norwegian children (birthweight <2000 grams), of which 72 were randomised to an intervention arm, and 75 FT born control children and their parents, Landsem et al. (2014) reported lower parenting stress scores (PSI) and improved child socio-emotional and developmental outcomes between ages of one and nine years in the preterm born intervention group in comparison with the other two groups. The authors suggested that this was a result of improved maternal adjustment to their child (Landsem et al., 2014). In another RCT among Australian families, regardless of not-sustained improvements in child outcomes in the intervention group, parent mental health (anxiety and depression) improved most among the primary caregivers who had received the intervention. Overall, the effect sizes were modest and the groups of parents reported levels of anxiety and depression that were within normal or mildly elevated ranges (Spittle et al., 2018). Both authors acknowledged the methodological challenges of conducting RCT among populations

that, for example, may simultaneously have long-term contact with other healthcare services and parenting support (Landsem et al., 2014; Spittle et al., 2018).

There has been a rapid recent increase in parent outcome research among preterm populations (Zeitlin et al., 2020), with most studies focusing on the first months and years following the birth and hospitalisation. At these times, the influence of the perinatal experience may still be active, and these studies do not reveal how parents' feelings, perceptions or mechanisms of coping may change over time (Janvier et al., 2016). Parents may reflect their early experiences differently when their children's long-term outcomes are known (Widding et al., 2020), in comparison with parental experiences reported during hospitalisation (Wraight, McCoy, & Meadow, 2015). Instead of potential positive family transformations, individual growth, and/or resilience following preterm birth, the overall theme in literature has been to focus on negative outcomes (Janvier et al., 2016; Lou, Pedersen, & Hedegaard, 2009). The nature of premature birth proposes negative outcomes instead of positive experiences, which may have contributed towards a general negativity about the outcomes studied.

1.7 Pathways of Influence in Parental Experiences: Birth and Early Childhood

Although short and of selected sources, the discussion above has demonstrated that when long-term parent outcomes or parental experiences are examined within the context of prematurity, a common starting point has been the potential impact of the adverse birth experience and the infant's prolonged hospitalisation. Studies have hypothesised based on a 'challenged' parent-child relationship in the belief that initial separation produces difficulties (Hallin et al., 2012; Taylor, Klein, Minich, & Hack, 2001) and has a long-lasting impact on the parental emotional responses of stress and anxiety (Miles & Holditch-Davis, 1997). A majority of the studies referenced above were identified during the process of screening publications to be included in the systematic review of long-term parent outcomes following EP and VP birth presented in the next chapter (for the exact search strategy please see page 57 in the next chapter). Web of Science was used to identify recent publications that had referenced the previously identified reports.

In their Preterm Parental Distress Model, a conceptual framework of pathways of influence in parenting prematurely born children, based on a review of previous research on prematurity, Miles and Holditch-Davis (2000; 1997) begin by discussing the impact that pre-pregnancy and perinatal factors, such as maternal age, SES, history of infertility, previous experiences of loss or/and high-risk pregnancy, may have on the emotional responses that parents display in the event of preterm birth and infant hospitalisation. Following the infant's admission to NICU,

parental experiences are shaped by loss of anticipated parental role, and the uncertainty about their child's outcome and clinical status. These experiences are further affected by social and environmental factors such as the family's ability to visit (economic resources, physical distance, work commitments, needs of siblings), the way in which staff approach the infant and the parent-infant dyad and communicative progress, and the physical environment of the unit (medical equipment, noises, lights). These factors influence the magnitude of emotional responses of anxiety, stress, fear, depression, worry or/and feelings of loss of control. Miles and Holditch-Davis then conclude that "these early neonatal experiences result in a different orientation to parenting" (Miles & Holditch-Davis, 1997, p. 261). This 'different orientation' is driven by the perception that the child is 'vulnerable' or 'special' because of the birth and the neonatal course. Within this process model, the parent-infant relationship becomes altered, and parents may, for example, display 'compensatory' parenting behaviours because of initial perceptions of 'fragility'. As a result, these altered parenting practices impact child health and developmental outcomes long term (Holditch-Davis & Miles, 2000; 1997). In this model, the proposed associations between parental responses to stress, parenting behaviour, and child outcomes are similar to the parenting stress models presented by Abidin (1992) and Deater-Deckard (1998) (please see Figure 1-3 earlier).

These frameworks have clearly influenced research hypotheses. Since then, the research focus in prematurity has expanded from the adverse birth experience and infant hospitalisation to early childhood. Generally, the first few years of childhood have been regarded as important in influencing the child's cognitive outcomes, emotional status, and social competence through to adolescence and adulthood (Macvarish et al., 2014). Lower quality of parent-child interaction and parenting behaviour, such as reduced parent responsiveness and/or sensitivity or increased parental control may lead to impaired child outcomes, whereas increased parent sensitivity and responsiveness can promote these outcomes among prematurely born children (Beckwith et al., 1992; Bilgin & Wolke, 2015; Jaekel et al., 2012; Wolke, Jaekel, et al., 2013). Following the psychoanalytic framework and attachment theory (Maccoby, 1992), the effects of the parent-child relationship established after birth and during early childhood are regarded as relatively stable over time (Beckwith et al., 1992; Hallin et al., 2012). Yet factors such as child independence, parental control, individual behaviours (e.g., child behavioural disorders) and/or family context (e.g., divorce) or contextual factors (e.g., changes to healthcare services or governmental support systems) may change over time and modify the parent-child relationship.

There is evidence that parents of preterm born children may recall the birth and hospital-period as stressful years later (Widding et al., 2020). Further evidence indicates that parents of

preterm born young children may interact differently with their child in comparison with parents of children born full-term (Beckwith et al., 1992; Jaekel et al., 2012; Wolke, Jaekel, et al., 2013). This altered parent behaviour may be a beneficial response to the child's needs (Jaekel et al., 2012), and it may impact the relationship that the parent and the child have in early adolescence (Beckwith et al., 1992) as well as influence adolescent outcomes, such as academic achievements (Wolke, Jaekel, et al., 2013). Conceptual frameworks, such as those presented above, may be valuable as they suggest possible points for interventions, for example, they may help to direct long-term support or guide support services in potentially more stressful times such as discharge from hospital (Granero-Molina et al., 2019) or transition to adolescence (Beckwith et al., 1992). The models promote improved long-term outcomes among both the children and their parents (Deater-Deckard, 1998). However, it is important to consider that studies following causal models like these are limited by their methodology, such as investigating limited data on preconception, pregnancy or/and perinatal factors, or wider family context over the years that might influence findings. Although there is a tendency to use such models, other research foci have equal value, such as positive transformations following adverse events (Scorgie, Wilgosh, & Sobsey, 2004). Lastly, if the defining experiences of birth and early childhood are viewed as deterministic, they may seem less relevant for parents and adolescents with current challenges, as those events have already taken place (Macvarish & Lee, 2019). For example, challenges among adolescents who display risky behaviours, such as substance abuse, may be viewed as 'inevitable' due to adverse early childhood environment (Barnes & Power, 2012).

1.8 The EPICure Studies

The EPICure studies are national birth cohorts in the UK established to describe survival and later life outcomes following EP birth (EPICure and EPICure 2). The first EPICure cohort collected data on all births less than 26 completed weeks of gestation in the UK and Northern Ireland over a 10-month period in 1995. The 309 long-term survivors who were admitted for neonatal intensive care have been followed up at 2.5 years (n=283; 92%), 6 years (n=241; 78%), 11 years (n=219; 72%) and at 19 years of age (n=192; 42%) (Suonpera, 2019).

Since the first EPICure cohort, research has led to significant advances in neonatal care, such as provision of antenatal steroids to promote the infants' lung maturation, use of surfactant replacement to improve their lung functioning, and measures taken to avoid hypothermia and improve infant nutrition. In the early 2000s, neonatal care in England was organised into hospital networks to centralise the care of most immature infants into tertiary centres with most experience. These improvements in care supported the decision to establish a second

cohort to determine any changes in outcomes among the survivors in line with advances in practice. The EPICure 2 cohort was established in 2006 for infants born at 22-26 weeks of gestation to mothers resident in England (Costeloe et al., 2012).

The neonatal and child outcomes between the cohorts have been compared at discharge from hospital (Costeloe et al., 2012) and at 2.5 years of age (Moore et al., 2012). EPICure 2 has shown improved survival at all gestations even though the proportion of major short-term morbidities has remained similar between the cohorts. In comparison to the first EPICure cohort, 13% more infants born between 22-25 weeks of gestation were discharged alive from hospital after being admitted for neonatal intensive care. Survival increased with increasing gestational age from 9.5% at 23 weeks to 16.0% at 25 weeks. The proportion of neonatal morbidities at discharge, such as major cerebral injury, were similar between the cohorts, although infants born in 2006 were more often discharged when still receiving supplementary oxygen and having undergone laser treatment for retinopathy of prematurity (increases of 7% and 8%, respectively). The consistency in the patterns of early morbidity between the cohorts has been taken to suggest consistency in the long-term predictors of outcomes among the survivors born in 1995 and those born a decade later in 2006 (Costeloe et al., 2012).

The finding of an increased survival accompanied by stable levels of neonatal morbidity is in line with other similar birth cohort studies (Torchin et al., 2020); a French study compared the outcomes of infants born at less than 34 weeks of gestation in 1997 and 2011. Although showing increased severe-morbidity-free survival at older gestations, the outcomes for infants born less than 25 weeks of gestation remained stable (Ancel et al., 2015). Infants born with birthweights less than 1000 grams in Finland between 1996-1997 and 1999-2000 had similar survival rates and comparable levels of oxygen dependency at term (Tommiska et al., 2007). Recent studies have also suggested that the levels of morbidity in early (Pierrat et al., 2017) and later childhood (Marlow et al., 2021) have remained stable. Meta-analyses have indicated that despite increased survival, no improvement in cognitive outcomes in childhood for VP born survivors have been demonstrated (Twilhaar et al., 2018).

A majority of studies assessing long-term parent outcomes following preterm birth have included families of infants born in the 1980s and early 1990s. However, increased infant survival and recent advances in neonatal care may have increased parental expectations and altered the parents' early experiences, perhaps altering the current trajectory of long-term parent outcomes in comparison to the older cohorts (Drotar et al., 2006). In the EPICure cohorts, infants born in 2006 were more likely to survive the first seven days following birth in comparison to those who were born in 1995. There were no differences in the survival rates

between the cohorts after the first week. This may have led to a circumstance, in which infants who previously would have not survived, now lived at risk of more complications than previously. Among those infants in the EPICure 2 cohort who were alive at seven days, deaths from infection and enterocolitis increased in comparison to infants born in 1995, perhaps increasing the uncertainty among the parents (Costeloe et al., 2012). This may have provoked further parental stress and anxiety during the infant's lengthy hospitalisation in the neonatal intensive care unit. Parents' experiences of the hospital stay and the memory of having a newborn baby who is in a need of intensive care can be long-lasting (Widding et al., 2020). Consequently, despite the apparent consistency over time in the patterns of early morbidity among the infants, the comparability of parent outcomes between earlier and more contemporary cohorts has remained unclear.

1.9 Defining 'Good' and 'Poor' Outcomes Following Preterm Birth

In the context of prematurity, the aim of neonatal medical care has been to promote survival of the newborn babies, minimise their ill-health, and decrease long-term morbidities experienced by these infants. Thus, survival rate and the level of long-term morbidities have been regarded as the discipline's 'success measures' (Marlow, Hoy, Peacock, & Kamphuis, 2020). In the literature, the importance of survival and low levels of later life morbidities have been discussed in the context of public health (the cost of health care or burden on education and social services) and health care ethics (informed decision-making regarding infant care together with the parents) (Costeloe et al., 2012). What has been regarded as a 'good' or a 'poor' long-term outcome following EP birth has predominantly been defined by focusing on the child outcomes, perhaps overlooking outcomes among the parents.

Survival has been the principal outcome measure, often measured from 'live at birth' to 'discharged alive' from hospital. In the EPICure 2 cohort in 2006 it was 2% (n=3) for babies born at 22 weeks of gestation, 19% (n=66) at 23 weeks, 40% (n=178) at 24 weeks, 66% (n=346) at 25 weeks, and 77% (n=448) among infants born at 26 weeks of gestation (Costeloe et al., 2012). Survival is naturally impacted by the care decisions made at birth, such as the infant's active stabilisation, mortality being practically 100% among those infants who do not receive active care. In addition, the choice of denominator impacts the survival rate; whether those surviving are compared to those who were 'alive at the onset of labour', 'born alive', 'admitted for intensive care' or those who survived the first few days following birth. At all 'stages' the outcomes have been poorer for less mature infants (Costeloe et al., 2012; Torchin et al., 2020).

In the short-term, child outcomes of interest have been neonatal morbidities including continued need for oxygen supplement beyond becoming a full-term infant, diagnosed as bronchopulmonary dysplasia (BPD), abnormal findings in cerebral ultrasonography suggestive of long-term neurological difficulties such as cerebral palsy, retinopathy of prematurity increasing the risk of blindness and necrotising enterocolitis, a bowel disorder associated with high rates of mortality. In addition, growth measures have been of interest (Costeloe et al., 2012). The long-term outcomes of focus have generally been respiratory problems, learning difficulties, cerebral palsy, and behavioural problems. The risk of poor long-term outcomes has increased with decreasing maturity and male sex (Johnson et al., 2009; Moore et al., 2012). The challenges such as low motor functioning, poor growth, low IQ and/or behavioural difficulties, such as attention deficit, have generally persisted to adolescence and early adulthood (Marlow et al., 2021; Ni et al., 2020; Taylor et al., 2015).

Even though the neonatal outcomes have been suggestive of the long-term outcomes, the link between them has not been definite (Hallin, Hellstrom-Westas, & Stjernqvist, 2010; Torchin et al., 2020). The level of immaturity has been highly predictive of later life outcomes. Nevertheless, among the infants in the EPICure 2 cohort, in contrast to other neonatal morbidities, a severe abnormality reported on cerebral ultrasonography was not related to gestational age (Costeloe et al., 2012). A unique aspect of prematurity has been its simultaneous relation to several physical, psychological and cognitive health concerns, creating uncertainty in the long-term outcomes (Drotar et al., 2006). Parents of EP children may have experienced uncertainty in relation to their children's capabilities in comparison to their full-term born peers as they approach school age (Torchin et al., 2020), for example, parental expectations of 'catching up' in developmental outcomes have been reported (Marlow et al., 2020).

A general trend in neonatal intensive care has been to promote increased parent involvement in their infants' care (Shaw et al., 2019; Widding et al., 2020) and to endorse joint decision-making between the medical professionals and the parents in care decisions such as those relating to the infant's life sustaining treatment (Shaw, Stokoe, Gallagher, Aladangady, & Marlow, 2016). Even though the parent's role is to make 'informed' decisions, due to the uncertainties in the long-term outcomes, both the child's and the parent's, the decisions may be difficult to regard as fully 'informed'. Parents' experiences of the decision-making process may change over time as the child grows older (Allen, 2014) and more time passes since the decision (Caeymaex et al., 2011; O'Neill, 2019). Parents have experienced the joint decision-making process differently depending on whether the treatment choices were communicated to them as 'recommendations' or 'options' (Shaw et al., 2016). Retrospectively parents have

felt it important that they did not bear sole responsibility for the choices made regarding their infant's care, but that their recollection is that decisions were collaboratively made together with the medical professionals (Caeymaex et al., 2011). Nevertheless, after the infant has been discharged from hospital, the consequences of the joint decisions have largely remained on the parents. Parents' initial focus may have been on the survival of their child, rather than on any consideration of long-term outcomes (Einarsdottir, 2009). In addition, even though the healthcare professionals have been able to inform parents of possible long-term sequelae, the meanings and perceptions of entities such as 'disability' may have varied between the parents and the professionals (Einarsdottir, 2009; Lou et al., 2009; Marlow et al., 2020).

1.10 Representations of Prematurity

In the wider public discourse, when discussing 'prematurity' a metaphoric language using terms such as 'fragile', 'tiny', 'miracle' or 'fighter' have commonly been employed (Isaacson, 2002; O'Neill, 2019). Since EP infants are being born immaturely, it may have seemed natural to refer to them as 'fragile' or 'tiny'. On occasions survival at the limit of viability has been framed as a 'miracle'. To the parents, the initial experience of vulnerability followed by a 'miraculous' survival may pose additional expectations for their roles as parents (O'Neill, 2019). It may be culturally normative to regard preterm born children as 'fragile' (Tallandini et al., 2015) which may further emphasise the parental role to support child outcomes.

In addition to wider media (BBC, 2004b), metaphoric language has appeared in research publications (Isaacson, 2002). Studying cultural artifacts published over a ten-year period in the US, Isaacson (2002) found that most commonly preterm birth was referred to in a positive tone. A fifth of the newspaper, magazine, and research articles she identified utilised the term 'miracle' when referring to the survival of the preterm infant. In its incontestability the 'miracle' metaphor may have functioned beneficially; it frames an unfortunate, often dangerous event of preterm labour in a positive light, shadowing the possible struggles that the infant or the parents may have experienced, both in the short and long term (Isaacson, 2002). Parents of children born with morbidities have linked the 'miracle' metaphor to a narrative of hope, in which their infant's developmental challenges are regarded as temporary, something that they can 'fight through', 'repair', and overcome (Kantrowitz-Gordon et al., 2016; Landsman, 1998). In the long-term, parents of these initially 'fragile' children may feel additional pressures to ensure their child's future success. Within the framework of parental determinism, by parenting their 'survivor' children 'optimally', the experience of 'saving' them may be prolonged. A study among parents of children with long-term chronic conditions suggested that the parents may become so called 'expert carers' to their children. Due to their

child's morbidity, resource-heavy and time-consuming intensive parenting practices had become a necessity (Webster, 2018).

The discursive representation of the preterm born child as a 'miracle', 'fragile', or 'fighter' may also provide tools for the parents to utilise when re-building their perceptions of 'parenthood' following the adverse birth experience and prolonged hospital stay. Perceiving one's child as initially vulnerable may help to better understand potential later life challenges and/or adjust initial aspirations and expectations for one's own parenthood or the child's development (Wilson & Cook, 2018). Extreme prematurity is not a natural state of being, but rather the continuity of life is ensured with medical, technological, and social input (Orfali, 2017). Consequently, in public discourse, the survivors are often stated to "owe their lives to modern neonatal intensive care" (BBC, 2004a). However, in addition to the focus on survival, it is important to acknowledge and better understand the pivotal roles of parents in making the "miracles" happen. We should not forget long-term parental inputs and the consequences of those efforts behind the flagship success stories surrounding extreme prematurity.

1.11 'Parenting' versus 'Parent Outcomes'

The discussion that I have presented in this introductory chapter, for example on the themes of attachment and early childhood, have demonstrated that, in line with wider discourse about 'parenting', research into the effects of prematurity has embraced the concept of parental determinism; the designs of previous studies have presumed the parent behaviour to have a direct impact on, either to promote or hinder long-term child outcomes such as cognitive development or social competence (please consider the earlier examples of Day, Dobson, et al. (2018); Day, Schmidt, et al. (2018); Hallin et al. (2012); Treyvaud et al. (2020)). In neonatology, in the context of preterm birth and the importance of early childhood, parenting has been framed as an 'intervention' that can be employed to promote child outcomes. Consequently, many studies into the effects of prematurity have assessed the influences that the parent (while 'parenting') may have in determining child development but disregarded the influences that the task of 'parenting' may have on parent wellbeing or quality of life. Perhaps to remain within the 'boundaries' of the discipline, many studies assessing long-term parent outcomes (not 'parenting') following preterm birth, have articulated their purposes to be to promote the parent-child relationship and consequently to improve child outcomes (for example Taylor et al. (2001)). Even though framed as investigations of 'parent outcomes', such as depression, anxiety or stress, the focus frequently remains on the consequences for the child, and the effects on parents have remained secondary and therefore, intrinsically parent outcomes lack value. This, a kind of 'metanarrative' of research on long-term parent outcomes

following preterm birth, will become more evident when discussing the findings from the systematic review presented in Chapter 2.

I regard ‘parent outcomes’ and ‘parenting’ as two different, although interlinked, entities that have in some research agendas mistakenly been viewed as one. Research on long-term ‘parent outcomes’ examines outcomes, such as stress, mental health, social outcomes (economic status, work, educational attainment, reproductive behaviour), parental experiences, perceptions or/and feelings among individuals in relation to their roles as parents. Here the child or children and the task of ‘parenting’ are factors that may impact the outcomes among the individuals. Research on ‘parenting’, however, examines the impact of parent behaviour, the set of actions that is called ‘parenting’, commonly on child health and development. Here the parent behaviour is the exposure (or intervention) impacting the outcomes of child health or/and development. As per the process models of parenting behaviour discussed earlier, ‘parenting’ is impacted by child, parent, and family factors, as well as by wider contextual factors (Abidin, 1992; Deater-Deckard, 1998; Miles & Holditch-Davis, 1997). It seems that much of research among prematurity has focused on ‘parenting’.

By its nature, the research on long-term outcomes following preterm birth has relied on long causal pathways and complex models of trajectories. Consequently, a degree of uncertainty has been inherent in its findings. Similarly, the modern social world is characterised by uncertainty, by being built on defined and ever re-defined hypotheses rather than stable facts (Giddens, 2013, p. 8). Parents execute their roles in the social context of modern life that is evolving and changing. Therefore, when complex entities such as ‘parental experiences’ are investigated, rather than approaching the topic within a given discipline, a multi-disciplinary perspective may provide deeper understanding.

At 11 years of age the EPICure2@11 Study re-evaluated key psychological, medical and parent outcomes using standardised established measures at an important phase of the participants’ and their families lives – the transition to adolescence and secondary education. This study provided an outstanding opportunity for in-depth investigation of parental experiences previously missing from the literature. The aim of this inquiry was to locate the parent outcomes within their lived experiences.

1.12 Research Aims and Questions

This study aimed to improve our understanding of experiences of parents of children born EP entering adolescence. This overall aim was divided into three specific quantitative, qualitative

and mixed methods research aims and questions (Teddlie & Tashakkori, 2006). The aim of the analysis was to explore the extent to which the quantitative and qualitative results converged and to determine how quantitative outcomes can be related to parental experiences.

The objectives of the present study were to:

1. Identify sources of parenting stress among primary caregivers of EP YA and their age, gender and school class matched control children's primary caregivers
2. Explore parental experiences of parents with children born EP in transition to adolescence at 11 years of age
3. Explore whether, and if so, how parenting stress and associated sociodemographic, child and parent health factors converge with parental experiences

In meeting these objectives, this study addressed three related research questions:

1. How do parents experience parenting their child born EP during the transition to adolescence at around 11 years of age? (qualitative)
2. Is parenting stress associated with birth status, sociodemographic, child health or parental health factors? (quantitative)
3. How do parental experiences relate to parenting stress and associated sociodemographic, child health and parental health factors? (mixed)

To conclude, this chapter has discussed the parent-child relationship, general research on parenting behaviour, experiences of parents of preterm born children over time, and the theoretical frameworks of parental determinism and parenting stress, with relevant empirical research examples. A commonality between the various frameworks presented in this chapter, is that they are process models of parenting behaviour in which child factors, parent factors, family factors and contextual factors are in an interplay influencing the parent-child relationship and parenting behaviour (please see Figure 1-4 below). Parental determinism can be regarded as a part of the wider sociocultural context in which parents operate and, similarly, as a contextual factor it can impact parenting behaviour. Studies have, for example, examined how parenting stress, an EP birth as an adverse birth experience, prolonged infant hospitalisation as a risk factor for insecure attachment, and parental perceptions of child

vulnerability may alter parenting behaviour. Some studies have examined the process model as causal; how it may impact child outcomes. However, in this study, I postulated the movement in the model to be bidirectional. For example, child outcomes can be regarded as child factors (indicated by a long dash dot line) that can impact parenting behaviour and provoke parenting stress or impact other parent outcomes such as mental health outcomes or health-related quality of life (HRQoL). In the present study, the focus was not on how parenting behaviour may impact adolescent outcomes, but what factors in the bidirectional model of parenting behaviour may impact parent outcomes among parents of EP YA at a particular point in time in early adolescence. Chapter 2 will systematically examine literature on parent outcomes among parents of adolescents born VP. Following a presentation of research methodology in Chapter 3, Chapter 4 will take the particular parent outcome of parenting stress and examine what child, parent, family, and contextual factors are associated with parenting stress reported in adolescence (indicated by the section inside a broken line). Chapter 5 will explore parental experiences; how EP parents talk about their parental experiences and examine these descriptions against the conceptual framework of parental determinism (indicated by the section inside a dotted line). Inclusion of ‘adult competence’ inside a broken line box indicates that parenting behaviour is ultimately aimed at child socialisation (Maccoby, 1992; Ogbu, 1981) as discussed at the beginning of this thesis. The concept of future adult competence (i.e., not yet achieved) is further discussed in Chapter 5. Finally, narrative data integration is presented in Chapter 6 (Figure 1-4).

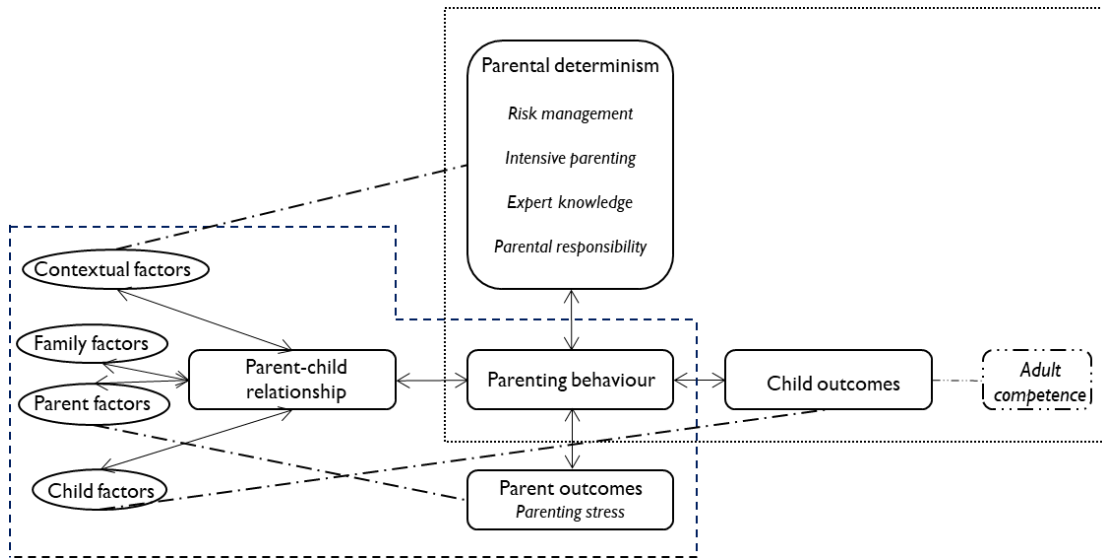


Figure 1-4 The bidirectional model of parenting behaviour

Arrows linking varying factors, parent-child relationship, parenting behaviour, parental determinism and parent and child outcomes indicate bidirectional movement between these constructs. Long dash dot lines between child outcomes and child factors, parent outcomes and parent factors and contextual factors and parental determinism indicate that these constructs can be regarded as the same due to the bidirectional movement in the model. The section inside the broken line will be explored in Chapter 4. The section inside the dotted line will be explored in Chapter 5.

Chapter 2 Long-term Parent Outcomes Following Very Preterm Birth: A Systematic Narrative Review of Qualitative and Quantitative Evidence

2.1 Introduction

The potential for impairment following preterm birth is well described and the risk rises with decreasing gestational age at birth (Ancel et al., 2015; Costeloe et al., 2012; Moore et al., 2012). As discussed in the preceding chapter, the experience of intensive care and concerns about child's survival may provoke parental distress (Caporali et al., 2020). The impact of prematurity on the parents has been assessed during the period in neonatal intensive care (Baia et al., 2016; Wraight et al., 2015) and through the early years of childhood (Carson et al., 2015; Granero-Molina et al., 2019; Treyvaud et al., 2011). However, as the child grows the experience of parenting a preterm born child may change as the child enters new developmental phases and the parents become more experienced. For example, as discussed in Chapter 1, increased parental experience of caring for a preterm born child may relate to the cumulative experience in the parent-child dyad, as well as to the increased parental knowledge of the child's health profile (Thomasgard & Metz, 1997). Parents of adolescents with complex care needs have reported continuous learning in their roles as parents as their children's care needs change over time (Woodgate et al., 2015). Thus, earlier parental experiences may change over time (Treyvaud, 2014). In this systematic review of literature, I will collate findings from current publications describing and/or assessing long-term parent outcomes following EP or VP birth.

2.2 Background

Adolescent behaviour - Adolescence is a key phase in child development; the period when the child transitions to adulthood characterised by increased emotional and social self-control and greater drive for peer socialisation and acceptance (Casey, 2015; Taylor, 2020). During this phase, the cognitive, social-emotional, and self-regulation skills developed throughout the childhood with the support of parents (Kuppens & Ceulemans, 2019) can lead to autonomous adulthood (Casey, 2015). Emotional support previously primarily acquired from the parent-child relationship may in adolescence be gained from peer relationships (Best, Manktelow, & Taylor, 2014; Yerkes et al., 2019). Consequently, adolescence is an important time for forming and sustaining friendships (Best et al., 2014) when children face new cognitive, physical, and social challenges (Casey, 2015). Although parental childrearing goals are highly contextually varied, for example in relation to the culture and families' SES, Western parents have generally valued fostering their children's independence, happiness, and academic or professional skills

and competence in their parenting practices (Suizzo, 2016; Tulviste, Mizera, De Geer, & Tryggvason, 2007).

During adolescence, families may face unexpected challenges as new expectations for the child's social and emotional skills appear, particularly among families with children born preterm (Greenley et al., 2007; Taylor, 2020). As introduced in the preceding chapter, higher rates of neurodevelopmental disabilities, psychiatric illness, and emotional or behavioural challenges among EP adolescents in comparison with adolescents born FT (D'Onofrio et al., 2013; Hallin et al., 2010) may challenge socialisation in peer groups. Early adolescents born EP/VP preterm (12 years of age) have reported more social difficulties with their peers, having smaller social networks, and being bullied more frequently in comparison with their FT born peers (Ritchie, Bora, & Woodward, 2018). Among 151 British adolescents born VP (<33 weeks' gestation) the proportion of children categorised with cognitive and/or neurological impairment/s increased between the ages of eight and 15 years (e.g., from none to mild disabilities), and the proportion of VP born adolescents receiving special educational support grew over this time period (O'Brien et al., 2004). Independent of the parents' educational level or marital status, EP/ELBW adolescents, particularly young men (Goddeeris et al., 2010), were less likely to complete their college education and less commonly started university courses in comparison with their FT born peers (Goddeeris et al., 2010; Hallin et al., 2010). Preterm born adolescents may also adjust their educational aspirations by selecting practical rather than academic programmes to study which may hinder their future earning potential and promote prolonged family dependence (Hallin et al., 2010), although confounders such as families' SES may help to explain educational outcomes among preterm born populations (D'Onofrio et al., 2013).

Developmental disorders, such as attention deficit-hyperactivity disorder (ADHD) and autism spectrum disorder (ASD), that are prevalent among EP/VP born populations, may adversely impact interactions with both the family and peers (Lund, Vik, Skranes, Brubakk, & Indredavik, 2011; Saigal et al., 2020). For children with ASD, the family members may form the closest social contact group (Thompson & McFerran, 2015) which may further increase concerns among the parents as they aspire to promote their child's independence. ADHD symptoms may worsen at the time of adolescence (Hallberg, Klingberg, Reichenberg, & Möller, 2009). Yet, in the family context, preterm born adolescents' behaviour may be more complying (high agreeability and consensus) than the behaviour of their FT born peers (Kajantie et al., 2008). Preterm born adolescents may consume less alcohol and/or smoke less often (Kajantie et al., 2008; Saigal, Pinelli, Hoult, Kim, & Boyle, 2003). Consequently, parent-adolescent conflicts, commonly associated with increased adolescent independence and risky

behaviours, such as substance abuse, may be less frequent or appear later among families with adolescents born preterm (Moore, Taylor, Klein, Minich, & Hack, 2006; Singer et al., 2007; 2010).

As discussed in the introductory chapter of this thesis, researchers among prematurity have hypothesised that the impacts of adverse birth experience, parental perception of child vulnerability, and potentially altered early parent-child relationship among parents of EP/VP born children may carry forward as altered parenting behaviour. These experiences may impact parent outcomes by increasing parental concerns of child health and/or abilities. Preterm born adolescents have rated their own HRQoL more favourably in comparison with their parents' reports (Hallin et al., 2010; Indredavik, Vik, Heyerdahl, Romundstad, & Brubakk, 2005; Lund et al., 2011; Saigal et al., 2003). Although the favourable adolescent ratings may be impacted by preterm born children's increased readiness to give socially acceptable answers (Allin et al., 2006) and by exclusion of adolescents with most severe morbidities from follow-up studies (Wolke, Chernova, et al., 2013). Nonetheless, mothers of 16 to 17-year-old EP/ELBW born Danish adolescents perceived their preterm born adolescent to have more social anxiety in comparison with the mothers of their FT born peers, while the adolescent self-ratings were comparable (Somhovd, Esbjorn, Hansen, & Greisen, 2018). Parental ratings of child anxiety and social difficulties remained stable over time from childhood (8 years) to adolescence (14 years) among parents of ELBW adolescents from Cleveland, Ohio (Taylor et al., 2015).

Adolescence and parent outcomes – Parents and the familial environment can have a pivotal role in supporting child adaptation over this period (DiClemente et al., 2001; Gross, Mettelman, Dye, & Slagle, 2001). For example, among the general population, age-appropriate parental control and monitoring may reduce adolescent risk-taking behaviours (Anderson, 2008; DiClemente et al., 2001). Yet being a parent of an adolescent may impact parental wellbeing, for example provoke parenting stress (Anderson, 2008). Although all parents experience stress, the evidence has suggested that parenting stress may be more severe among parents of children with long-term health concerns or behavioural problems (Barroso, Mendez, Graziano, & Bagner, 2018). Parenting stress can be adaptive if it prompts the parent to find new coping strategies to adjust to the child's changed behaviour and, consequently support the parent-child relationship. Stress, however, may be harmful if it alters parenting behaviour in a way that hinders the parent-child interaction (Almogbel et al., 2017) and consequently provokes prolonged negative parental experiences. Among mothers of 7-year-old Canadian children born VP (≤ 32 weeks' gestation) higher parenting stress scores were associated with escape/avoidant coping style (Linden et al., 2015). As discussed earlier in this

thesis, a balance between parental perception of how demanding the child is to parent (impacted by both child and parent factors as well as contextual factors such as cultural appropriateness of child behaviour) and the resources available to support the parenting task (family and contextual factors) is important; perceptions of low levels of resources and challenging adolescent behaviour and/or life situation may provoke adverse parent outcomes (Deater-Deckard, 1998).

Parenting stress and associated factors - Although studies of parenting stress and adolescent behaviour have been uncommon (Barroso et al., 2018), the evidence has indicated that rather than physical chronic child health condition/s, or severity of those conditions, adolescent behavioural and/or psychological disorders are more strongly associated with increased parenting stress among at-risk populations (Barroso et al., 2018; Majnemer, Shevell, Rosenbaum, Law, & Poulin, 2007). Findings from a study among families with 9-year-old children with cerebral palsy (45% were born preterm, mean GA 31 weeks) suggested that child behavioural difficulties and psychosocial adjustment were more strongly associated with high parental distress (PSI) scores than the level of child physical disability (Majnemer et al., 2007). The lesser impact of child physical disability on parenting stress may have suggested that parental perception of child vulnerability associated with altered parenting behaviour and parental anxiety among parents of preterm born younger children (Miles et al., 1998) may become a lesser factor when the child gets older (Tallandini et al., 2015). Yet parental perception of child vulnerability may persist even though child's health status has improved (Thomasgard & Metz, 1997) and might be impacted more by maternal psychological health rather than child factors (Tallandini et al., 2015). Nonetheless, adolescents born EP/VP may experience multiple child risk factors simultaneously such as neurosensory impairments and psychiatric and neurodevelopmental disorders (D'Onofrio et al., 2013; Drotar et al., 2006), all which can pressurise the parent-child relationship.

Parents of 150 three to 18-year-old children (52.7% were above the age of 12 years) attending special educational needs (SEN) schools in the US, were more likely to report high parenting stress (PSI-SF score $\geq 85^{\text{th}}$ centile) if they perceived their children to have functional impairments (Columbia Impairment Scale score ≥ 16). This association was further strengthened if the parents reported having any personal health concerns but decreased among parents with higher than college level education. The authors postulated that parents with a higher level of education may have been cognitively more prepared to respond to their children's behaviours, but also that they may have had more financial resources available to manage the parenting task in comparison with parents with lower education (Almogbel et al., 2017). Their results demonstrated that both parent and child factors were impactful in

parenting behaviour and parent outcomes. However, in their study, all variables were parent-reported and therefore, it may be that the parents who experienced high stress were also more likely to perceive their child's behaviour as challenging. Empirical studies, as well as theory, have suggested that the relationship between parenting stress and child behavioural difficulties may be bidirectional (Barroso et al., 2018; Gerstein & Poehlmann-Tynan, 2015; Sameroff & Mackenzie, 2003). Among an at-risk population of fostered/adopted children, parent perceived child behavioural difficulties (Strengths and Difficulties Questionnaire (SDQ)) correlated highly with clinically significant levels of parenting stress (PSI-SF) (McSherry, Fargas Malet, & Weatherall, 2019). Thus, for parental experiences, the perception of (or appraisal of) the demandingness of child behaviour may be important, regardless of whether it may meet the diagnostic criteria (Guyard, Michelsen, Arnaud, & Fauconnier, 2017).

When considering resources to support the parenting task, Anderson (2008) examined child factors, parent factors, and contextual factors predicting parenting stress levels among 824 parents of 10 to 18-year-old adolescents at high risk of substance abuse recruited from communities across the US. According to their results, parent reported child factors, particularly social skills, and problem behaviour, were more predictive of parenting stress than parent factors such as gender, ethnicity, or parent health. In their analysis, in the final model, only presence of any parental health concern (yes/no) was associated with increased parenting stress levels (PSI). The contextual factors included family structure, family conflict, family cohesion (i.e., support, parental attachment, emotional closeness), family involvement, social support, parent education, and household income. Only lower parent educational attainment, increased family conflict, and decreased family involvement were associated with increased parenting stress scores (Anderson, 2008).

Guyard et al. (2017) assessed family adaptation to child disability among 220 families of 13 to 17-year-old adolescents with cerebral palsy recruited from population-based registries of four European countries. According to their results, family dysfunction, parental unemployment, adolescent behavioural disorders (more than motor impairments) and younger child age predicted higher PSI scores in the parental distress domain. Lower parental distress scores were associated with increased parental qualifications, respite from care, and positive parental attitudes (Guyard et al., 2017). In another study of predictors of parental distress (Brief Symptom Inventory) among parents of three to 12-year-old children with cerebral palsy, spina bifida, or limb deficiencies, the severity of child disability was not associated with parents' BSI scores, rather, among the parents with children with cerebral palsy, family roles (McMaster Family Assessment Device), recent stressful life events, and younger child age predicted higher BSI scores. The authors interpreted this to suggest that how families'

allocated tasks and adjusted to their child's disability were important factors in positive parental experiences (Wiegner & Donders, 2000). Indeed, division of household chores that is perceived as 'disproportioned' have been found to negatively impact couples' relationship (Eerola et al., 2021).

As an alternative to the so-called 'risk model' designs presented above, other approaches in research on parenting behaviour and child development have focused on better understanding factors such as resilience (understood as a set of factors allowing an individual to successfully overcome negative effects of risk exposures and to avoid negative trajectories relating to those risks) in promoting positive adaptations (Fergus & Zimmerman, 2005) which can buffer against negative outcomes among both the parents and their preterm born children (Treyvaud, 2014). In addition, as discussed in detail earlier, more recent scholarly attention has focused on critically examining the deterministic views of parenting behaviour (i.e., that the decisions made by parents and their actions directly impact the future 'success' of their child as an adult), and those of children as inherently 'at risk' of dangers external to the immediate parent-child relationship (Dermott, 2012; Smith, 2010).

Summary - In conclusion, adolescence is an important time of increased child independence. Adolescent behaviour among children born preterm may differ from adolescents born FT. Behavioural changes in adolescence can provoke parenting stress, particularly among parents of children born preterm when challenges are frequent. Increased parental distress may alter parenting behaviour and the parent-child relationship. Adverse parent outcomes in adolescence among parents of children with long-term morbidities have been associated more strongly with child behavioural difficulties and psychological disorders than with the child's physical health status. Family and contextual factors, such as higher parental educational level and family income may buffer against negative parent outcomes, for example by increasing parental capacity to respond to adolescent behaviours (i.e., support coping strategies). Lower parental physical health or increased family conflict may further provoke negative outcomes among parents and their adolescent children.

2.3 Systematic Review Aims and Questions

Predominant topics in the research on long-term parent outcomes following preterm birth have been the influence of birthweight and/or neonatal medical risk level, child disability, parental adaptation, child age, and family resources on various parent and family outcomes. As the outcome measures used to assess the parent impact have described merely aspects of the parenting experience, they have resulted in a mixture of recommendations for possible

interventions or future research. Assembling findings from different studies may help to better describe parental experiences as a whole and improve understanding of factors that influence parents' experiences and their interlinkages. The purpose of this review was to collate previous research findings. As studies assessing long-term parent outcomes following preterm birth have generally been non-randomised studies with varying designs, settings, and outcome instruments, a narrative approach was chosen as the most appropriate method of data synthesis (Mays, Pope, & Popay, 2005; Rodgers et al., 2009; Tacconelli, 2010).

The aim of this systematic review of literature was to identify all studies describing and/or assessing long-term parent outcomes following EP/VP birth as their primary outcome. Due to infrequent survival, birth cohorts that have reported long-term outcomes for strictly EP survivors have been scarce. Until more recently, studies have included infants based on their birthweight and lacked data on gestational age. Consequently, these longitudinal cohort studies have not been able to distinguish outcomes between EP and VP born participants. Thus, studies with both EP and VP cohorts were included in this review. The review included all studies of parents of children 1) who were born extremely (less than 28 weeks of gestation) or very (28 to 32 weeks of gestation) preterm or/and had extremely (<1000 grams) or very (<1500 grams) low birthweight (World Health Organisation, 2018), 2) who were aged between 8 to 25 years at the time of the study assessment and 3) the study described and/or assessed parent outcomes as its primary outcome measure.

This review addressed two review questions:

Primary review question: What are the parent outcomes at the time the preterm born child reaches adolescence?

Supportive review question: What factors are associated with the long-term parent outcomes?

This chapter describes the methods of this systematic narrative review of literature and presents a summary of the search results. I will describe the characteristics of the identified studies and discuss the relationships between the findings.

2.4 Methods

2.4.1 Eligibility criteria

To meet the inclusion criteria the study had to describe or assess one or more parent outcomes as its primary outcome measure. The primary outcomes of interest were any parent or family

outcomes including (but not restricted to) parenting stress, parental distress, parental anxiety, parent physical and mental health outcomes, parent behaviour, parenting skills and competence, parenting style, parent-child relationship, parent experiences, family impact, family burden, family functioning and families' socio-economic or social outcomes. All study designs, except interventional studies, assessing or describing long-term (child age between 8 and 25 years) parent outcomes following extremely or VP birth (as defined above) were included. Interventional studies were excluded due to the difficulty to extract outcome data into a narrative data synthesis for parents in the non-interventional arm only.

Since survival of EP infants has been infrequent until relatively recently, studies of families with children born VP were also included. Table 2-1 below details the general classifications of EP and VP birth as defined by the WHO (World Health Organisation, 2018) and the stricter inclusion criteria employed in the EPICure cohorts (Costeloe et al., 2012). EPICure 2 study's eligibility criteria were restricted to births less than 27 weeks of gestation as the medical management of infants born at 27 weeks of gestation had generally been successful and the limited size of the cohort better facilitated long-term follow up (Costeloe et al., 2012). In this review, the WHO definitions were utilised.

Table 2-1 Definitions of extremely and very preterm birth

Category	Definitions (weeks + days)		
	WHO	EPICure	EPICure 2
EP (EP)	≤27 + 6	22 to 25 + 6	22 to 26 + 6
VP (VP)	28 to 31 + 6	-	-
Extremely low birthweight (ELBW)	<1000 g	-	-
Very low birthweight (VLBW)	<1500 g	-	-

The age range was decided based on previous research among EP/VP survivors; the age of 8 years was considered to be the earliest possible onset of pubertal growth changes (Wehkalampi et al., 2011), whereas EP/VP survivors have been found to transition to independent living and adulthood at later ages compared to FT born adolescents (Kajantie et al., 2008). Studies with mixed age populations, such as preterm born children aged 6 to 12 years, were included if the age range studied involved children aged between 8 and 25 years. When multiple publications reported findings by using data of children from the same birth cohort this was clearly stated. Some studies reported findings for children with varying degrees of prematurity, in those cases, only the results for children born EP or VP were reviewed. If this was not possible, the study was excluded.

The data synthesis included all identified studies written in the English language and published since 2000 in peer-reviewed journals. The year 2000 was chosen as a preliminary review of literature had indicated that few studies of long-term outcomes were published earlier and, on occasions, were characterised by low quality of reporting. Non-English language studies were initially reviewed, so long as the studies provided an English language abstract, to consider whether there was a potential language bias, but these studies were not included in the data synthesis. Similarly, conference papers were included in the initial searches in order to identify ongoing research but were excluded from the synthesis (Tacconelli, 2010). The eligibility criteria are detailed in Table 2-2 below.

Table 2-2 Eligibility criteria for the systematic narrative review of literature on long-term parent outcomes following very preterm birth

	Child birthweight <1500 grams
	Child gestational age <32 weeks
	Child age 8 to 25 years
Inclusion criteria	Primary outcome measure parent outcome/s
	A peer-reviewed publication
	Publishing language English
	Year of publishing 2000 or later
Exclusion criteria	Interventional study design

2.4.2 Search strategy

Advice from a subject librarian was sought when planning and finalising the search strategy. The searches were carried out on Medline, Embase and PsychINFO databases using Medical Subject Headings (MeSH) with the year of publication 2000 or later as a restriction. High level MeSH terms for prematurity (premature, low birth weight), parent outcomes (parent, family, mother, maternal, father, paternal, sibling) and adolescence (adolescent, young adult, puberty, development) were searched using the OR operator and ‘explode’ function to include all sub-headings. The searches were combined using the “AND” operator (Tacconelli, 2010). The MeSH search terms, and the search configuration are detailed in Appendix A.

The MeSH terms assigned to the relevant articles already selected into the review were screened in order to ensure that they matched the subject headings used in the search (Tacconelli, 2010). A citation search was conducted by screening bibliographies of the articles already included in the review and by screening citation indexes using the Web of Science (Denison et al., 2013; Tacconelli, 2010). The references were managed using both Mendeley and EndNote programmes, they were charted using Microsoft Excel and the final narrative data synthesis was carried out in NVivo 12 Software. The searches were first carried out in April 2018 and repeated in May 2019 to incorporate most recent publications.

2.4.3 Data extraction

The data extraction progressed in two stages. Stage 1 included an initial screen in which titles and abstracts of the articles were reviewed. In Stage 2 the full papers, identified as potentially relevant, were read and data were extracted if the inclusion criteria were met.

The supportive review question: *“What factors are associated with long-term parent outcomes”* yielded a large amount of potentially relevant studies assessing 1) EP/VP born adolescents’ (aged 8 years or above) outcomes or 2) parent or child outcomes before the EP/VP born child reached the age of eight years. As the purpose of this review was to collate the findings of long-term parent outcomes following EP/VP birth in adolescence, and a narrative synthesis of such a large number of manuscripts was not possible, the publications in the supportive review question were included by retrospectively reading and reviewing them. This was done to assess how they related to the main themes that emerged from the core sample of articles in the main review question: *“What are the parent outcomes at the time the preterm born child reaches adolescence”*. The articles in the supportive review question were used to support the Discussion section. The items presented in Table 2-3 below were extracted from the articles (data extraction tool based on Denison et al. (2013)). The data extraction tool was piloted before its use.

Table 2-3 Data extraction items for the systematic narrative review of literature on long-term parent outcomes following very preterm birth

Article details	Paper title
	Authors
	Type of publication
	Journal name
	Year of publication
	Country of origin
Study details	Study design
	Aims and objectives of the study
	Methodological orientation and theory of qualitative studies
	Study time period
	Study geographical location
	Inclusion and exclusion criteria
	Length of follow-up
	Recruitment procedures used
	Target population and final number of subjects studied for outcome
Data collection time points	
Participant characteristics	Family socio-economic status and SES measurement tool
	Child age
	Child gender
	Child ethnicity
	Child gestational age
	Birthweight
	Multiple status
	Parent age
	Parent gender
Parent ethnicity	
Study results	Description of outcome measurements
	Description of measurement instruments
	Confounding factors adjusted for
	Results of study analysis
	Qualitative data analysis methods used
	Narrative findings of qualitative studies
	Conclusions of study

2.4.4 Risk of bias assessment

According to the Centre for Reviews and Dissemination (CRD) systematic review guide (2010) the quality assessment of published research should consider whether the study design has been appropriate considering the research objectives described, and assess risk of bias and other issues related to quality, such as the choice of outcome measures or generalisability of the results (Tacconelli, 2010). The risk of bias assessment supports the interpretation of findings from varying studies and explains possible differences in the findings (Tacconelli, 2010; Walsh & Downe, 2006).

I assessed risk of bias of all eligible quantitative studies using an instrument published by the CLARITY (Clinical Advances Through Research and Information Translation) group, as this instrument included most relevant criteria to assess risk of bias in cohort studies (CLARITY Group, 2021). The instrument presents an overall assessment of risk of bias without a summary score by utilising eight questions relating to selection of exposed and non-exposed cohorts, assessments of exposure, prognostic, and outcome variables and possible presence of the outcome at the start of the study, and considerations of follow-up rates and possible co-interventions. Each question is rated as definitely yes (low risk of bias), probably/mostly yes, probably/mostly no, or definitely no (high risk of bias). The risk of bias was evaluated at study level and each item was considered as equally important. A copy of the instrument is included in Appendix B. The quality of qualitative studies' reporting was assessed using a tool presented by Walsh and Downe (2006). This instrument was chosen as it supported narrative data synthesis particularly well.

2.4.5 Data synthesis and reporting

The data analysis in NVivo 12 followed methods of thematic analysis (Nowell, Norris, White, & Moules, 2017). Articles were uploaded to NVivo 12 and read while coding all findings referring to 1) long-term parent outcomes and 2) the authors' interpretations of those findings. The quotes were then read and re-read while the study findings and authors' interpretations were coded into emerging themes such as 'disability', 'parental adjustment' and 'family resources'. First, a preliminary data synthesis was performed by charting all findings of long-term parent outcomes by type of outcome, such as psychological impact, family impact, and/or parent-child relationship outcomes, and by direction of impact (negative, positive, and/or no impact). Effect sizes were presented when data were available in the original publication. Then, relationships between findings and the authors' interpretations of these findings were narratively explored per the emerged key themes (Rodgers et al., 2009; Tacconelli, 2010).

2.4.6 Ethics and study registration

This review did not contain any studies with human participants performed by the author. Details of the protocol for this systematic review were registered on PROSPERO and can be accessed at https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42019132802. As two separate publications of findings of this review are intended as per the review questions, the current PROSPERO registration relates only to the main review question. The registration has been updated to reflect protocol changes of exclusion of RCT and a change in the risk of bias assessment tool. Amendment history is recorded on the website.

2.5 Results

2.5.1 Study characteristics

Figure 2-1 below presents the screening process and the search results. The database search in PsycINFO returned 428 potentially relevant publications, in Medline 1840 publications and in Embase 15675 publications. The initial screening of all these items resulted in 166 potentially relevant publications. During the first stage, 64 of these publications were excluded as they did not meet the inclusion criteria, mostly because they involved preterm born children from older gestational ages. The one hundred and two remaining publications were assigned to their relevant review questions; 71 were assigned to the supportive review question and 31 articles to the main review question. The bibliography screening and the citation searching yielded two more eligible publications in the main objective. After reading the 33 full manuscripts, further 16 articles were excluded as they did not meet the inclusion criteria. The final sample consisted of 17 publications of which four were longitudinal studies, 11 had a cross-sectional design, one was a nationwide population-based registry cohort, and one was a qualitative study. Table 2-5 below presents descriptive summaries of the included studies.

The identified publications included 10 longitudinal birth cohorts of children born less than 32 weeks of gestation in Germany (1 publication) (Jaekel et al., 2012), Australia (2 publications) (Burnett et al., 2017; Yaari, Treyvaud, Lee, Doyle, & Anderson, 2019), Norway (1 publication) (Indredavik et al., 2005), Finland (1 publication) (Pyhala et al., 2011), Canada (2 publications) (Saigal et al., 2010; Saigal, Rosenbaum, et al., 2000) and in the US (7 publications of 4 birth cohorts; 3 cohorts with 2 publications (Drotar et al., 2006; Moore et al., 2006; Singer et al., 2007; 2010; Taylor et al., 2001; Wightman et al., 2007) and a cohort with one publication (Brown, Kilbride, Turnbull, & Lemanek, 2003). Six of the publications from the US reported findings for children born in Cleveland, Ohio. The publications reported both longitudinal and cross-sectional analyses of the birth cohorts' data. The four longitudinal

publications compared outcomes between two or more timepoints. These timepoints were from one month to 14 years (7 timepoints) (Singer et al., 2010), from two to 13 years (3 timepoints) (Yaari et al., 2019), from 11 to 14 years (4 timepoints) (Moore et al., 2006) and at six and eight years (Jaekel et al., 2012). Of the 10 birth cohorts, seven were born before 1990, one recruited infants born between 1989 and 1991, and three in the 1990s and after (see Table 2-4 below). Sample sizes varied from 26 parents of EP and FT born children to 267 parents of VP born children and 298 parents of their FT born comparator children. At the time of the studies the children were aged between 8 and 25 years.

The only cross-sectional study not based on a birth cohort reported findings for 78 mothers whose children were born between 1995 and 2006 in Israel (Zerach, Elsayag, Shefer, & Gabis, 2015). At the time of the assessment the children were between four and 16 years. The families were identified from a hospital database and did not have a comparison group. The nationwide population-based registry cohort study reported parent outcome of altered reproductive behaviour following preterm birth for caregivers of 1,019 children born less than 32 weeks of gestation between 1987 and 1990 in Finland (Alenius et al., 2018). The qualitative study identified interviewed parents of children born before 28 completed weeks of gestation in Denmark between 1992 and 1995 (Lou et al., 2009). At the time of the interviews the children were between seven and 10 years of age.

The identified studies assessed parent outcomes of mothers (6 studies), primary caregivers (3 studies) and parents (8 studies) of children born EP/ELBW or VP/VLBW.

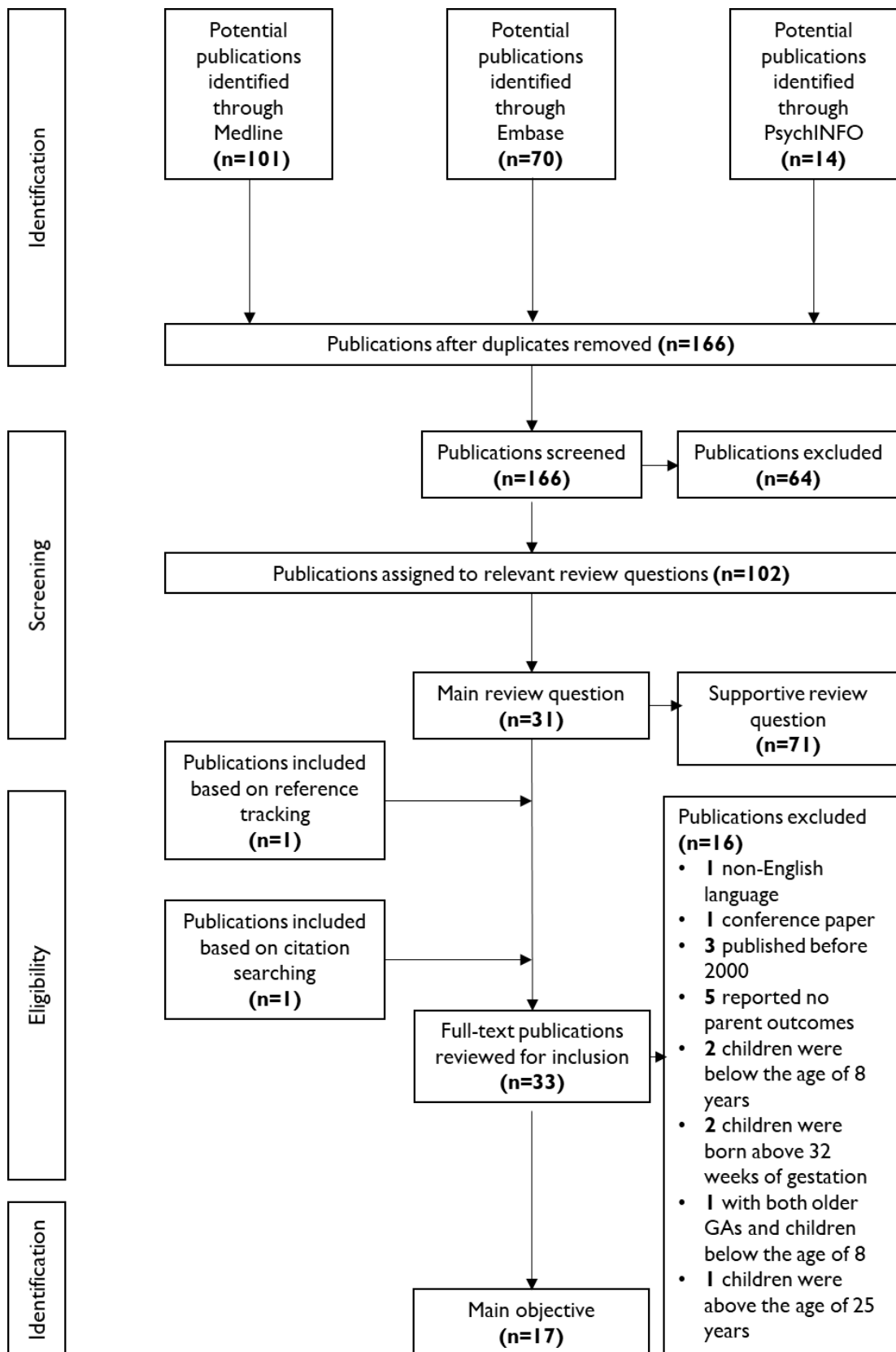


Figure 2-1 Flowchart showing the search results and screening process for the systematic narrative review of literature on long-term parent outcomes following VP birth

Table 2-4 Publications based on longitudinal birth cohort studies by author, recruitment area and period (n=10)

	Authors	Recruitment area	Years of recruitment
1	Saigal et al; 2010 Saigal et al; 2000	Hamilton, Ontario	1977-1982
2	Pyhala et al; 2011	Helsinki, Finland	1978-1985
3	Taylor et al; 2001 Moore et al; 2006	Cleveland, Ohio	1982-1986
4	Brown et al; 2003	Kansas City, Missouri	1983-1984
5	Jaekel et al; 2012	South Bavaria, Germany	1985-1986
6	Indredavik et al; 2005	Trondheim, Norway	1986-1988
7	Singer et al; 2007 Singer et al; 2010	Cleveland, Ohio	1989-1991
8	Burnett et al; 2017	Victoria, Australia	1991-1992
9	Drotar et al; 2006 Wightman et al; 2007	Cleveland, Ohio	1992-1995
10	Yaari et al; 2019	Melbourne, Australia	2001-2003

Table 2-5 Studies describing or assessing long-term parent outcomes following VP birth (n=17) included in the systematic review of literature by type of publication and child's age at the assessment

Publication	Country	Description of study aim/s	Participants	Child age	Main results
Longitudinal reports					
Singer et al. 2010	USA	To assess the longitudinal changes of parenting stress and coping among mothers of VLBW children from birth to adolescence	Mothers of 113 VLBW children with BPD Mothers of 80 VLBW children with no BPD Mothers of 122 FT born children)	Assessed at 7 timepoints between 1 month and 14 years	1) VLBW affected several parent outcomes such as parenting stress, coping mechanism, feelings of mastery and financial resources 2) Mothers of VLBW children had slower and lower educational attainment after the childbirth compared to the mothers of NBW children 3) Mothers of VLBW-BPD children experienced higher levels of mastery compared to NBW children's mothers at child's age of 14 years 4) Parenting stress levels among mothers in all groups varied over time
Yaari et al. 2019	Australia	To assess trajectories of psychological distress in mothers of VP and FT born children and examine predictors of distress among the VP mothers over time	185 mothers of 224 VP children 77 mothers of 79 FT born children	Assessed at 3 timepoints between 2 and 13 years	1) At all timepoints mothers of VP children reported higher psychological distress; the distress was stable over time and group differences persisted 2) Maternal and family factors, such as social risk level, were more strongly associated with maternal psychological distress compared to child neurodevelopmental impairment or multiple birth
Jaekel et al. 2012	Germany	To assess the quality of mother-child interaction among dyads of VLBW/VP and FT born children and their mothers	267 VLBW/VP born children and their mothers 298 FT born children and their mothers	Assessed at 2 timepoints at 6 and 8 years	1) Mothers of VLBW/VP children were less sensitive and more verbally controlling compared to the FT born children's mothers; maternal behaviour differences disappeared when controlled for child IQ 2) Increased maternal sensitivity at 1 st timepoint promoted increased child persistence during the 2 nd assessment; the association increased with decreasing child IQ, demonstrating that the mothers positively adjusted their parenting behaviours 3) Lower maternal sensitivity and higher verbal control were not associated with higher family adversity

Table 2-5 (continued)

Publication	Country	Description of study aim/s	Participants	Child age	Main results
Moore et al. 2006	USA	To investigate changes in family sequelae of VLBW across four follow-up assessments between children's ages of 11 and 14 years	Caregivers of 44 children born with BW <750 grams Caregivers of 42 children born with BW between 750 and 1499 grams Caregivers of 47 children born FT ¹	Assessed at 4 timepoints between 11 and 14 years	1) Parents of VLBW adolescents reported more long-term family burden and parent psychological distress than parents of NBW adolescents 2) The family burden and parental distress varied differently according to the level of low BW and family resources and SES 3) Several families with children born with VLBW did not report family burden or parental psychological distress
Cross-sectional reports					
Drotar et al. 2006	USA	To compare the family impact of school-aged children born ELBW or NBW and to assess predictors of family impact amongst the ELBW children's families	Primary caregivers of 219 ELBW children Primary caregivers of 176 NBW children	8 years	1) Total negative family impact, including financial impact, caretaker burden and familial burden were greater among ELBW children's families than in families with children born with NBW 2) The differences in the family impact were not explained by differences in family resources 3) Child's functional limitations and neurodevelopmental impairments were most predictive of higher negative family impact, whereas BW or neonatal risk level had no effect
Singer et al. 2007	USA	To compare the levels of stress and coping, and their determinants, among mothers of VLBW children with and without BPD, and their FT born controls' mothers	Mothers of 94 VLBW children with BPD Mothers of 71 VLBW without BPD Mothers of 97 FT born children	8 years	1) Mothers of VLBW children reported fewer family strains and less parent-child conflict compared to the mothers of FT born children 2) Mothers of VLBW children were more concerned about their child's health than mothers of FT born children 3) Mothers of VLBW children had adapted their coping styles since previous assessments and presented less denial or mental disengagement as coping strategies 4) VLBW birth reduced mothers' further educational attainment and work opportunities

Table 2-5 (continued)

Publication	Country	Description of study aim/s	Participants	Child age	Core results
Wightman et al. 2007	USA	To assess the level of parental protectiveness and associated factors among parents of ELBW children and parents of NBW children	Primary caregivers of 217 ELBW children Primary caregivers of 176 NBW children	8 years	1) Parents of ELBW children reported higher total parental protectiveness and control than the parents of NBW children; an association that diminished when families with children with NSIs were excluded from the analyses 2) Parents with ELBW NSI children reported significantly more overprotection than parents with ELBW children with no NSIs
Taylor et al. 2001	USA	To assess long-term effects of VLBW on families with children with varying extent of LBW and neonatal medical risk level	Parents of 60 children born with BW <750 grams Parents of 55 children born with BW between 750 and 1499 grams Parents of 49 children born FT with NBW	11 years	1) Parents with children born <750 grams reported more difficulties in child attachment, less parenting competence, and higher rates of adverse family outcomes than parents with children born FT 2) The outcomes for families of children born 750-1499 grams fell between those of children born <750 grams and FT, suggesting a gradient effect of BW 3) Concurrent child health problems were the strongest predictor of negative family sequelae, stronger than BW or neonatal medical risk level
Brown et al. 2003	USA	To assess parents' perceptions of the impact of ELBW birth on long-term family functioning	26 ELBW adolescents and their parents 26 NBW adolescents and their parents	14 years	1) No differences in family impact between the BW groups were found 2) Parents reported having experienced negative family impact on social life and financial domain in child's infancy, but not in adolescence
Indredavik et al. 2005	Norway	To assess the effect of BW on adolescent quality of life, parent-child relationship and parent mental health amongst mothers and fathers of adolescents born VLBW, SGA or NBW	56 VLBW adolescents and their parents 60 SGA adolescents and their parents 83 NBW control adolescents and their parents	14 years	1) Parents mental health did not vary between the groups of parents; all parents perceived their own parenting practices similarly 2) Parents of VLBW adolescents reported higher impact on their emotional health and limited time for personal needs compared to parents of NBW adolescents; the impact was increased if the child had CP or psychiatric disorder 3) All study groups reported similar levels of relational warmth

Table 2-5 (continued)

Publication	Country	Description of study aim/s	Participants	Child age	Core results
Burnett et al. 2017	Australia	To compare ELBW and NBW adolescents and their parents' perceptions of family functioning	193 EP/ELBW adolescents and their 178 parents 151 NBW adolescents and their 145 parents	15-20 years	1) EP/ELBW families perceived their family relations similarly or more positively than NBW families 2) Both the EP/ELBW adolescents and their parents perceived their families to be more protective (organised and controlled) 3) Parent reported family control or conflict were not associated with increased anxiety or depression among the EP/ELBW adolescents
Zerach et al. 2015	Israel	To assess perceived levels of parenting stress and PTSD symptoms among mothers of ELBW born children with varying levels of morbidities	Mothers of 78 ELBW children	4 to 16 years	1) Mothers of ELBW children with disabilities reported higher levels of parenting stress than mothers of ELBW children with no developmental difficulties 2) Lower level of education, negative life events and child disability (regardless of level) predicted higher parenting stress 3) A quarter of the mothers reported symptoms of PTSD at a level of diagnosis 4 to 16 years after the birth
Saigal et al. 2000	Canada	To compare the impact of burden of illness on families of adolescents born with ELBW with those whose children were born with NBW	Parents of 145 ELBW adolescents Parents of 123 NBW adolescents	12 to 16 years	1) Mothers of ELBW teenagers had adjusted well to family life and work 2) Mothers of ELBW adolescents experienced negative financial impacts following EP birth 3) Mothers of ELBW adolescents reported increased impact on their emotional health compared to the mothers of NBW children; negative emotional impact diminished when children with impairments were excluded 4) Higher proportion of parents of impaired children reported that the experience of having an ELBW child had improved their feelings of themselves
Pyhala et al. 2011	Finland	To examine differences between recalled parenting behaviour by VLBW YAs and their parents and NBW YAs and their parents	98 mothers and 77 fathers of VLBW YAs 95 mothers and 77 fathers of FT YAs	Mean age 22.5 years	1) Parents of VLBW YAs recalled their parenting to have been more supportive 2) VLBW YAs recalled their parents to have been more protective; especially women who perceived their parents to have been also more authoritarian

Table 2-5 (continued)

Publication	Country	Description of study aim/s	Participants	Child age	Core results
Saigal et al. 2010	Canada	To assess the impact of illness on families of YAs born with ELBW or NBW; to assess changes in family burden since adolescence and its association with NSI	Mothers of 130 ELBW YAs Mothers of 126 NBW YAS	22-25 years	1) Mothers in both groups reported similar scores in family impact, family dysfunction, state anxiety, social support, and maternal physical and mental health 2) Families with ELBW YAs with NSIs reported less family dysfunction compared to the families of ELBW YAs with no NSIs 3) Mothers of ELBW YAs reported significant negative impact on parents' work
Population-based registry study					
Alenius et al. 2018	Finland	To assess the impact of preterm birth across all gestational ages on the number of subsequent children	Mothers and fathers of 328 EP children and 691 VP born children	n/a	1) Preterm birth had altered the reproductive behaviour of both parents by reducing the final number of children in the family 2) A gradient effect with level of prematurity and number of subsequent children existed; mothers of surviving EP children had fewest subsequent children 3) The associations were not affected by SES or high-risk pregnancy factors; infant disability had only a minor effect on subsequent parent reproductive behaviour
Qualitative study					
Lou et al. 2009	Denmark	To explore positive family experiences following EP birth	Mothers and fathers of 14 EP children	7 to 10 years	1) Parents experienced that their EP children had contributed positively to the family life 2) The mothers and fathers of EP children viewed their child's performance differently to clinical diagnoses
¹ number of families assessed at all 4 timepoints BPD Bronchopulmonary dysplasia BW Birthweight CP Cerebral palsy ELBW Extremely low birthweight FT Full-term LBW Low birthweight			NBW Normal birthweight NSI Neurosensory impairment PTSD Post-traumatic stress disorder SES Socio-economic status VLBW Very low birthweight VP Very Preterm YA Young adult		

2.5.2 Risk of bias

As presented in Figure 2-2 below, with two exceptions (Alenius et al., 2018; Jaekel et al., 2012), I considered all cohort studies to have probably or definitely high risk of bias on two or more of the items when assessed with the Tool to Assess Risk of Bias in Cohort Studies (CLARITY Group, 2021). This indicated some methodological limitations in most studies. Most common methodological challenges were the selection of exposed and non-exposed cohorts from different populations (Q1), uncertainty regarding the presence of the outcome of interest at the start of the study (Q3), high lost to follow-up rates (Q7), and uncertainty regarding other possible co-interventions during long follow-up periods (Q8) (Figure 2-2).

Publication	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	
Singer et al. 2010	++	++	-	+	+	+	-	+	++ Definitely yes (low risk of bias) + Probably / Mostly yes - Probably / Mostly no -- Definitely no (high risk of bias)
Yaari et al. 2019	+	++	--	+	+	+	-	-	
Jaekel et al. 2012	++	++	++	++	+	++	+	-	
Moore et al. 2006	-	+	--	+	+	+	+	+	1 Was selection of exposed and non-exposed cohorts drawn from the same population? 2 Can we be confident in the assessment of exposure? 3 Can we be confident that the outcome of interest was not present at start of study 4 Did the study match exposed and unexposed for all variables that are associated with the outcome of interest or did the statistical analysis adjust for these prognostic variables? 5 Can we be confident in the assessment of the presence or absence of prognostic factors? 6 Can we be confident in the assessment of outcome? 7 Was the follow up of cohorts adequate? 8 Were co-Interventions similar between groups?
Drotar et al. 2006	-	++	--	++	+	+	++	+	
Singer et al. 2007	++	++	-	+	+	+	-	+	
Wightman et al. 2007	-	++	--	++	+	+	++	+	
Taylor et al. 2001	-	++	--	+	+	+	+	+	
Brown et al. 2003	--	++	--	--	+	+	--	-	
Indredavik et al. 2005	++	++	--	+	+	+	+	-	
Burnett et al. 2017	++	++	++	+	+	+	-	-	
Zerach et al. 2015	<i>n/a</i>	++	--	+	-	+	-	-	
Saigal et al. 2000	-	++	++	+	+	-	++	-	
Pyhala et al. 2011	++	++	++	+	+	+	--	-	
Saigal et al. 2010	-	++	--	+	+	+	++	+	
Alenius et al. 2018	++	++	++	++	++	++	++	+	

Figure 2-2 Risk of bias of cohort studies included in the systematic review of literature on long-term parent outcomes following VP birth

Six birth cohorts selected control children from the same population at birth (Alenius et al., 2018; Burnett et al., 2017; Indredavik et al., 2005; Jaekel et al., 2012; Pyhala et al., 2011; Singer et al., 2007; 2010) limiting the risk of selection bias (Q1). One study recruited controls at birth as well as at the children's age of two years (Yaari et al., 2019), and other studies at

school age. At school age, the index and control children were commonly matched for age, sex, and ethnicity by utilising either an element of random selection, for example, studies stated to have ‘selected at random’ from a list of eligible students (Saigal, Burrows, Stoskopf, Rosenbaum, & Streiner, 2000), or by distributing recruitment flyers at schools (Brown et al., 2003). One longitudinal birth cohort study had re-matched lost to follow-up control participants (Hack et al., 1994; Moore et al., 2006; Taylor et al., 2001), which might have introduced a particularly high risk of selection bias, especially as attrition was higher among families with low SES; a factor independently associated with the outcome of interest (Moore et al., 2006; Taylor et al., 2001).

Overall, three birth cohorts, including the population-based registry cohort study, reported high follow-up rates ($\geq 80\%$) (Alenius et al., 2018; Drotar et al., 2006; Saigal, Burrows, et al., 2000; Saigal et al., 2010; Wightman et al., 2007) and three studies reported good follow-up rates ($>60\%$) (Indredavik et al., 2005; Jaekel et al., 2012; Moore et al., 2006; Taylor et al., 2001) (Q7). In some cases, attrition was reported imprecisely; for example, studies stated the total rates of lost to follow-up without detailing rates per groups of parents (Singer et al., 2007; Singer et al., 2010; Yaari et al., 2019). Brown et al. (2003) followed up less than 50% of their original cohort, whereas Pyhala et al. (2011) reported outcome data for approximately 32% ($n=102/313$) of parents of VLBW adolescents in the original birth cohort. Details of whether or how lost to follow-up were considered in the analyses was not always reported.

Many of the publications did not mention previous parent health factors or parental eligibility criteria at birth (Q3). Thus, the outcomes of interest, such as maternal psychological disorders, could have been present at the start of the study or pre-existed the birth. As an exception, Singer et al. (2007; 2010) excluded mothers who had a mention of previous ‘major psychiatric illness’ in their medical records. Previous parental health concern was the most common prognostic factor not matched or adjusted for in the analyses.

In relation to the possible impact of co-interventions (Q8), some studies considered the influences of the wider family environment by measuring parent perceived social support, social risk, and/or life stressors and changes, but none of the publications mentioned considerations of other possible follow-up services that families might have taken part in over the years. Consistent close follow-up care among families with children born preterm, particularly among those families with children with morbidities, might have influenced parent outcomes and parenting behaviour and thus, impact outcomes among the groups of parents differentially.

For a majority of the studies, I considered the assessment of the exposure to be definitely low risk of bias (Q2) as the studies' eligibility criteria were based on infant birthweight rather than assessment of appropriateness of infant size for gestational age. However, when considering differences in reported long-term parent outcomes between the studies, another important issue to consider is the inability of several of the studies to distinguish participants born adequate for gestational age (AGA) from those born small for gestational age (SGA). As many of the studies recruited participants before the early 1990s, they lacked accurate data on gestational age. Only three publications separated families whose children were born SGA from those born AGA (Alenius et al., 2018; Indredavik et al., 2005; Pyhala et al., 2011). Findings from these publications further supported previous conclusions in research that intrauterine growth restriction and associated biological risk factors may influence the outcomes among these children, and potentially among their families (Indredavik et al., 2005; Jaekel et al., 2012; Pyhala et al., 2011).

The quality of reporting in the included publications was generally good. With two exceptions (Brown et al., 2003; Singer et al., 2010), the publications reported their aims and objectives clearly, they detailed the outcome measures used, and characterised the participants included in the study. Moore et al. (2006) did not report the exact definition of 'full-term' children in their study. Studies may have had sufficient power to detect a clinically important effects in the main outcome, but due to small sample sizes, many lacked power to justify numerous sub-group analyses conducted (Moore et al., 2006; Taylor et al., 2001).

Contrary to the generally good quality of reporting, the external validity of many of the studies was limited. Generalisability of the findings from the studies varied as some publications relied on cross-sectional data on families recruited from a single hospital, whereas others reported longitudinal follow-up findings based on whole population samples (Alenius et al., 2018; Jaekel et al., 2012). Whether the outcomes for families of infants born in a single hospital during a limited recruitment period can be considered representative of EP/VP born children and their families' outcomes in other settings is questionable. As seen in Table 2-4 above, the 10 publications based on longitudinal birth cohort studies reported findings from six countries, but with one exception, were restricted to a city or district.

In the single qualitative study included in this review, the authors did not explicitly state their analytical approach and failed to demonstrate reflexivity which might have hindered the reliability of their findings (Lou et al., 2009; Walsh & Downe, 2006).

2.5.3 Outcome measures used to assess long-term parent outcomes

A wide range of instruments were used to measure long-term parent outcomes (see Table 2-7 below). With one exception (Jaekel et al., 2012), the instruments relied on parent self-report. Family impact was assessed using six different questionnaires, of which one, the Impact on Family Scale (IOF), was used by three birth cohorts from Cleveland, Ohio (differing versions). As the IOF has been designed to measure the impact of child's chronic condition/s on the family, Singer et al. (2007; 2010) modified the instrument to be also applicable to families with healthy children. Taylor et al. (2001) and Drotar et al. (2006), used the same instrument, but did not report any modifications, even though their studies also included families with healthy children. The other five instruments that assessed family impact were either used by one study only, by two studies, but only sub-scales of the instrument, or reported twice for the same cohort of families.

In the family context, some of the instruments aimed to assess whether factors other than EP/VP birth, such as perceived social support, influenced long-term parent outcomes. The Life Stressors and Social Resource Inventory-Adult Form (LISRES-A) was one such instrument used by three birth cohorts. Parent psychological health was assessed by 13 questionnaires. The Brief Symptom Inventory (BSI) and the Center for Epidemiologic Studies Depression Scale (CES-D) were both used by two birth cohorts. Different sub-scales or versions of the Parenting Stress Index (PSI) were used by three birth cohorts. To assess the parents' perceptions of child health, and the parent-child relationship or the parents' relationship, only the Child Health Questionnaire Parent Form 50 (CHQ PF50) and Vineland Adaptive Behavior Scale (VABS) were used by more than one cohort. It is important to note that these instruments measure outcomes for overlapping areas, for example the PSI measures both parent psychological health and parent-child relationship. Hereafter, when referring to an instrument the corresponding abbreviation is used. A list of instruments and their abbreviations is presented in Table 2-6 below.

Table 2-6 List of instruments to measure long-term parent outcomes and their commonly used abbreviations

BSI	Brief Symptom Inventory
CBCL	Child Behaviour Checklist
CES-D	Center for Epidemiologic Studies Depression Scale
CHQ-PF50	Child Health Questionnaire Parent Form 50
COPE	COPE Questionnaire
CSI	Child Symptom Inventory
DAS	Dyadic Adjustment Scale
FAD	McMaster Family Assessment Devise
FBI	Family Burden Interview
FES	Family Environment Scale
FILE	Family Inventory of Life Events and Changes
FIQ	Family Impact Questionnaire
FS-II R	Functional Status Measure – Revised
GHQ	General Health Questionnaire
HADS	Hospital Anxiety and Depression Scale
IOF	Impact on Family Scale
LISRES-A	Life Stressors and Social Resource Inventory-Adult Form
MSPSS	Multidimensional Scale of Perceived Social Support
PBI	Parent Behavior Inventory
PBI-SF	Parental Bonding Instrument Short Form
PPQ	Perinatal Post-Traumatic Stress Disorder Questionnaire
PPS	Parent Protection Scale
PSI	Parenting Stress Index
PSI-SF	Parenting Stress Index Short Form
QuICCC	Questionnaire for Identifying Children with Chronic Conditions
SCL-90-R	Symptom Checklist-90-Revised
SF-36	Health Survey Short Form 36
SIPA	Stress Index for Parents of Adolescents
STAI	Spielberger State-Trait Anxiety Inventory
VABS	Vineland Adaptive Behavior Scale
-	Bradburn Affect Balance Scale
-	Etch A Sketch
-	Impact on the Family
-	Life Events Questionnaire
-	Ontario Child Health Study Questionnaire
-	Social Support Index

Table 2-7 Instruments used to assess long-term parent outcomes following VP birth by type of impact assessed and publication

	Singer et al; 2010	Yaari et al; 2019	Jaekel et al; 2012	Moore et al; 2006	Drotar et al; 2006	Singer et al; 2007	Wightman et al; 2007	Taylor et al; 2001	Brown et al; 2003	Indredavik et al; 2005	Burnett et al; 2017	Zerach et al; 2015	Saigal et al; 2000	Pyhala et al; 2011	Saigal et al; 2010
Family impact															
The Impact on Family Scale ^[1, 2]	x ^a				x	x ^a		x ^c							
The McMaster Family Assessment Devise ^[4]								x							x ^j
The Family Burden Interview ^d				x				x ^d							
Family Impact Questionnaire ^[5]									x ^g						
The Family Environment Scale ^[6]											x ^h				
Impact on the Family ^k													x		x ^k
Family context															
The Social Support Index ^[7]															x
The Family Inventory of Life Events and Changes ^[8]						x									
The Life Stressors and Social Resource Inventory-Adult Form ^[9]				x	x		x								
Life Events Questionnaire ^[10]												x			
The Multidimensional Scale of Perceived Social Support ^[11]	x ^f					x									

Table 2-7 (continued)

	Singer et al; 2010	Yaari et al; 2019	Jaekel et al; 2012	Moore et al; 2006	Drotar et al; 2006	Singer et al; 2007	Wightman et al; 2007	Taylor et al; 2001	Brown et al; 2003	Indredavik et al; 2005	Burnett et al; 2017	Zerach et al; 2013	Saigal et al; 2000	Pyhala et al; 2011	Saigal et al; 2010
Parent psychological health															
The Brief Symptom Inventory ^[12]				x		x		x							
The Parenting Stress Index ^[13]	x					x		x ^b							
The Stress Index for Parents of Adolescents ^[14]	x ^e														
The Parenting Stress Index Short Form ^[13]												x			
The COPE Questionnaire ^[15]	x					x									
The Center for Epidemiologic Studies Depression Scale ^[16]					x		x								x
Symptom Checklist-90-Revised ^[17]										x					
The Bradburn Affect Balance Scale ^[18]															x
The Spielberger State-Trait Anxiety Inventory ^[19]															x
The Health Survey Short Form 36 ^[20]															x
Perinatal Post-Traumatic Stress Disorder Questionnaire ^[21]												x ^l			
The General Health Questionnaire ^[3]		x ⁿ													
The Hospital Anxiety and Depression Scale ^[34]		x													

Table 2-7 (continued)

	Singer et al; 2010	Yaari et al; 2019	Jaekel et al; 2012	Moore et al; 2006	Drotar et al; 2006	Singer et al; 2007	Wightman et al; 2007	Taylor et al; 2001	Brown et al; 2003	Indredavik et al; 2005	Burnett et al; 2017	Zerach et al; 2013	Saigal et al; 2000	Pyhala et al; 2011	Saigal et al; 2010
Parents' perceptions of child health															
Child Health Questionnaire Parent Form 50 ^[22]						x				x ^m					
The Questionnaire for Identifying Children with Chronic Conditions ^[23]					x		x								
Vineland Adaptive Behavior Scale ^[24]					x		x	x							
The Child Symptom Inventory ^[25]							x								
Functional Status Measure - Revised ^[26]									x						
Child Behaviour Checklist								x							
Parent-child relationship / parenting behaviour															
The Etch A Sketch ^[27]			x												
Parental Bonding Instrument Short Form ^[28]										x					
The Parent Behavior Inventory ^[29]														x	
The Parent Protection Scale ^[30]							x								
Parents' relationship															
The Dyadic Adjustment Scale ^[31]						x									
The Ontario Child Health Study Questionnaire ^[32, 33]															x ⁱ

^a: modified version to be applicable to healthy children as well

^b: The Competence and Attachment scales only to assess the quality of parent-child interaction

^c: Impact on Family Scale version G

^d: interview schedule developed for the study based on Burgess ES, Drotar D, Taylor HG, Wade S, Stancin T, Yeates KO. The Family Burden of Injury Interview (FBI). *J Head Trauma Rehabil.* 1999;14:394-405

^e: administrated at 14 years only

^f: measured between years 2 and 14 only

^g: also used a modified version of the Family Impact Questionnaire to assess parents' perception when their children were infants (less than 3 years)

^h: partially used

ⁱ: partially used; 4 items only

^j: partially used: The general functioning subscale only

^k: a non-validated 17-item questionnaire encompassing work-, marital-, and family related issues developed for the study

^l: non-validated Hebrew translation

^m: emotional and time impact scales were used to assess also parental psychological health

ⁿ: utilised at 2 years only

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2.5.4 Long-term parent outcomes following very preterm birth

In the long-term, the experience of parenting an EP/VP born child altered the parents' reproductive behaviour by reducing the number of subsequent children (Alenius et al., 2018), modified the family life by increasing child-related family burden (Moore et al., 2006; Taylor et al., 2001) and parenting stress (Singer et al., 2007; 2010; Zerach et al., 2015). The parents reported decreased emotional health (Indredavik et al., 2005; Saigal, Burrows, et al., 2000), but generally did not experience increased mental health problems compared to parents of FT born children (Indredavik et al., 2005; Singer et al., 2007; Wightman et al., 2007). The experience of parenting an EP/VP born child also altered perceived parenting behaviour; both the parents and their preterm born adolescent children reported increased family control and protective parenting (Burnett et al., 2017; Indredavik et al., 2005; Pyhala et al., 2011; Wightman et al., 2007). Mothers who saw themselves to have parented their EP/VP born adolescents successfully reported improved feelings of themselves (Saigal, Burrows, et al., 2000). Table 2-8 below summarises directions of impact of preterm birth on long-term parent outcomes by type of impact and assessment instrument.

2.5.4.1 Parent psychological health

Studies commonly reported no difference in parent psychological health outcomes between the groups of parents (Table 2-8). Yet diverse assessment instruments (n=13) and various group comparisons based on infant birthweight, neonatal medical risk level, family SES, or/and the level of families' social resources challenged comparability. Moore et al. (2006) found that parents of adolescents born with BW less than 750 grams and low SES (Hollingshead Four Factor Index) reported poorer mental health (BSI¹) scores in comparison with parents of FT born adolescents at the children's ages of 11 and 12 years, but not at the ages 13 and 14 years. Yaari et al. (2019) reported higher maternal distress scores (HADS²) among parents of VP born children in comparison with FT born children's parents, although the effect size was modest (mean difference 0.27, 95% CI = 0.03, 0.66, $p = 0.03$). Indredavik et al. (2005) reported generally no differences between the groups of parents (VLBW, SGA, FT) in perceived parent psychological problems (SCR-90-R)³, but found that VLBW fathers reported higher phobic anxiety scores in comparison with fathers of adolescents born FT.

¹ The Brief Symptom Inventory

² The Hospital Anxiety and Depression Scale

³ Symptom Checklist-90-Revised

VLBW parents also reported reduced emotional health (CHQ-PF50⁴) (parental emotional health: VLBW $M = 73.6$, $SD 22.4$ & FT $M = 90.1$, $SD 12.9$ FT, $p \leq 0.001$) (Indredavik et al., 2005). Over time, mothers of children born with VLBW at high neonatal medical risk level (a diagnosis of BPD; in the study defined as supplementary oxygen required for more than 28 days and radiographic evidence of chronic lung disease) reported reduced denial and mental disengagement coping (COPE⁵) scores in comparison with mothers of FT born children (Singer et al., 2010) (Table 2-8).

Parents of high medical risk preterm born children reported increased child-related parenting stress (PSI⁶) scores in comparison with parents of children born preterm at low risk (no BPD) or FT (HR-VLBW $M = 99.6$, $SD 19$ & LR-VLBW $M = 92.4$, $SD 27$ & FT $M = 94.2$, $SD 21$, $p < 0.003$) (Singer et al., 2007; 2010); as well as did those parents with ELBW children with severe morbidities (Zerach et al., 2015). Singer et al. (2007; 2010) found no difference in reported parental distress (PSI) among the groups of parents (HR-VLBW $M = 112.5$, $SD 22$ & LR-VLBW $M = 106.7$, $SD 27$, FT $M = 110.1$, $SD 23$, $p > 0.05$), but Taylor et al. (2001) reported lower parent perceived competence and poorer attachment scale scores (PSI) among parents of children born with BW <750 grams in comparison with parents of FT born children. As seen above, in both Singer et al. reports LR-VLBW parents reported lowest child-related and parental stress scores in comparison with the other two groups of parents, although the differences in the average stress scores were modest. Some evidence of decreasing parenting stress scores over time among parents of high neonatal medical risk VLBW children were reported, whereas parents of full-term born children reported increasing parenting stress scores over time (Singer et al., 2010) (Table 2-8).

⁴ Child Health Questionnaire Parent Form 50

⁵ The COPE Questionnaire

⁶ The Parenting Stress Index

Table 2-8 Direction of impact of EP/VP birth on long-term parent outcomes by type of impact and outcome instrument used in longitudinal birth cohort studies assessing long-term parent outcomes following VP birth

Type of impact	Negative impact	No difference between the groups	Positive impact	Evidence of change over time
Parent psychological & physical health				
The Brief Symptom Inventory	Moore et al. 2006: <750g BW low-SES vs. FT at 11 & 12 years	Singer et al. 2007: VLBW vs. FT ($F = 0.61, p < 0.55$) Moore et al. 2006: VLBW high-SES vs. FT at all ages Taylor et al. 2001: $M 48.17 \pm 7.94$ <750g BW ADV, $M 57.11 \pm 9.76$ <750g BW DIS, $M 50.78 \pm 9.63$ 750-1499g BW ADV, $M 53.33 \pm 7.58$ 750-1499g BW DIS, $M 50.46 \pm 10.08$ FT ADV, $M 50.79 \pm 11.89$ FT DIS		Moore et al. 2006: Statistically non-significant changes over time
The COPE Questionnaire			Singer et al. 2007: Denial score at 8yrs $M 17.7 \pm 3.85$ HR-VLBW vs. $M 19.55 \pm 5.00$ FT ($F = 5.3, p < 0.01$) Singer et al. 2010: Denial score at 14yrs HR-VLBW vs. FT ($p = 0.03$)	Singer et al. 2010: HR-VLBW maternal denial and mental disengagement coping reduced over time
The Center for Epidemiologic Studies Depression Scale		Drotar et al. 2006 & Wightman 2007: <16 score 23% ELBW vs. 20% NBW Saigal et al. 2010: $M 9.5 \pm 8.6$ ELBW vs. $M 9.6 \pm 8.0$ NBW		
Symptom Checklist-90-Revised	Indredavik et al. 2005: VLBW paternal phobic anxiety \uparrow vs. FT fathers ($p < 0.05$)	Indredavik et al. 2005: VLBW & SGA vs. FT mothers		
The Bradburn Affect Balance Scale		Saigal et al. 2010: $M 1.8 \pm 2.8$ ELBW vs. $M 1.7 \pm 2.4$ NBW		
The Spielberger State-Trait Anxiety Inventory		Saigal et al. 2010: $M 33.1 \pm 9.4$ ELBW vs. $M 33.0 \pm 9.0$ NBW		

Table 2-8 (continued)

Type of impact	Negative impact	No difference between the groups	Positive impact	Evidence of change over time
Parent psychological & physical health				
Perinatal Post-Traumatic Stress Disorder Questionnaire		Zerach et al. 2015: ELBW severe disability vs. ELBW mild/moderate disability & ELBW non-impaired		
The Hospital Anxiety and Depression Scale	Yaari 2019: VP vs. FT β 0.27, 95% CI 0.03, 0.66 ($p = 0.03$)			
Child Health Questionnaire Parent Form50	Indredavik et al. 2005: parental emotional impact M 73.6 \pm 22.4 VLBW vs. M 90.1 \pm 12.9 FT ($p \leq 0.001$); parental time impact M 92.3 \pm 12.2 VLBW vs. M 99.1 \pm 5.6 FT ($p \leq 0.001$)			
The Parenting Stress Index				
Child Domain	Singer et al. 2007: M 99.6 \pm 19 HR-VLBW, M 92.4 \pm 27 LR-VLBW, M 94.2 \pm 21 FT ($p < 0.003$) Singer et al. 2010: HR-VLBW vs. FT			Singer et al. 2010: LR-VLBW improving levels over time vs. HR-VLBW & FT
Parent Domain	Taylor et al. 2001: competence & attachment scales <750g BW vs. FT	Singer et al. 2007: M 112.5 \pm 22 HR-VLBW, M 106.7 \pm 27 LR-VLBW, M 110.1 \pm 23 FT ($p > 0.05$) Singer et al. 2010: VLBW vs. FT		
The Stress Index for Parents of Adolescents				Singer et al. 2010: At 14 years LR-VLBW improved levels vs. HR-VLBW & FT; at 14 years FT increasing stress over time
The Parenting Stress Index Short Form	Zerach et al. 2015: ELBW severe disability vs. ELBW mild/moderate disability & ELBW non-impaired			
The Health Survey Short Form 36		Saigal et al. 2010: ELBW non-NSI vs. ELBW NSI vs. NBW		

Table 2-8 (continued)

Type of impact	Negative impact	No difference between the groups	Positive impact	Evidence of change over time
Parent-child interaction / Parenting behaviour				
The Etch a Sketch	Jaekel 2012: VP/VLBW vs. FT maternal sensitivity ↓ & verbal control ↑			Jaekel 2012: VP/VLBW if child IQ ↓ then maternal sensitivity ↑ over time
Parental Bonding Instrument Short Form		Indredavik et al. 2005: VLBW mothers & fathers vs. SGA mothers & fathers vs. FT mothers & fathers		
The Parent Behavior Inventory		Pyhala et al. 2011: VLBW vs. FT maternal ($p = 0.67$) & paternal ($p = 0.52$) hostile parenting	Pyhala et al. 2011: Maternal supportive parenting VLBW vs. FT β 0.30, 95% CI 0.01, 0.59 ($p = 0.04$); paternal supportive parenting VLBW vs. FT β 0.34, 95% CI 0.02, 0.66 ($p = 0.04$)	
The Parent Protection Scale	Wightman et al. 2007: total parental protection M 31.0±6.9 ELBW vs. M 29.8±5.2 NBW ($p < 0.05$)			
Family impact				
The Impact on Family Scale	Drotar et al. 2006: total impact ELBW vs. NBW Singer et al. 2007: total impact HR-VLBW vs. LR-VLBW ($p < 0.02$); HR-VLBW vs. FT ($p < 0.02$) Taylor et al. 2001: <750g BW vs. FT ($p < 0.05$) Singer et al. 2010: total impact HR-VLBW low social support vs. LR-VLBW & FT		Singer et al. 2010: Mastery VLBW vs. FT	Singer et al. 2010: total impact VLBW & FT declined over time

Table 2-8 (continued)

Type of impact	Negative impact	No difference between the groups	Positive impact	Evidence of change over time
Family impact				
The McMaster Family Assessment Devise		Taylor et al. 2001: <750g BW & 750-1499g BW vs. FT Saigal et al. 2010: general functioning subscale <i>M</i> 20.0±5.4 ELBW vs. <i>M</i> 20.9±5.8 NBW	Saigal et al. 2010: general functioning subscale <i>M</i> 18.9±6.3 ELBW NSI vs. <i>M</i> 20.3±5.0 ELBW no-NSI (<i>p</i> = 0.006)	
The Family Burden Interview	Taylor et al. 2001: Any burden 61% <750g BW, 51% 750-1499g BW vs. 32% FT ($X^2=8.68$, <i>p</i> < 0.05) Moore et al. 2006: <750g BW low-resources vs. FT at 11 & 12yrs; <750g BW high-resources vs. FT at all ages; 750-1499g BW high-resources vs. FT at 14yrs			
Family Impact Questionnaire		Brown 2003: ELBW vs. FT		Brown 2003: ELBW social (<i>p</i> = 0.005) and financial (<i>p</i> = 0.012) impact reported in infancy diminished by adolescence
The Family Environment Scale	Burnett 2017: parent reported control EP/ELBW vs. NBW <i>β</i> 3.4, 95% CI 0.7,6.0 (<i>p</i> = 0.01)	Burnett 2017: parent reported cohesiveness, expressiveness, conflict, organisation EP/ELBW vs. NBW (<i>p</i> > 0.07)		
Family context / environment				
The Social Support Index		Saigal et al. 2010: <i>M</i> 51.8±8.1 ELBW vs. <i>M</i> 52.0±7.6 NBW		
The Family Inventory of Life Events and Changes			Singer et al. 2007: <i>M</i> 2.5±2.8 HR-VLBW, <i>M</i> 2.8±2.6 LR-VLBW & <i>M</i> 3.5±2.8 FT (<i>F</i> = 7.5, <i>p</i> < 0.02)	

Table 2-8 (continued)

Type of impact	Negative impact	No difference between the groups	Positive impact	Evidence of change over time
Family context / environment				
The Life Stressors and Social Resource Inventory-Adult Form		Drotar et al. 2006 & Wightman et al. 2007: ELBW vs. NBW		
The Multidimensional Scale of Perceived Social Support		Singer et al. 2010: VLBW vs. FT at all time-points		
Parents' relationship				
The Dyadic Adjustment Scale	Singer et al. 2007: Consensus scale VLBW vs. FT ($F = 5.26, p < 0.006$)			
The Ontario Child Health Study Questionnaire		Saigal et al. 2010: $M 7.2 \pm 2.6$ ELBW vs. $M 7.7 \pm 2.6$ NBW		
Parental divorce rate		Singer et al. 2007: 16% HR-VLBW, 7% LR-VLBW & 12% FT ($p < 0.33$)		
Change in original marital status		Saigal et al. 2010: 35% ELBW vs. 37% NBW		
Parent education				
Years of maternal educational attainment	Singer et al. 2007: at 8yrs $M 13.5$ VLBW vs. $M 14.2$ FT ($p = 0.01$) Singer et al. 2010: at 14yrs $M 13.82$ VLBW vs. $M 14.82$ FT ($p = 0.04$)			Singer et al. 2010: Lower and slower educational attainment among VLBW mothers after birth

NOTE: Absolute values are reported when data were available; if study reported $p > 0.05$ as statistically significant these were excluded from the table; excluded instruments ($n=3$): Impact on the Family: no summary measure (Saigal et al. (2000, 2010)); The General Health Questionnaire: reported at 2 years only (Yaari et al. (2019)); Life Events Questionnaire: no data reported (Zerach et al. (2015)

↑ higher; ↓ lower; ± Standard Deviation; M Mean; F F Statistic; θ Mean Difference; p Probability; CI Confidence Interval; X^2 Chi Square Test Value

ADV sociodemographic advantage & DIS sociodemographic disadvantage (Taylor et al. 2001); BW Birthweight; ELBW Extremely low birthweight; FT Full-term

HR high neonatal medical risk level = diagnosis of Bronchopulmonary dysplasia (BPD) & LW low neonatal medical risk level (no BPD) (Singer et al. 2007, 2010); NBW Normal birthweight; NSI Neurosensory impairment; SES Socio-economic status; SGA Small for gestational age; VLBW Very low birthweight; VP Very Preterm

2.5.4.2 Parent-child relationship and parenting behaviour

The direction of impact of EP/VP birth on the parent-child relationship or/and parenting behaviour varied. Parents of EP/VP children perceived themselves as more protective (PPS⁷) (total parental protection: ELBW $M = 31.0$, $SD 6.9$ & NBW $M = 29.8$, $SD 5.2$, $p < 0.05$ (Wightman et al., 2007) and supportive (PBI⁸) (maternal supportive parenting: mean difference 0.30, 95% $CI = 0.01, 0.59$, $p = 0.04$; paternal supportive parenting: mean difference 0.34, 95% $CI = 0.02, 0.66$, $p = 0.04$ (Pyhala et al., 2011). No group differences were found in parent recall of parent-child bonding (PBI-SF⁹) (Indredavik et al., 2005) or hostile parenting (PBI) (Pyhala et al., 2011). It may be worth noting that although these studies found statistically significant differences between the groups of parents, the effect sizes were small, and their clinical relevance remained uncertain. In a rated mother-child interaction session (Etch A Sketch), mothers of children born VP/VLBW were less sensitive and more verbally controlling in comparison with mothers of FT born children, but positively adjusted their parenting behaviour over time as per their child's cognitive ability (Jaekel et al., 2012) (Table 2-8). It is important to acknowledge, however, that a level of subjectivity in interpretation of parenting behaviour outcomes exists; for example, as discussed in the preceding chapter, high parental control or/and protectiveness may be regarded as both a positive as well as negative parental adaptation to the child's behavioural needs (Wightman et al., 2007).

2.5.4.3 Family context, family impact, and social outcomes

Parents with children born EP/VP perceived their family context, such as social resources and support (LISRES-A¹⁰; MSPSS¹¹; Social Support Index), similarly to the parents of full-term born adolescents (Drotar et al., 2006; Saigal et al., 2010; Singer et al., 2010; Wightman et al., 2007). Parents of preterm born children reported their families to have experienced fewer stressful life events or changes (FILE¹²) in comparison with parents of children born FT (HR-VLBW $M = 2.5$, $SD 2.8$ & LR-VLBW $M = 2.8$, $SD 2.6$ & FT $M = 3.5$, $SD 2.8$, $p < 0.02$) (Singer et al., 2007). Parents of preterm born adolescents reported negative family impact or burden, although the impact varied based on child birthweight and families' SES (Moore et al., 2006; Taylor et al., 2001) and perceived level of social support (Singer et al., 2010). Parents of EP and VP born children had fewer subsequent children following the preterm birth than parents of children born full-term (Alenius et al., 2018). All parents rated their families to generally

⁷ The Parent Protection Scale

⁸ The Parent Behavior Inventory

⁹ Parental Bonding Instrument Short Form

¹⁰ The Life Stressors and Social Resource Inventory-Adult Form

¹¹ The Multidimensional Scale of Perceived Social Support

¹² Family Inventory of Life Events and Changes

function well (Saigal et al., 2010; Taylor et al., 2001). Yet parents of ELBW young adults with neurosensory impairments perceived their families' functioning more favourably in comparison with parents of ELBW adolescents without impairments (Saigal et al., 2010). A declining trend in total family impact over time among all families was reported (Singer et al., 2010). Parents of adolescents born preterm reported comparable divorce rates with parents of FT born children (Saigal et al., 2010; Singer et al., 2007). Maternal educational attainment was slower and lower following preterm birth in comparison with mothers giving birth FT (Singer et al., 2007; Singer et al., 2010) (Table 2-8).

2.5.5 Factors influencing long-term parent outcomes

Predominant themes in the literature about long-term parent outcomes following EP/VP birth were the influence of birthweight and/or neonatal medical risk level, child disability, parental adaptation, child age and family resources on various parent and family outcomes. As the outcome measures used described merely aspects of parenting experience, they resulted in a mixture of recommendations for possible interventions or future research.

2.5.5.1 The influence of prematurity and neonatal medical risk level

The long-term parent outcomes following EP/VP birth were linked to child's level of prematurity (Alenius et al., 2018; Moore et al., 2006; Taylor et al., 2001). The level of prematurity and neonatal medical risk level are also associated; less mature infants have a higher risk of complications (Ancel et al., 2015; Costeloe et al., 2012; Moore et al., 2012).

In a study of long-term effects of preterm birth on families of children with varying extents of low birthweight and neonatal medical risk levels, more difficulties with child attachment (PSI¹³), less parenting competence (PSI) and higher rates of negative family impact (IOF¹⁴; FAD¹⁵) were reported among parents of 11-year-old adolescents born with BW <750 grams compared to parents of adolescents born with BW between 750 and 1499 grams and parents of full-term born adolescents. Between the latter two BW groups the reported parent outcomes were similar (Taylor et al., 2001).

Consistent with their previous findings, another longitudinal study of the same families at the children's ages of 11, 12, 13 and 14 years, found greater child-related family burden (FBI¹⁶)

¹³ Parenting Stress Index

¹⁴ Impact on Family Scale

¹⁵ McMaster Family Assessment Devise

¹⁶ Family Burden Interview

within the families with adolescents born <750 grams and low family resources (LISRES-A¹⁷) at the child ages of 11 and 12 years, and within families with high resources in the lowest BW group at all ages. As presented earlier, psychological distress (BSI¹⁸) scores also increased among the parents of adolescents with BW <750 grams and low SES compared to controls parents' scores at the children's ages of 11 and 12 years (Moore et al., 2006).

Taylor et al. (2001) examined the possibility that the associations between BW and/or neonatal medical risk level and long-term family outcomes were mediated by child's chronic conditions and/or neurodevelopmental morbidities. They found that the families with children with health concerns reported poorer long-term family outcomes, and when these families were excluded from the analyses, the association between BW and/or neonatal medical risk level and family outcomes diminished (Taylor et al., 2001). Moore et al. (2006) did not assess mediation in their study, but contemplated on a longer causal relationship between low birthweight and pre-pregnancy factors such as poor maternal health, long neonatal hospitalisation and a challenged parent-child relationship, and increased prevalence of child morbidities among the smallest infants, all resulting in a challenged parenting experience later (Moore et al., 2006).

Even though a clear advantage was their repeated measures of family impact over time, findings were evaluated with a non-validated instrument developed for the study (FBI). In addition to the risk of bias resulting from re-matching of control participants (Hack et al., 1994), the two reports were based on a relatively small sample of families: the index study reported outcomes for families of 60 children with BW <750 grams, 55 with BW between 750 and 1499 grams and 49 families of full-term born controls; the second study suffered attrition, which was more marked among low-SES families, and reported findings for caregivers of 44 children born <750 grams, 42 children born between 750 and 1499 grams and 47 children born full-term. Small group sizes may have prevented the authors from detecting more subtle differences between the groups, such as between the middle birthweight group and the full-term born controls (Moore et al., 2006; Taylor et al., 2001).

A study assessing the impact of ELBW on general health outcomes, family stressors and family impact among parents of 8-year-old ELBW (n=219) and NBW (n=176) children concluded that lower BW or GA did not predict higher family impact (IOF). Actually, BW, GA, and neonatal risk factors were not related to scores on the IOF questionnaire, but negative family impact was associated with child's neurodevelopmental disability, in fact the functional impact of child's chronic condition/s were the most predictive of higher negative family

¹⁷ Life Stressors and Social Resource Inventory-Adult Form

¹⁸ Brief Symptom Inventory

impact (Drotar et al., 2006). This finding supports both Taylor et al. (2001) and Moore et al. (2006) contentions that the impacts of lower BW and higher neonatal medical risk level are mostly experienced through their relation to increased prevalence of child disability in these families (Moore et al., 2006; Taylor et al., 2001).

Following the framework of influences on parental experiences after preterm birth (Miles & Holditch-Davis, 1997), Taylor et al. (2001) commented further that the experiences of difficult birth and neonatal intensive care may have a long-lasting impact on the family, even though the initial parent stress seems less during the early years (Singer et al., 2007; 2010). They speculated that initial distress may compromise the parent-child relationship, which together with challenged child behaviours bidirectionally worsens the outcomes among the parents and the child (Taylor et al., 2001). This hypothesis of long-lasting impact of birth experience was not supported by Drotar et al. (2006). In their predictive model which included composite variables for SES/parent risk, birth risk, neonatal risk, child's neurodevelopmental outcomes, and functional impact of chronic conditions on the family, rather than the neonatal variables, the SES/parent risk together with child neurodevelopmental disability were more predictive of the long-term parent outcomes. Participants in their study were families with children born with lowest birthweights, at potentially higher risk of complications, when the neonatal experiences could also be assumed to have been challenging. Although, Drotar et al. (2006) final predictive model only accounted for 17% of the total variance in family impact, indicating that other factors not assessed in their study also affected the family impact (Drotar et al., 2006). Additionally, it may be that the instruments used by Drotar et al. (2006), somewhat different to Taylor et al. (2001) (see Table 2-7) did not capture the particular impact of the birth experience, which may relate more to parenting behaviour than to the overall family impact.

Another longitudinal study assessed parenting stress (PSI) among mothers of VLBW children with a diagnosis of BPD (n=113), VLBW children without a diagnosis of BPD (n=80) and full-term born controls (identified at birth) (n=122) at seven timepoints between one month and 14 years of age using the same measurement tools (IOF, PSI, MSPSS¹⁹, COPE²⁰). The VLBW families were recruited when their infants were treated in level III neonatal intensive care units of hospitals in Cleveland, Ohio between 1989 and 1992 (same geographical area as Taylor et al. (2001)). The mothers of VLBW children with BPD reported the highest levels of child-related parenting stress, whereas the stress levels were similar among the mothers with VLBW children with no BPD and the mothers of full-term children until eight years of age.

¹⁹ Multidimensional Scale of Perceived Social Support

²⁰ COPE Questionnaire

At school-age, the stress levels among the mothers of full-term born controls increased bringing the group closer to the stress levels of mothers in the BPD group, whereas the child-related stress among parents in the no-BPD group decreased over time. Children with BPD had more disabilities, chronic health problems and lower IQs than the children in the other two groups. Overall, parenting stress in the child and parental distress sub-scales were most consistent over time among the mothers of VLBW children with BPD (Singer et al., 2010).

Zerach et al. (2015) measured parenting stress (PSI-SF²¹) among mothers of ELBW children aged four to 16 years. They found the highest levels of overall parenting stress among mothers of ELBW children with severe developmental difficulties and lowest levels of stress in mothers of ELBW children with normal development (Zerach et al., 2015). Although Zerach et al. (2015) utilised a cross-sectional study design among children from a wide range of ages with no control comparison, their findings supported Singer et al. (2010) in regards to the association between child disability and parenting stress (Singer et al., 2010; Zerach et al., 2015).

2.5.5.2 *The influence of child disability*

The level of prematurity and neonatal medical risk have a direct link to child's chronic conditions and neurodevelopmental outcomes; lower gestation or birthweight and higher neonatal medical risk level increase the risk of morbidities (Ancel et al., 2015; Moore et al., 2012; Pierrat et al., 2017; Serenius et al., 2013).

Drotar et al. (2006) assessed the “global impact of paediatric disability on the family” (Drotar et al., 2006, p. 2009) using the IOF, and found that the child's neurodevelopmental impairments (definition included a diagnosis of cerebral palsy, ‘*psychometric testing*’ and parent report on VABS²²) and the functional impact of child's health on the family (QuICCC²³) had the greatest impact on the family outcomes. Other family stressors (LISRES-A) did not explain the differences in the outcomes between the families with children born EP and the control families, but increased maternal depression (CES-D²⁴) scores were associated with higher family impact. Drotar et al. (2006) concluded that the impact of child disability on families was overarching rather than segmented in particular areas of family life. They noted that the differing aspect of prematurity is its simultaneous relation to several physical, psychological and cognitive health concerns such as cerebral palsy and behavioural disorders

²¹ Parenting Stress Index Short Form

²² Vineland Adaptive Behavior Scale

²³ Questionnaire for Identifying Children with Chronic Conditions

²⁴ Center for Epidemiologic Studies Depression Scale

that impact the families on multiple levels such as family resources, parental time and the family's internal relationships (Drotar et al., 2006). Yet Moore et al. (2006) found evidence that among preterm born adolescents even more subtle developmental difficulties affected parents' experiences. This was demonstrated by the inclusion of disability factor (diagnosis of neurosensory impairments and/or child IQ <70) in their analytic model which did not alter the group effects between the <750 grams, 750-1499 grams and the full-term born children (Moore et al., 2006).

In a cross-sectional analysis of a longitudinal birth cohort study from Hamilton, Ontario, Canada, Saigal, Burrows, et al. (2000) evaluated family impact of EP birth on the parents of 12 to 16 years old adolescents with and without neuro-sensory impairments (NSI), defined as one or more of the following diagnoses: cerebral palsy; hydrocephalus; significant cognitive impairments; autism; unilateral blindness; bilateral blindness and sensorineural deafness. Overall, the study found negative family impacts. However, these were reduced after exclusion of participants with NSIs, showing no difference in parent outcomes between the parents of EP and the full-term born adolescents. Although utilising a non-validated outcome measure, the study assessed overall impact on work, marital and family life. The parents with adolescents with NSIs perceived that their emotional health had suffered due to their child's health, but also that the experience of caring for an EP child had improved their feelings of themselves (Saigal, Burrows, et al., 2000).

In addition to clinically defined child disability, also parental perceptions of their child's abilities influenced the outcomes. This could be related to Deater-Deckard's (1998) parenting stress model of parental appraisal of appropriateness of child behaviour, as presented in the introductory chapter of this thesis. A study from Trondheim, Norway assessed the influence of VLBW on quality of life, parent-child relationship and parent mental health (PBI-SF²⁵, SCL-90-R²⁶, CHQ-PF50²⁷) at the child's age of 14 years (Indredavik et al., 2005). VLBW parents reported more behavioural and psychosocial problems in their child compared to control parents, even though both groups of adolescents rated their own quality of life similarly. The adolescents' perceived quality of life was not altered by their level of IQ (Indredavik et al., 2005). In line with Saigal, Burrows, et al. (2000) parental concern about their child's health impacted their own emotional health and restricted the time they had for themselves. The negative emotional impact was further strengthened among both mothers and fathers if the VP born adolescent had a diagnosis of cerebral palsy or psychiatric disorder, but

²⁵ Parental Bonding Instrument Short Form

²⁶ Symptom Checklist-90-Revised

²⁷ Child Health Questionnaire Parent Form 50

did not impact parent mental health (SCL-90-R) or relational warmth in the family (PBI-SF), as all parents reported similar levels for these outcomes (Indredavik et al., 2005).

The authors noted a potential information bias; parents of preterm born adolescents may have been more aware of risks relating to prematurity and therefore were more likely to report health concerns compared to term-born controls. However, the parents did perceive that the worry impacted their personal health, which should be regarded as an outcome in itself. Parental concern about their child's quality of life may have altered parenting behaviour, as VLBW adolescents reported their families to be more protective than those of controls (Indredavik et al., 2005).

In contrast, Yaari et al. (2019) found that mothers of VP born children from Melbourne, Australia reported higher levels of psychological distress at all ages (assessed at three timepoints between two and 13 years) compared to mothers of FT born children (GHQ²⁸, HADS²⁹). However, instead of child neurodevelopmental impairment, maternal and family factors, such as social risk level, were more strongly associated with increased maternal psychological distress scores. Their findings signal the importance of acknowledging the influence that prenatal maternal health and preceding family factors may have on long-term outcomes after the EP/VP birth; a factor that few studies were able to consider.

2.5.5.3 The influence of parental adaptation

Child disability alters parental experiences, both by modifying the experience of how the child is to parent and perception of success of the task of parenting.

Singer et al. (2010) found that symptoms of depression and anxiety found at birth and at two years among mothers with VLBW children with BPD had decreased by eight years and the mothers reported similar levels of parental distress (BSI³⁰) as mothers of VLBW children without BPD and mothers of full-term born children (Singer et al., 2007; 2010). No differences in maternal coping mechanisms were found in previous assessments, but at eight years the mothers with VLBW BPD children reported less denial and disengagement in coping than the mothers in the other two groups. This was interpreted by the authors as a positive adaptation in parenting behaviour. This demonstrated that despite the early mentally burdening parenting experience, the mothers were able to adapt successfully into their parenting roles (Singer et

²⁸ General Health Questionnaire

²⁹ Hospital Anxiety and Depression Scale

³⁰ Brief Symptom Inventory

al., 2007; 2010). The parents of VLBW BPD adolescents reported higher satisfaction with their parenting and feelings of mastery (IOF) than the parents of full-term born children at 14 years. This finding is in line with Saigal, Burrows, et al. (2000) who reported that parents of adolescents with NSIs experienced improved feelings of themselves and their parenting task.

Singer et al. (2007; 2010) discussed the possibility that the decreasing parental distress with child age and the adaptations in the parenting behaviour among the mothers with children with BPD may have resulted from reduced adolescent-parent conflict due to differing age-related behaviour among VLBW BPD, VLBW with no BPD and/or full-term born adolescents. Among VLBW BPD adolescents, functional limitations and the resulting reduced activities outside the home may have created an environment where parental monitoring and child compliance were increased, and therefore some of the typical child-parent relationship stressors were not present or appeared later (Singer et al., 2007; 2010). Even though there were low levels of parent-child conflict, the VLBW parents reported more marital disagreement at the child's age of eight years than controls (Singer et al., 2007).

Taylor et al. (2001) emphasised that potential reporting bias may reduce the validity of findings like above: the parents of VLBW children with disabilities may either adapt their parental ambitions as more 'appropriate' for their child's abilities and/or do not perceive their child's functional limitations as problems, and therefore be less likely to report negative family impact (Taylor et al., 2001). In the qualitative study of positive family outcomes among parents of EP children aged seven to 10 years, Lou et al. (2009) found that whereas clinicians assessed the child's performance in a certain task at a given timepoint, the parents understood their child's performance as a continuous dynamic process built on past concerns and progress. They also reported positive adaptations in the families where from a clinical perspective the child had functional limitations, but from a social perspective the family had adjusted successfully; a child with chronic lung disease was unable to play football, but performed well as a goal keeper (Lou et al., 2009, p. 572).

Jaekel et al. (2012) assessed the differences in the quality of mother-child interaction between VLBW and full-term born mother-child dyads at two timepoints of six and eight years, and therefore, directly addressed the hypothesis of a differing mother-child interaction among the VLBW families. The VLBW children were less persistent and socially active in their behaviours. Their mothers were less sensitive and more verbally controlling compared to the mothers of full-term born children. This association was mostly accounted for by the child's level of cognition. Increased maternal sensitivity at the first assessment promoted increased child persistence during the second assessment, and this positive association increased with a

decreasing child cognition, demonstrating that the mothers positively adapted and adjusted their parenting behaviour according to their children's needs (Jaekel et al., 2012).

Wightman et al. (2007) assessed parental protectiveness at the children's age of eight years and concluded that higher total Parental Protection Scale (PPS) scores among the parents of ELBW children were explained by the presence of child NSI (defined as diagnoses of cerebral palsy and/or bilateral blindness or deafness corrected with hearing aids). The authors postulated that parental overprotection among children with disabilities might represent a beneficial support adaptation. The authors also noted that in contrast to studies in other clinical groups, overprotective ELBW parents did not report increased levels of child behavioural disorders such as hyperactivity or attention-deficits among their children compared to the families with less protective parents. The authors did note that they failed to assess child psychopathology, a factor that was together with cerebral palsy most influential in negative parent outcomes in the Indredavik et al. (2005) report presented earlier. Similar to Indredavik et al. (2005), the prevalence of maternal depressive symptoms (CES-D) did not differ between the mothers of ELBW and NBW children (Wightman et al., 2007).

In a study of 15 to 20-year-old EP/ELBW adolescents and their parents from Victoria, Australia, both reported more organisation and control in their families and less family conflict (FES³¹) compared to their full-term born control adolescents and their parents (Burnett et al., 2017). Families with EP/ELBW adolescents also had a higher parent-adolescent agreement level between the FES scores compared to the families of control adolescents. Both adolescent and parental views about protective and controlled family environment did not change when participants with low IQs were excluded from the analysis. The original study sample had suffered attrition where non-participants had lower IQs and higher rates of childhood NSIs. Additionally, the ELBW adolescents taking part were more often from single-child families or were less likely to be living with both birth parents than the control adolescents. Having more controlling parents did not affect mental health outcomes of EP/ELBW children differently to the outcomes of full-term born adolescents (Burnett et al., 2017).

Pyhala et al. (2011) examined how VLBW survivors and their parents recalled their parenting behaviour 22 years after the birth in the Helsinki Study of Very Low-Birth-Weight Adults. The parents of VLBW young adults recalled their parenting behaviour to have been more supportive compared to the parents of FT born young adults, whereas the VLBW young adults remembered their parents as more protective, especially the young women. In their discussion,

³¹ Family Environment Scale

the authors suggested a link between overprotective parenting behaviour and decreased adolescent mental health (Pyhala et al., 2011). However, since they failed to examine mental health outcomes among either the parents or their children, the significance of finding a slightly differing recall of parenting behaviour among the groups of parents over a 16-year period remained unclear.

2.5.5.4 The influence of family resources

As presented earlier, Drotar et al. (2006) found that the primary caregivers of 8-year-old ELBW children reported higher total negative family impact (IOF) than the parents of NBW comparator children. The authors concluded that the excess negative family impact was not attributable to family resources (LISRES-A), which did not differ between the study groups (although this was borderline ($p = 0.052$)). Further, the study used a composite variable to account for family disadvantage (the variable included SES as Black ethnicity; less than high school education; living in an area with >30% of families living below the poverty level and parent risk as maternal depression (defined as a total CES-D score of ≥ 16)). The SES/parent risk composite variable predicted higher negative family impact, but without detailing which of the included factors were most influential (Drotar et al., 2006).

In contrast, Moore et al. (2006) stated that family stressors (LISRES-A) were associated with increased family burden (FBI) that varied by the level of resources, BW group and child age. Among low resource families, parents of adolescents born with BW <750 grams reported higher family burden than control families at the ages of 11 and 12 years. Among the high resource families, parents in the lowest BW group reported higher family burden at all ages, and the families of adolescents born between 750 and 1499 grams reported higher burden than controls at the age of 14 years. The authors interpreted this to indicate late emergence of burden in the high resource middle BW group families, and that among the lowest BW families with high resources the burden was more persistent than among the low resource families (Moore et al., 2006).

This study additionally investigated the effects of SES (Hollingshead Four Factor Index) on parental distress (BSI) and found significant group differences only among low SES families in the lowest BW group compared to the families of full-term born adolescents at the ages of 11 and 12 years (Moore et al., 2006). This finding differed from Indredavik et al. (2005) conclusion that the relationship between low BW and parental distress (SCL-90-R) at 14 years was further strengthened if the family was from a high socio-economic position (Hollingshead

Two Factor Index). Although the different outcome instruments utilised in these studies make direct comparisons difficult.

Taylor et al. (2001) found that the association between a BW <750 grams and increased parental distress (BSI), higher parenting stress scores (PSI Competence and Attachment scales) and increased negative family impact (IOF) was stronger among disadvantaged families compared to advantaged families (defined as Social Disadvantaged Index >1; included education, marital and minority statuses) (Taylor et al., 2001). Although Moore et al. (2006) study was conducted among the same families, they did not utilise the IOF in the same way and therefore direct comparisons are not possible between the two studies. In contrast, an association between lower SES and higher PSI scores was found by both Singer et al. (2007) and Zerach et al. (2015).

Singer et al. (2007) concluded that lower maternal education attainment was associated with negative parent coping mechanisms (COPE) (Singer et al., 2007; 2010). Similarly, in Wightman et al. (2007) report higher parental protectiveness (PPS) was associated with education lower than high school level, minority ethnic background, single parenthood and poverty. Both studies reported findings for hospital-based cohorts from Cleveland, Ohio. In contrast, Jaekel et al. (2012) stated that in their study based on a whole population sample of VLBW/VP births in Bavaria, Germany, higher parental control or less sensitive parental behaviour were not associated with higher family adversity (a composite variable including education, mother's age, single parenthood, family size, overcrowding, foster care, no emergency contacts and parent mental health; measured at birth), but was rather related to the child's cognitive functioning (Jaekel et al., 2012).

Overall, EP/VP birth hindered the financial and social progress commonly experienced in families with NBW children (Brown et al., 2003), especially among mothers (Singer et al., 2007; 2010). Singer et al (2007, 2010) found that further educational attainment after childbirth was reduced among the mothers of VLBW children compared to the mothers of full-term born children, regardless of their educational level achieved before the delivery, maternal IQ, and the medical risk status of the infant. The mothers of VLBW children also reported reduced job opportunities and income (Singer et al., 2007; 2010). Saigal et al. (2010) supported this finding by stating that over 20 years after giving birth extremely prematurely, the mothers reported negative impacts on their work opportunities, regardless of their child's disability status (Saigal et al., 2010). Consequently, the levels of family resources commonly measured at adolescence were not independent factors of EP/VP birth.

2.5.5.5 *The influence of child age*

According to Singer et al. (2010), total child-related family impact (IOF), regardless of parents' perceived level of social support or child's prematurity status, decreased over time among all families. The authors found that after the initial stress of having an EP/VP born child, parenting stress levels remained relatively constant until the age of three. Towards school-age, differences in parenting stress levels narrowed between the groups of parents (Singer et al., 2010).

Also, Moore et al. (2006) reported decreased family burden over time among all families, except among those high-resource families with adolescents born between 750 and 1499 grams. Moore et al. (2006) interpreted this to indicate different levels of family conflict, and parent and adolescent responses to conflict in high and low resource families. When children grow, life tasks become more demanding. The families with adolescents born <750 grams may have experienced challenges throughout their child's life due to higher levels of child disability, and therefore may not have perceived the transition to adolescence as stressful. Similarly, the adolescents born full-term may have tackled the new tasks more easily and therefore not burden the family, whereas the new developmental demands may have seemed more unexpected to the middle-BW group families where adolescents may have had more subtle developmental or social difficulties (Moore et al., 2006).

Saigal et al. (2010) concluded that by the time the participants in their cohort had reached the age of 22 to 25 years no differences in reported levels of family dysfunction (Impact on the Family) among the parents of EP and full-term born young adults were found. Also, different to the assessment at adolescence, at this timepoint inclusion of the participants with NSIs in the analyses did not alter the results. The authors hypothesised that these findings demonstrated that the parents had had more time to reflect on the family impact, but also that they may have felt a sense of completion as their EP child had successfully reached adulthood and therefore the impact of EP birth on the parents and the family had ceased (Saigal et al., 2010).

2.6 Discussion

When synthesising findings from the 17 studies, I found that long-term parent outcomes following EP/VP birth differed from those of control parents of full-term born YA. This association was largely accounted for by the presence of child disability, and altered by child age, family resources, and parental adaptations among the families with adolescents born EP/VP. The reviewed studies had found increased child-related family burden (Moore et al.,

2006; Taylor et al., 2001), negative family impact (Drotar et al., 2006; Taylor et al., 2001) and parenting stress (Singer et al., 2007; 2010; Zerach et al., 2015). The studies also found that parents of preterm born children had fewer subsequent children following the preterm birth (Alenius et al., 2018). Parents experienced long-term financial implications such as limited job opportunities and loss of family income (Saigal, Burrows, et al., 2000; Saigal et al., 2010; Singer et al., 2007; 2010) and reduced educational attainment after preterm birth (Singer et al., 2007; 2010). Positive family transformations were reported (Lou et al., 2009; Saigal, Burrows, et al., 2000; Saigal et al., 2010). Further attention should be given to the interpretations and limitations of these findings.

2.6.1 Summary of long-term parent outcomes

Altogether, this review found that the identified publications reported a trend towards increased family impact among the families with YA born preterm, particularly among those families with children with long-term morbidities. EP/VP birth seemed to have a negative impact on family life as a whole rather than on a particular segment of life (Drotar et al., 2006). However, it is important to note that, with one exception (Burnett et al., 2017), the findings of increased child-related family burden and impact (Drotar et al., 2006; Moore et al., 2006; Taylor et al., 2001), as well as parenting stress (Singer et al., 2007; 2010) were based on cohorts of families from Cleveland, Ohio, together with one cross-sectional hospital-based report from Israel (Zerach et al., 2015). As discussed earlier, many of these studies had considerable risk of bias.

In this review parent psychological health outcomes were assessed in several publications. Those from Ohio, US, Ontario, Canada and Trondheim, Norway showed similar findings of no differences in mental health outcomes between the groups of parents. Yet diverse assessment instruments and various group comparisons based on infant birth status, neonatal medical risk level, and/or family resources challenged the comparability. In contrast to the other publications, Yaari et al. (2019) from Melbourne, Australia, a more recent birth cohort from the early 2000s, showed increased psychological distress scores among VP parents that was consistent over time. Although, the average group scores were within the normal range of anxiety and depressive symptoms among both groups of parents. Instead of child neurodevelopmental impairment, maternal and family factors, such as social risk level, were more strongly associated with maternal psychological distress in their study (Yaari et al., 2019). Yet, this symptomology could have pre-existed the VP birth.

Despite generally comparable mental health outcomes, in this review, I found that parents of EP/VP children reported decreased emotional health and perceived that their concerns about their child's health impacted their personal health (Indredavik et al., 2005; Saigal, Burrows, et al., 2000). As discussed in the introduction to this chapter, parents of preterm born children may view their children's quality of life more critically in comparison with the view of adolescents themselves (Indredavik et al., 2005; Wolke, Chernova, et al., 2013). The findings from this review suggested that parental concerns of their adolescent children's quality of life may have impacted their parenting behaviour. VLBW adolescents reported their families to be more protective than their peers (Indredavik et al., 2005). Parents of EP/VP adolescents perceived their parenting behaviour to have been more supportive (Pyhala et al., 2011), more protective (Indredavik et al., 2005), but also more controlling (Burnett et al., 2017) than parents of children born FT. Yet high attrition in the study samples and small effect sizes must be considered. Nevertheless, these differences in parenting behaviour might have indicated beneficial adaptations. Over time, parents of preterm born children reported reduced denial and mental disengagement coping in comparison with mothers of FT born children (Singer et al., 2010). Also, the only study not relying on parent self-report, reported positive behavioural adaptations among parents based on their children's cognitive ability (Jaekel et al., 2012). The presence of child disability impacted parenting behaviour demonstrating that in parental experiences, child ability, parent perception of child ability, as well as perceptions of successfulness of one's own parenting behaviour may be interlinked. Yet the included studies less commonly distinguished between varying adolescent morbidity, such as cerebral palsy or/and behavioural and psychiatric disorders. These factors may have been present simultaneously (D'Onofrio et al., 2013; Drotar et al., 2006). Adolescent behavioural challenges, rather than chronic health concerns, have been shown to be associated with increased parenting stress among other at-risk populations (Barroso et al., 2018; Majnemer et al., 2007). Future research among families with adolescents born preterm could further explore potential associations between parent outcomes and varying adolescent morbidities.

Findings from the reviewed studies suggested that the family environment, in terms of social and financial resources, was associated with parental coping styles and/or adaptation over time. Among all parents, lower parent educational level was associated with negative coping styles (Singer et al., 2007; 2010). Families with higher social risk, such as single parents and/or parents from black ethnic minority groups, reported more protective parenting behaviour (Wightman et al., 2007). This may relate to contextual factors such as neighbourhood safety (Kotchick & Forehand, 2002). On the contrary, Jaekel et al. (2012) found that families' social risk factors were not associated with parenting behaviour, rather that child disability was a more predictive factor. Jaekel et al. (2012) measured their family risk factors, such as maternal

education and housing conditions, at birth. Yet families in their study lived in a setting where low-cost healthcare services were available to all. As findings from this review have demonstrated, EP/VP birth may impact parents' educational attainment and income prospects after the birth, and thus, the level of family resources commonly measured at adolescence are not independent factors of EP/VP birth.

2.6.2 Construction of parental experiences

In this review I found that, since the outcome measures utilised to assess long-term parent outcomes following EP/VP birth largely relied on self-report, they described aspects of parenting experience; how the child was perceived and how the parents' perceived their own parenting behaviour at the time of the assessment. Changes in experiences resulted in variations in the measures of long-term parent outcomes, which only partially captured the experience, as other factors were associated with parenting behaviour. These factors, such as family resources or child disability, either promoted or hindered the parents' perception of successfulness. These interlinkages are visualised in Figure 2-3 below.

At first glance, birthweight and/or neonatal medical risk level seemed influential to long-term parent outcomes following EP/VP birth. On a closer examination, the associated child disability was more predictive of outcomes in the families with EP/VP children long term. However, although clinically defined child disability influenced long-term parent outcomes, parental perceptions of their child's ability impacted impressions; how the child was to parent and how successful the parent perceived their parenting behaviour to be. These perceptions resulted in changes in parents' behaviour. Family resources, as social and financial means, provided a context for the family that either hindered or promoted parental experiences. As the child grew, parent outcomes fluctuated as the experiences changed; by adulthood both positive and negative perceptions of family impact, burden, or parenting stress had largely diminished, perhaps as the active parenting task had ended. Those long-term parent outcomes that were irreversible, such as the impact on level of education or loss of job opportunities, remained. Whilst these outcome measures captured what were believed to be the issues facing parents, investigations from the parents' perspectives were largely missing. With one exception, no qualitative studies were identified that conveyed the perspectives of parents and situated these in their current lived experiences. These are urgently needed to gain a deeper understanding of how having a preterm born child affects parents.

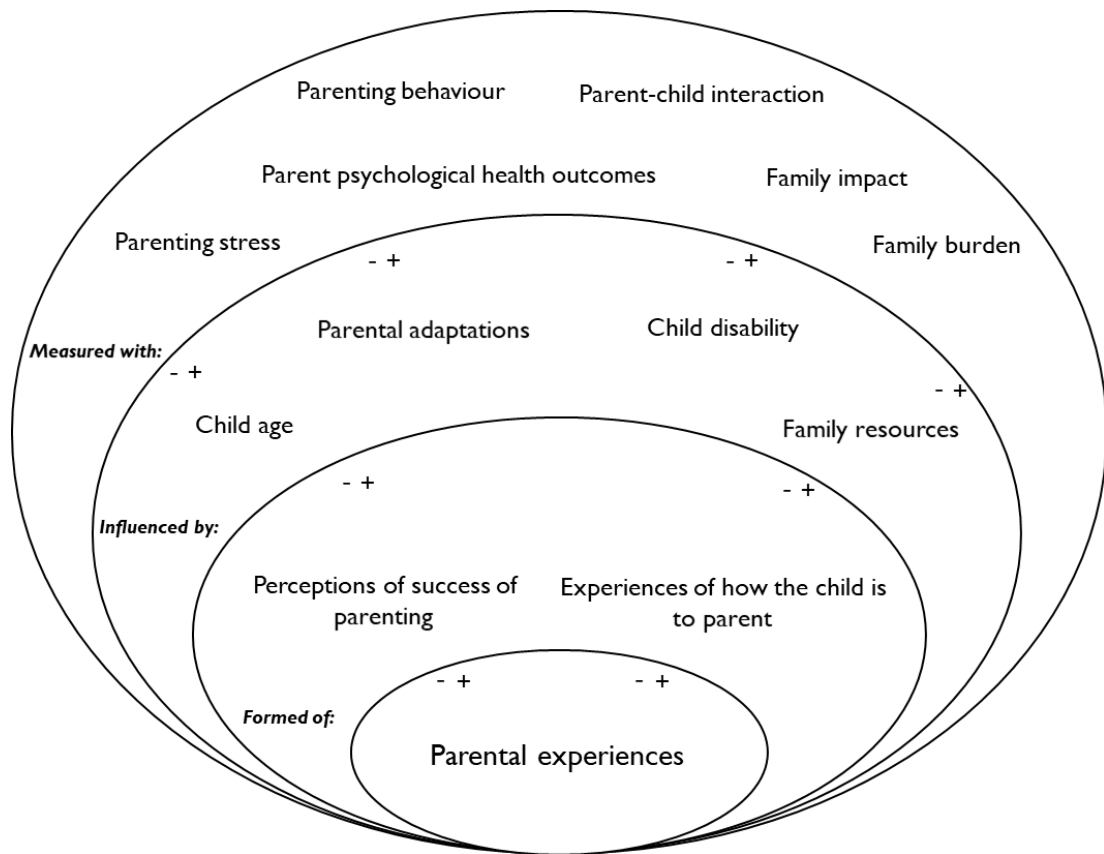


Figure 2-3 Construction and influences in the long-term parental experiences following EP/VP birth

2.6.3 Limitations in current research

This review has demonstrated that current research on long-term parent outcomes following EP/VP birth has limitations. Methodological disparities between the studies challenged my ability to synthesise findings. The studies utilised varying outcome measures with minimal overlap between the birth cohorts, study findings were described for children from wide age ranges, and on occasions without utilising comparison groups. A similar difficulty in the synthesis of long-term family outcomes has been reported in a review of family impact of preterm birth on families with mostly pre-school-aged children (Treyvaud, 2014). Additionally, generalising findings from the longitudinal cohort studies is challenging as the birth cohorts were relatively small. Furthermore, several of them recruited infants from small geographically restricted areas, and the studies were from varying societal backgrounds; for example, the potentially differing impact of Germany's, Canada's, Finland's and Norway's universal low-cost healthcare system on family resources compared to the birth cohorts from the US should not be disregarded (Saigal, Burrows, et al., 2000). Families from the US have reported having to make large financial sacrifices to care for their VP born children (Kantrowitz-Gordon et al., 2016). The children in this review were born between 1972 and

2003 and the experiences of EP/VP birth and the neonatal intensive care, and therefore potentially the long-term parent outcomes, might differ between the families in the older and more recent birth cohorts. For example, an increased focus on family-centred care in the NICU context has been a more recent phenomenon (Shaw et al., 2019).

As a methodological limitation of this review, the literature search could have been further limited with cohort age and/or type of impact to ensure the comparability of included studies. However, this might have compromised the review's aim to improve understanding of parent outcomes more widely. Also, despite a wide termed search and generous inclusion criteria, the final sample included only 17 studies and eight settings, suggesting that information on the effect of EP/VP birth on long-term parent outcomes is scarce. Future research could focus on particular parent outcomes such as mental health outcomes and examine findings in detail over time. Previous research has noted a challenge that studies have commonly examined various parental psychological health outcomes (Misund et al., 2013) often utilising screening tools instead of clinical diagnostic instruments (Treyvaud, 2014).

This review has showed that research among prematurity has mainly focused on examining factors contributing towards negative family outcomes. This may be because preterm birth has been understood as a risk exposure whose effects (i.e., negative family outcomes) are to be prevented/mitigated among the future generations. Consequently, in the literature, the focus of the research has been on challenged outcomes and little attention has been paid to factors that promote robustness in family outcomes. The studies included in this review predominantly concluded that among families with children with disabilities parent outcomes were worse than among families with healthy EP/VP children or full-term born children's families. Among the latter two groups the outcomes were generally similar. However, several studies used instruments, such as the Impact on Family Scale, particularly designed to assess the impact of child's chronic condition/s on the family, and therefore the finding of a dominant impact of child disability can be largely expected. As mentioned earlier, studies may have not examined the impact of the varying type of child disability. The findings of no differences in parent outcomes between the families of EP/VP born and the full-term born adolescents' families were often attributed to either reporting or recall biases. Lou et al. (2009) noted in their discussion that the research on long-term parent outcomes following EP/VP birth has mainly hypothesised on negative family impacts, overlooking potential positive family transformations. More recently, the general research on long-term effects of preterm birth has given more attention to better understanding factors such as resilience (Janvier et al., 2016). Schappin et al. (2013) have pointed out that despite the often-hypothesised life changing

impact of preterm birth, some studies assessing stress levels among parents of preterm born children have not found the expected poor parent outcomes.

Associated research among preterm born populations has presented long-term consequences. A qualitative study of psychological and social responses of parents of younger VP born children (≤ 7 years) found that the parents' fear of their child's physical and cognitive development lasted for years after the birth. The parents went to extreme measures to promote their child's optimal development; some described a decline in their personal health due their child's limitations as a 'natural' cause of parenthood. The parents also reported weakened relationships with extended family members and friends as they felt that others were unable to fully comprehend the unique experience of parenting a preterm born child. The study did not detail whether these findings were associated with child disability (Kantrowitz-Gordon et al., 2016).

In this review, I found that understanding long causal pathways had further challenged the research on long-term parent outcomes. Some studies in this review noted that prenatal factors, such as maternal health, may have been more influential among families with smaller infants, those with more morbidities. Therefore, due to these prenatal factors, rather than those in the child's adolescence, the long-term parent outcomes may have differed from the outcomes of families with EP/VP born children of higher BW or the outcomes of full-term children's families. Yet, as the risk of bias assessment demonstrated, many studies did not have data on parental mental health outcomes before or during pregnancy, or at the time of birth. As discussed in the introductory chapter of this thesis, maternal mental illness prior to and during pregnancy may also impact symptomology postpartum (Kee et al., 2021; Misund et al., 2013). Parental adjustment to preterm birth may have been different between the parents from varying socio-economic backgrounds as a result of available resources and/or other simultaneous family stressors (Jaekel et al., 2012; Voss, Jungmann, Wachtendorf, & Neubauer, 2012). However, the findings from this review have showed that the lack of data on pre-pregnancy parental factors is a general limitation in research on long-term parent outcomes following EP/VP birth.

2.6.4 Long-lasting impact of adverse birth experience

In this review, I found that against the hypothesis of a prolonged impact of adverse birth experience, the studies, on occasions, reported similar long-term parent outcomes among parents of EP/VP adolescents and the families with full-term born adolescents; in the Moore et al. (2006) study (same cohort of families as in Taylor et al. (2001)) 29% of the parents with

children born with BW <750 grams reported no negative family burden (FBI) or parental distress (BSI) at the child's age of 12 years. Alenius et al. (2018) concluded that the finding that 63% of mothers of EP children did not have further children (previous interbirth interval length considered) was not fully accountable to high-risk pregnancy factors, maternal severe clinical outcomes, or infant disease, suggesting that factors different from the adverse birth experience or infant morbidity altered the parents' reproductive decisions following EP/VP birth. Yet, as discussed in the introductory chapter of this thesis, during the early experiences, severity of infant disease may not directly correlate with the level of parental distress (Brummelte et al., 2011; Caporali et al., 2020). Later, parents of toddlers (at 24 months of age) with higher neonatal medical risk may report lower levels of parenting stress in comparison with families with preterm born infants with lower medical risk (Gerstein & Poehlmann-Tynan, 2015). This may relate to a sense of parental relief following infant survival and to the effects of continuous support received from follow-up services by those families with preterm born children with more severe chronic conditions. In addition, increased personal growth following a traumatic life event has been associated with the stressfulness of the event (Noy, Taubman-Ben-Ari, & Kuint, 2015). Factors such as these may help to explain findings reported by Moore et al. (2006).

Yet, in this review, I found that the hypothesis of a long-lasting impact of adverse birth experience was a prevalent starting point in the research among prematurity. Some studies in this review hypothesised that the parents' perception of their child's health as poorer than clinically established (Indredavik et al., 2005; Singer et al., 2007; 2010) may have resulted from the high-risk birth and neonatal period which had left a long-lasting parental perception of child vulnerability (Taylor et al., 2001). This perception of child vulnerability was understood to alter parenting behaviour (Brown et al., 2003; Indredavik et al., 2005). For example, Taylor et al. (2001) speculated on a long-lasting impact of adverse birth and prolonged neonatal intensive care, and that the initial parental distress may have compromised the parent-child relationship, which together with challenged child behaviours bidirectionally worsened outcomes among the parents and the child. Drotar et al. (2006), however, showed that the present parent risk factors and child disability were more influential in family impact than birth or neonatal risk factors. Parents of adolescents born preterm and full-term recalled their 'bonding experiences' with their children similarly (Indredavik et al., 2005).

As discussed earlier, in adolescence, child behaviour and psychosocial health may be more impactful factors in parental experiences than physical child disability (Barroso et al., 2018; Majnemer et al., 2007). These factors may challenge peer relations (Ritchie et al., 2018). Thus, the particular challenges in adolescence, such as peer relations, may appear as more pressing

concerns for parents in comparison with recall of adverse birth experience or prolonged infant hospitalisation, although theory may suggest that the impact of these experiences is more innate (Maccoby, 1992). At this point in time, those EP/VP families whose adolescents' challenges in relation to behaviour, independence, or/and peer relations are new or unexpected, may report poorer parent outcomes in comparison with families with EP/VP born adolescents who, due to their child's chronic condition/s, expected further difficulties (Moore et al., 2006).

Perhaps to remain within the 'boundaries' of the discipline, many studies in this review articulated their purposes to be to promote the parent-child relationship and consequently to improve child outcomes (for example consider Taylor et al. (2001) discussion). Even though framed as investigations of 'parent outcomes', such as depressive symptoms, anxiety, or stress, I found that the focus in these studies frequently remained on the consequences for the child, and the effects on parents remained secondary and therefore, intrinsically parent outcomes lacked value. Generally, the theoretical bases of these studies were not explicitly stated (Rodgers et al., 2009), but the reports implied to assess the consequences of adverse birth experience and consequently 'challenged' early parent-child relationship. The risk exposures of adverse birth and challenged early parent-child relationship were understood to impact outcomes among both the parents and children, and to generate a more adverse environment for the child to grow. These rationales referred to the psychoanalytic and attachment theories, as well as to the framework of parental determinism, as discussed in detail in the introductory chapter of this thesis.

In this review, studies conceptualised the preterm birth as a risk exposure causing changes in parenting behaviour and framed parents as relatively 'passive receivers' of the 'effects of prematurity'. Although deterministic thinking in relation to the parents' impact on child outcomes was evident in these studies, the active decision-making parental role was largely invisible, in both improving or hindering the preterm born child's outcomes long term. For example, parents were not considered to uptake behaviours such as intensive parenting practices to promote their children's outcomes, but rather they were seen as 'parenting' in a manner determined by their earlier life experiences. In parenting research among other populations, so called goal-orientated parenting behaviour, such as intensive parenting practices, has been a widely discussed discourse (Lee et al., 2014, p. 26-27).

In this review, a deterministic view of parental perception of child vulnerability altering parenting behaviour among the families with EP/VP born adolescents was inherent in the reports. For example, Indredavik et al. (2005) discussed the balance between parenting an 'initially fragile' child 'appropriately' and parental overprotection. Wightman et al. (2007)

after finding an association between ELBW and higher parental protectiveness at the child's age of eight years, although only among the families with ELBW NSI children, highlighted the importance in future research to examine the relationship between parental protection and monitoring and reduced risk taking among adolescent ELBW survivors. A detailed review of all the varying questionnaires utilised to assess parenting behaviour would have, perhaps, assisted me to pinpoint what exact parenting behaviours, dimension, or practices (Kuppens & Ceulemans, 2019) were included in concepts such as 'hostile parenting' (Pyhala et al., 2011) in the reports that assessed parenting behaviour. Nevertheless, these studies suggested a link between parenting practices and adolescent mental health outcomes (Burnett et al., 2017; Pyhala et al., 2011). Although hypothesised by Pyhala et al. (2011), Indredavik et al. (2005) and Burnett et al. (2017) both showed that the differing parenting behaviour among the parents of preterm born adolescents (aged 14 years and 15 to 20 years, respectively) did not result in poorer mental health outcomes among either the preterm born participants themselves or their parents, in comparison to the control families. Consequently, if attempts are made to attribute poor mental health outcomes or challenging social behaviours among EP/VP adolescents to the effects of 'ineffective' parenting (as defined by Maccoby (1992)) in relation to the level of child socialisation, caution may be warranted.

Yet previous research has demonstrated that a more protective family environment may function as a buffer against negative outcomes for VLBW children (Treyvaud, 2014); a relationship that has increased importance for adolescents with the lowest birthweights (Greenley et al., 2007). A study assessing the relationship between early adolescent family conflict and later adjustment difficulties following VLBW birth reported that adolescents at the highest medical risk defined as birthweight <750 grams were also more negatively affected by potential family conflict in early adolescence than controls (same cohort of families as Moore et al. (2006); and Taylor et al. (2001). Increased perceived family conflict at the age of 11 years was associated with increased negative changes in total behavioural functioning and externalising behaviours at the age of 17 years. However, it is worth noting that this association was found only among adolescent-perceived family conflict, whereas no relationship was found between parent reports and adolescent adjustment difficulties (Greenley et al., 2007). Generally, the families with EP/VP children included in this review reported low levels of family conflict (Burnett et al., 2017; Indredavik et al., 2005; Pyhala et al., 2011; Wightman et al., 2007).

2.6.5 Limitations of narrative data synthesis

The methodological approach to this systematic review had limitations. As mentioned above, due to a wide-focused review question, studies included in this review varied widely. Despite this, the review question helped me gain a greater understanding of the research field to date (Mays et al., 2005). Yet it challenged my ability to collate the findings from the reviewed studies. For example, due to the methodological disparities between the studies, it was not possible to undertake a meta-analysis. In addition, the presentation of findings from the thematic analysis of factors influencing long-term parent outcomes had a level of subjectivity in it. In addition to the key themes discussed in length, such as neonatal risk level and child disability, other themes also emerged during the analysis. Due to the length and scope of this report, not all themes were narratively explored in this report. The selection of most topical themes to be presented in this narrative review was supported by a second independent coder (ASM) who confirmed the themes' priority (Rodgers et al., 2009).

2.6.6 Conclusions and recommendations for future research

This review has concluded that research on long-term parent outcomes following EP/VP birth has been scarce and the findings are inconclusive, largely due to methodological limitations. The 17 identified publications assessing long-term parent outcomes following EP/VP birth reported a trend towards increased family impact among families with YA born preterm, particularly among those families with children with long-term morbidities. Yet, the reviewed studies generally did not distinguish between varying adolescent morbidity, such as cerebral palsy and behavioural disorders. The findings from this review suggested that parental concerns of their adolescent children's quality of life among EP/VP parents may have impacted their parenting behaviour. Generally comparable mental health outcomes between the groups of parents were found. Studies also reported positive family transformations. EP/VP birth impacted parents' educational attainment, job opportunities, and income. Thus, the level of family resources commonly measured at adolescence were not independent factors of the EP/VP birth.

Understanding long causal pathways had challenged the research on long-term parent outcomes. Methodological disparities between the studies challenged data synthesis. Lack of data on pre-pregnancy parental factors was a general limitation among the studies. Investigations from the parents' perspectives were largely missing as no major attempts had been made to convey perspectives of parents to situate their long-term outcomes in lived experiences. The theoretical bases of studies were not explicitly stated, but commonly referred to the psychoanalytic framework and attachment theory and followed the rationale of parental

determinism. The reports postulated on a deterministic view of parental perception of child vulnerability altering parenting behaviour among the families with EP/VP born adolescents, triggered by the adverse birth experience and neonatal illness.

Future research among families with adolescents born preterm could explore the potential independent associations between parent outcomes and varying adolescent morbidities, and examine particular parent outcomes, such as parental psychological health outcomes, in detail over time. Further research could focus on standardising methods, such as using agreed outcome instruments and aim to minimise selection bias, to improve the understanding of outcomes from parents' perspectives, and to better understand the overall framing of prematurity and 'parenting' in influencing research agenda.

Key findings:

- 1) Publications reported a trend towards increased family impact among families with YA born preterm, particularly among those families with children with long-term morbidities
- 2) Similar findings of no differences in mental health outcomes between EP/VP and FT parents were reported
- 3) Parental concerns of their adolescent children's quality of life may have impacted parenting behaviour among EP/VP parents
- 4) EP/VP birth may negatively impact parents' educational attainment, job opportunities, and income
- 5) Current research on long-term parent outcomes following EP/VP birth has methodological limitations and the findings are inconclusive
- 6) Research among prematurity has mainly focused on examining factors contributing towards negative long-term family outcomes
- 7) A hypothesis of the long-lasting impact of adverse birth experience and neonatal illness was a prevalent starting point in the research on long-term parent outcomes
- 8) The theoretical bases of studies were rarely explicitly stated, but the reports implied features of the psychoanalytic and attachment theories and the framework of parental determinism

Chapter 3 Research Methodology

My narrative review indicated that the research on long-term parent outcomes following EP birth is relatively uncommon. Studies have shown varying results such as positive and negative outcomes, and that there is a poor understanding of how the reported outcomes are situated in parents' lived experiences. With a single exception, the studies had emerged from the positivistic research tradition. Hence, my decision to use mixed methods to examine the parent outcomes in this population and embed the perspectives of parents in their lived experiences. Thereby I aimed to improve both the "depth and breadth" (Bazeley, 2018, p. 12) of the current understanding of parenting an EP YA. As this mixed methods study was situated within an ongoing larger cohort study, with its own imperatives, pragmatic decisions concerning the study design, such as sampling, were made to be able to contribute towards the dual aims of the cohort study and this investigation, and to maximise the knowledge generation.

3.1 Mixed Methods Study Design

I conducted a convergent design mixed methods (MM) study (Teddlie & Tashakkori, 2006), in which I explored how data from qualitative semi-structured telephone interviews exploring parents' experiences at the time their child transitioned to secondary school related to cross-sectional survey data about parenting stress, sociodemographic, child and parent health factors collected as part of a longitudinal national birth cohort study EPICure 2. Since outcomes relating to parenting behaviour, parent health or parental experiences had not previously been collected as part of the EPICure studies, this study was both exploratory and cross-sectional in nature (Hennekens & Buring, 1987, p. 108-109). The qualitative and quantitative elements of this study were conducted in parallel to each other. The concurrent design was fixed rather than emergent in nature as the design was determined in advance (Alavi, Archibald, McMaster, Lopez, & Cleary, 2018).

In their typology of MM research designs, Teddlie and Tashakkori (2006), authors who have both contributed extensively to the theoretical and methodological development of mixed methods research since the early 1990s, have presented a Methods-Strand Matrix (Teddlie & Tashakkori, 2006, p. 15) that defines various study designs based on the approach taken and number of research strands utilised. With the term 'research strand', the authors refer to a study process that includes three phases of 1) conceptualisation; 2) methodology and analyses (experiential phase) and 3) interpretation and inferences (inferential phase). Their typology excluded considerations of the theoretical basis of the study such as constructivism and particular research functions such as triangulation. Following their typology, the present study

was designed as a mixed methods multistrand concurrent study. According to Teddlie and Tashakkori (2006) in a concurrent multistrand MM design “at least two relatively independent strands: one with QUAL questions and data collection and analysis techniques and the other with QUAN questions and data collection and analysis techniques” (Teddlie & Tashakkori, 2006, p. 20) are integrated following strand-specific research results to form “meta-inferences” (Teddlie & Tashakkori, 2006, p. 20). Following from the concurrent multistrand MM design of this study, I was able to simultaneously examine the prevalence and sources of parenting stress, such as child birth status and family SES, and to explore parental experiences among EP parents within a single study. Thus, the design allowed me to explore varying aspects of the construct of parental experience of parents of an EP YA, and therefore, to improve current understanding (Teddlie & Tashakkori, 2006).

Similarly to the present study, Carpentieri, Elliott, Brett, and Deary (2016) were challenged to consider a mixed methods study design and additional vital components to an existing longitudinal cohort study. They implemented a qualitative research strand in an existing longitudinal cohort study, the 6-Day Sample, to deepen understanding of successful ageing among 171 Scottish senior citizens (Carpentieri et al., 2016). The quantitative strand of their study included researcher-defined outcome measures to assess aspects of successful ageing such as mental health outcomes and HRQoL. Qualitative interviews were designed to explore older peoples’ own perceptions of successful ageing. The authors emphasised that with a quantitative survey method they were able to describe prevalence and distributions of variables such as depressive symptoms among the study population. The interview data allowed them to contextualise participants’ experiences and explore framings of concepts such as ‘depression’ among older people (Carpentieri et al., 2016). Aligned with this, in the present study, I examined parent outcomes among the parents of EP YA, but also conveyed parental perspectives to situate the outcomes in lived experiences. Thus, this study design enabled me to bring forward other influential factors in parental experiences in addition to the predefined measure of parenting stress.

Although Carpentieri et al. (2016) did not explicitly state their MM study design by referring to an existing typology, they described a sequential study process. Their investigation aimed to improve the overall understanding of the concept of ‘successful aging’ with two types of data collection and analyses that were brought together in the inferential phase (Teddlie & Tashakkori, 2003, p. 687). A key object of their study was to explore how the qualitative data could improve the current, somewhat ‘quantitative’, understanding of successful ageing. In their data integration they used methods of triangulation, illustration, exploration of processes, and explanation. However, even though the authors had planned to explore, to an extent,

quantitatively driven topics such as physical health and social functioning in their thematic analysis of the interview data, the qualitative interviews had resulted in a more narrative accounts of experiences of ageing than what the authors had anticipated. The authors then discussed the general challenge in integrating quantitative and qualitative data as in their results the qualitative findings, although narrative and detailed, had remained somewhat ‘superficial’. Carpentieri et al. (2016) stated that they planned to produce a separate narrative analysis of the interview data in addition to their integrated analysis, thus depart from the MM research plan. Overall, the authors concluded that their quantitative findings correlated with the qualitative themes, but that they were also able to explore contradictions in health profiles in the participants’ narratives.

In a Canadian study of school success in adverse context, Jang, McDougall, Pollon, Herbert, and Russell (2008) utilised a concurrent multistrand MM research design (Teddlie & Tashakkori, 2006) to describe school factors that allowed the institutions to achieve academic excellence despite various contextual challenges such as a high employee turnover rate. Similarly to the present study, they utilised a validated survey instrument (School Life Survey) as their main quantitative outcome measure to assess the “school culture and leadership practices” (Jang et al., 2008, p. 228) among all teaching personnel in the sampled schools. A randomly selected sub-sample of teachers were invited to participate in a semi-structured interview to explore teachers’ perceptions of the schools’ success and challenges in supporting students to achieve academically. The qualitative research strand also included focus groups with students and parents, and in-depth interviews with head teachers. The researchers described their types of data to complement each other as the “general description of school improvement from the survey was enriched, elaborated, and clarified with contextually specific accounts of school success from interviews involving multiple perspectives” (Jang et al., 2008, p. 226).

In their report, Jang et al. (2008) illustrated varying integrated analytic strategies they had used to synthesise the data. Separate research teams allowed them to conduct the quantitative and qualitative research strands independently until the inferential phase. They discovered that their quantitative data showed relatively harmonious perspectives of school improvement, whereas the qualitative data revealed more variability in experiences. Consequently, the authors turned to a data consolidation and case analysis approaches as methods to explore the origins of these inconsistencies. Their data were brought successfully together by comparing qualitative themes against narrative descriptions of the quantitative data resulting in eight consolidated themes and narrative school profiles. As a limitation of their design, the convergent nature was posed due to time and resource constrains, and the authors postulated

that a fully integrated sequential design could have provided a more comprehensive understanding, as the authors would have been able to use the qualitative findings to guide the selection of quantitative data collection instruments (Jang et al., 2008). The design of the present study will be further illustrated and discussed with relevant empirical research examples as they related to the methods of this study later in this chapter. First, I will present a short discussion on the ontological and epistemological positioning of this study to contextualise the pragmatic decisions made on research methods.

3.2 Pragmatic Research Approach in Mixing Methods

Mixing research methods arising from different ontological viewpoints requires further consideration (Bazeley, 2018, p. 13-14; Creswell & Plano Clark, 2011, p. 38-39; Morgan, 2007, 2014). In this study, the qualitative research strand emphasising a constructivist worldview was positioned within the existing positivistic main study (EPICure2@11), influencing research methods such as sampling, as will be discussed in detail below. In the methodological discussions arising from different ontological standpoints, mixed methods researchers have referred to a concept of ‘research paradigms’ (Kuhn, 1996, p. 175). Although definitions vary, ‘research paradigm’ is commonly understood as a consensual set of beliefs and practises accepted by a research community (Creswell & Plano Clark, 2011, p. 39; Kuhn, 1996, p. 175; Morgan, 2007, 2014). A recent dominant research paradigm has focused on careful consideration of ontology, epistemology, and methodology in determining research methods (Morgan, 2007). However, when the emphasis is placed on the ontological question of reality and the epistemological question of knowing about that reality followed by the methodology, a top-down model is built in which ontology inherently guides research methods and questions (Morgan, 2007). The epistemological paradigm eventually leads to a “paradigm incompatibility” (Morgan, 2007, p. 5) where mixing methods becomes impossible.

In his review, Morgan (2007) discusses a pragmatic approach as a paradigm shift away from the epistemological top-down paradigm in mixed methods research. Following Kuhn (1996), Morgan demonstrates how paradigm definitions are negotiated amongst groups with scholarly interests, their boundaries are arbitrary, and subject to favouritism. Therefore, rather than claiming research paradigms to be defined by fixed principles of philosophy of knowledge, they should be understood as negotiated pragmatic approaches (Morgan, 2007).

In line with this, the narrative review presented above demonstrated how research questions and methods were often determined by dominant agenda. A unifying feature of the reviewed studies was that prematurity was seen as a risk exposure whose effects were assessed with

instruments generally measuring negative parent and family outcomes. More recently, the research agenda has shifted towards appreciating complexity. For example, the research interest in the EPICure2@11 Study to better understand wider factors, such as family context or parenting behaviour, influencing preterm born survivors' outcomes, has in itself been a 'negotiated' response to a paradigm shift. Since individual understandings are built and shaped via social interaction (Giddens, 2013, p. 41-52), positivistic research agendas are influenced by the same social and political context through the 'scholarly community' conducting the research (Denscombe, 2008; Morgan, 2007, 2014).

The pragmatic approach presented by Morgan (2007) moves its focus from ontological discussions to shared meanings, joint action, and satisfaction with generated understandings created by communication and persuasion between methodologies. Thus, the question is how much shared understanding can be created and how this improved understanding can be used in practice. Contrary to the epistemological paradigm, pragmatism does not draw on external systems to explain beliefs and therefore no a priori basis exists to determine the limits of the 'communication' between methodologies (Morgan, 2007). Pragmatic research inquiry is regarded to generate cautious answers instead of ultimate truths as wider understandings gained from experience and experiments can be ever modified and complemented (Johnson & Onwuegbuzie, 2004). The appropriateness of results from pragmatic inquiries can be determined by future empirical enquiries (Morgan, 2007).

In pragmatic research the question directs the inquiry, followed by a careful consideration of 'what is possible', the "line of action" (Morgan, 2007, p. 20) and its "workability" (Morgan, 2007, p. 20) in research methods. The methodology I present in this thesis is a study of the 'line of action' (methods) chosen for this study to best answer the research question, which determined the need for a mixed methods inquiry. Thus, the approach is abductive, moving between induction and deduction (Morgan, 2007).

This study can be regarded as a pragmatic attempt to integrate constructivist and positivistic research methodologies as described by Morgan (2007). The interesting or most relevant question is not the conflicting ontologies of 'truth' in these worldviews, but rather the importance of the research question and what pragmatically works best. For example, the narrative review presented above showed how researchers commonly utilised the concept of 'recall bias' to explain conflicting research findings; as time had passed since the birth, the parents were unable to recall the negative effects. However, in a pragmatic sense, the most important or relevant question is not whether bias 'exists', but rather what significance does it have in the everyday lives of parents reporting positive or negative outcomes; how parents

experience, understand, or explain their social realities when they report these outcomes. In pragmatic research, individual's experiences of actions in the world are central to the inquiry (Morgan, 2007). The claim that the world exists outside of observation (positivism) or that everyone will have their own individual creations of the world (constructivists) are both meaningful as statements of "human experience" in pragmatic understanding (Morgan, 2014, p. 1048). When viewed in such a way, either the qualitative or quantitative section cannot be seen to 'add on' to the other, but rather that they 'work together' throughout the study design.

3.3 Methodological Applications Following Pragmatism

The overall design of the present study is illustrated in Figure 3-1 below. The figure is based on a diagram originally created by Teddlie and Tashakkori (2003; 2006, p. 688 & p. 21, respectively). As discussed earlier, the conceptualisation, experiential, and inferential phases for both quantitative (illustrated as rectangles) and qualitative (illustrated as ovals) research strands are shown. Although Teddlie and Tashakkori (2006) described concurrent MM studies to have two 'relatively independent' research strands, the present study departed from this design at two points of sampling and data analyses (illustrated as broken lines with arrows) for practical reasons to ensure the 'workability' (Morgan, 2007, 2014) of this study within the larger cohort study EPICure2@11. Although methodological appropriateness is paramount for the validity of research findings, also so called 'opportunistic designs' that may not rigidly follow predetermined research models have been considered appropriate in MM research (Jang et al., 2008; Teddlie & Tashakkori, 2006). Scholars have suggested that attempting to adhere to 'pure methodologies' to reduce the likelihood that the rigour of the study could be challenged, the need to 'qualify' the inquiry undertaken may hinder best answering the chosen research questions (Clarke & Visser, 2018; Long, McDermott, & Meadows, 2018; Thorne, 2011).

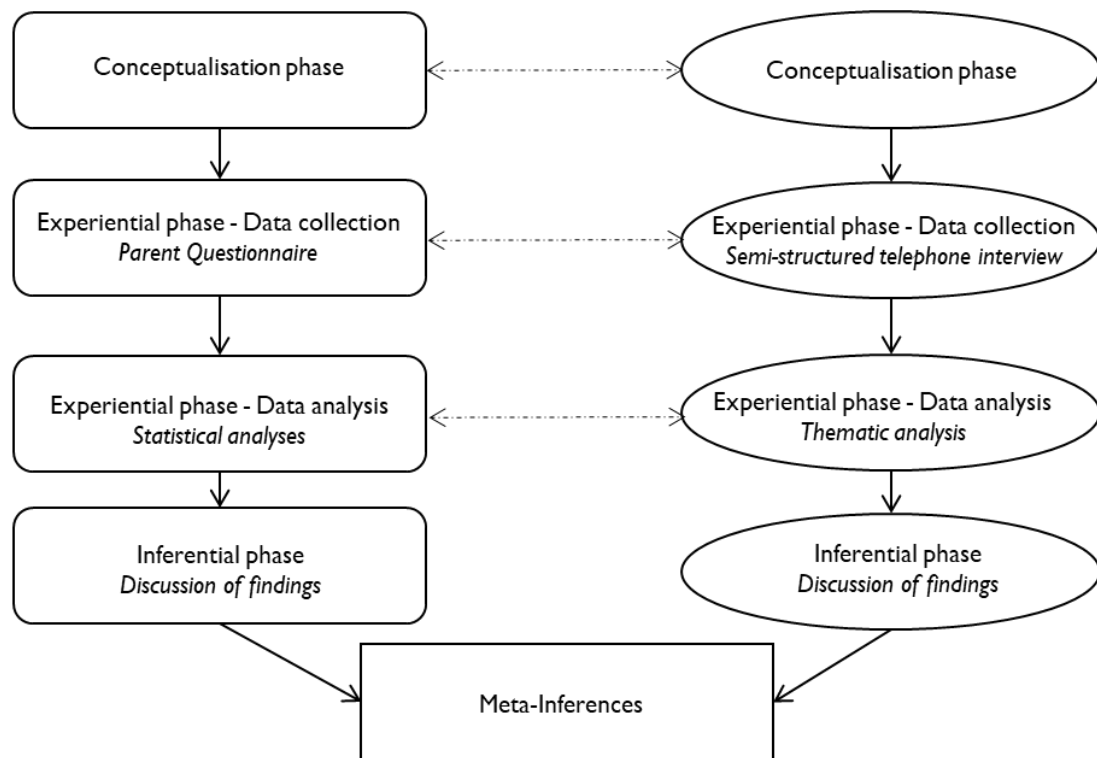


Figure 3-1 An illustration of the multistrand concurrent mixed methods design utilised in the study

Rectangles represented QUAN and ovals QUAL research strands. Arrows indicate progress in the research process. Broken lines with arrows represent points at which research strands interlinked (Teddle and Tashakkori 2003; 2006, p. 688 & p. 21, respectively).

In the conceptualisation phase of this study, research purposes and questions were formed based on previous theoretical and empirical research on the parent-child relationship, determinants of parenting behaviour, parent outcomes among parents of preterm born children over time, such as parenting stress, and culturally dominant framings of parenthood. As the preceding discussions in the introductory and review chapters of this thesis have indicated, the various conceptual frameworks of parenting stress, parental experiences of EP parents over time, and the framework of parental determinism are interlinked in their potential impact on parenting behaviour (as a reminder please see Figure 1-4 in Chapter 1). These interlinkages between the quantitative and qualitative research strands' conceptualisation phases are illustrated with a broken line with arrows in the Figure 3-1 above.

The quantitative and qualitative data were collected concurrently, but sampling occurred sequentially for the quantitative and qualitative research strands of this study (Teddle & Tashakkori, 2006). The quantitative strand employed probability sampling techniques as

designed for the EPICure2@11 Study. These are described in detail in Chapter 4 together with quantitative data analysis and results. Sampling for the semi-structured telephone interviews followed the quantitative sample and was a subset of it. The sampling strategy aimed to gain a maximum variation sample (Teddlie & Yu, 2007) as detailed in Chapter 5 together with qualitative data analysis and results.

The qualitative sampling method was impacted by the design of the EPICure2@11 Study and pragmatic choices regarding sampling methods were made. In the main project, the sampled families were invited to take part in the study in clusters during a lengthy data collection period (approximately 19 months). It was anticipated that some families would take longer to consider their participation in the main study, whereas others would be more eager to take part. For example, evidence has indicated that families with socio-economic and/or demographic risk factors, such as ethnic minority or multiple birth status, may take longer to provide data in follow-up studies of child neurodevelopment (Johnson et al., 2014). It was also anticipated that some child assessments would take longer to schedule. Due to these reasons, parents became eligible to be approached for the interview at varying times during the main study. Thus, an element of random sampling was introduced to the qualitative research strand to *begin* the sampling process to ensure that parents across the main study's data collection period had an opportunity to participate in the interview. When more parents had contributed data to the main study, and the pool of eligible participants grew, the case selection process for the interviews focused only on parental characteristics as detailed in Chapter 5, and the random element of sampling was dropped. Completed pen-and-paper Parent Questionnaires containing the quantitative data were visually screened to select parents with a range of socio-demographic characteristics to be approached for the interview. The design of the sampling process was finalised in consultation with my supervisors (NM, AL).

Regarding the decisions on sampling, to an extent, the design of the main study determined the design of the qualitative research strand. Alternative qualitative sampling methods in similar concurrent multistrand MM studies have been reported. For example, studies have randomly selected participants from larger quantitative research strands to also contribute data to the qualitative section of the study (Jang et al., 2008; López & Tashakkori, 2006; Roll-Pettersson, 2001). Yet, if the study is limited in size, as was the case in the present study, when participants are selected at random, opportunity to discover and present varying cases may be reduced (Teddlie & Yu, 2007). A qualitative study, which was similarly to the present study situated in a context of two longitudinal population-based follow-up studies of extreme and moderate prematurity in Sweden, invited all eligible parents from both cohorts to take part in a telephone interview exploring parents' birth stories when their children were aged between

10 and 12 years, resulting in a sample of 80 parents (Widding & Farooqi, 2016; Widding et al., 2020). An approach as such was not, however, feasible in the present study as I was the only interviewer; if all parents in the EPICure2@11 Study had been invited, I would have had to decline participation of some, potentially resulting in another selection process. The fact that none of the EP parents invited to take part in the interview declined to participate, even though they had already contributed their time to the main study (e.g., completed several questionnaires, assisted in liaising with the child's school, or/and hosted a home assessment for the child), may suggest that parents in the EPICure 2 cohort were generally interested to be interviewed about their experiences. Studies that have recruited so-called convenience samples (Teddlie & Yu, 2007) for example by advertising their projects with posters or online, may have attracted participants with particular motivations (Kantrowitz-Gordon et al., 2016) such as those who wished to discover new possible sources of support (Hallberg et al., 2009).

Carpentieri et al. (2016) used a sequential design in their sampling frame for the qualitative interviews about successful ageing in the study discussed earlier in this chapter. They first collected and scored the quantitative surveys to describe varying ageing profiles based on the respondents' physical and psychological functioning, general health status, and cognitive functioning in childhood. Then, the authors purposefully sampled 33 participants to interview based on their quantitative profiles to elicit varying experiences. In contrast to their design, parental characteristics considered in the purposeful qualitative sample of this study, were largely decided in advance based on a review of literature. Parent working status was added to the sampling frame retrospectively as it was missed in error. The design of the qualitative sample in the present study, however, was not based on prevalence or distribution of the quantitative variables, such as parenting stress scores, and it did not aim to represent the quantitative sample directly. To select parents to approach, I visually screened their Parent Questionnaires to identify parents with varying backgrounds. On occasions, I hand-scored the parenting stress index questionnaires to identify parents who reported particularly high levels of parenting stress (>91st centile). These parents were purposefully approached to include their experiences in the interview data as well. Based on evidence, parents with such a high stress scores were likely to be only a few in the quantitative sample. The sampling process will be further discussed in Chapter 5.

Carpentieri et al. (2016) qualitative research strand was a response to the limitations identified in a quantitative data only. Despite the carefully planned sampling frame, their sampling process was not without challenges. At the time of the interview, some participants' life situations had changed significantly in terms of personal health, or/and because of a death of their partner and therefore, their circumstances no longer represented their earlier quantitative

health profile. In fact, due to these reasons a few participants declined the interview, which limited the authors' original aim for the qualitative sample to represent the quantitative cohort. The authors noted that since the qualitative sample was only a subset of the quantitative cohort, attention needed to be paid when reflecting the types of data against each other, as, for example, the interviewed participants were cognitively better functioning than the total 6-Day Sample cohort. Their qualitative data brought forward personal experiences of successful ageing rather than generalisable facts.

As multistrand concurrent MM designs are generally resource-heavy, they are commonly utilised in large-scale studies with distinct quantitative and qualitative research teams (Teddlie & Tashakkori, 2006), as per the empirical research example discussed above (Jang et al., 2008). For example, data analyses (please see Figure 3-1 above) may trigger a return to the data collection phase as more strand-specific data are needed to answer the research question. As in the present study I worked alone simultaneously with both types of data, the data inevitably 'spoke' to each other (illustrated with a broken line with arrows in Figure 3-1 above) during the data analyses phase. Both types of data were analysed independently using appropriate methods best suited to answer the quantitative and qualitative research questions, respectively (Teddlie & Tashakkori, 2006). Even though I did not attempt to directly merge the types of data (e.g., transform data to be merged), as followed from the methodological approach employed in this study (i.e., a convergent design with a single researcher), the qualitative and quantitative data analyses, however, were in a conversation with each other. When analysing the qualitative data, a hypothesis of 'extensive caregiving efforts' was generated that was then examined in the quantitative data, and the quantitative data analysis revealed phenomena, such as increased likelihood of high parenting stress among parents with university degree education, whose presence was then explored in the qualitative data. This meant that the data integration occurred throughout the analysis phase as a bidirectional movement between the types of data (Morgan, 2007).

In a multistrand concurrent MM study, in the inferential phase, data convergency leads to meta-inferences (Teddlie & Tashakkori, 2006) in which the data types are integrated at a level of interpretation (Caracelli & Greene, 1993), such as demonstrated by Jang et al. (2008) as discussed earlier in this chapter. In this study, following the quantitative and qualitative analyses, the data were planned to be narratively interpreted in a light of how they answered the mixed methods research question (Creswell & Plano Clark, 2011, p. 212-213). The aim of the integrated analysis was to explore to what extent the quantitative and qualitative results converged, and if so, how the quantitative outcome measure, parenting stress, related to

parental experiences. This was to better understand parental experiences of parents of EP YA. The narrative data discussion is presented in Chapter 6.

In their reflective review of literature on pragmatic research, Clarke and Visser (2018) discussed, based on their own experiences, challenges with research rigour that early career researchers may experience when they are not following a 'pure' research methodology. The authors described pragmatic research as a step away from selecting a single methodology to selecting elements from possible methodologies. They concluded that the pragmatic research approach in their study had resulted in most 'sensible and practical' methods utilised to answer their research question. Yet this had only followed from a time-consuming extensive review process of literature during which it had been challenging to pinpoint the most valuable methods from all available options, and to fully assess the implications that the chosen methods would have on the end data, particularly when the authors had to alter their original data collection plan due to changes in circumstances (Clarke & Visser, 2018). Aligned with this, although my chosen convergent multistrand MM study design was influenced by both time and resource constraints, when deciding on the parallel design, I had not fully appreciated or expected the challenges that I may face when analysing the types of data alone concurrently. Neither had I fully realised what implications the mixing of quantitative and qualitative research strands on various points during the study could have on end data. I will discuss further these methodological limitations in the last chapter of this thesis.

3.4 Theoretical Positioning of the Study

The theoretical basis of this investigation lay in critical sociology with a social constructionist viewpoint (Marvasti, 2019). The theoretical approach influenced the design, analyses, interpretation of findings, and reporting of this study (Alavi et al., 2018). As the systematic review presented in the preceding chapter indicated, previous underlying metatheory in research on long-term parent outcomes following EP/VP birth has largely been positivistic. A particular viewpoint to explore the socially constructed aspects of phenomena such as appearance of dominant discursive framings of 'parenting' in research agenda, were largely lacking (Alavi et al., 2018; Marvasti, 2019).

In his theoretical review of utilisation of quantitative and qualitative data in constructionist research, Marvasti (2019) discussed varying forms of constructionism directing research practice. Forms of constructionism may vary from an understanding that nothing can be assumed a priori to a more pragmatic approach in which some entities can be treated as given. For example, in a strict form of constructionism words/talk are the context in that no outside

world/context exists; in other words, interviewees enact the context while they speak. However, following from a less strict understanding of constructionism, the main purpose of social constructionist research can be regarded as to understand how meanings are given to matters and how those meanings may change over time (Marvasti, 2019), such as in the case of the more recent framing of ‘parenting’ as ‘doing’ rather than ‘being’ (Smith, 2010), as discussed earlier in the introductory chapter of this thesis. In his own empirical research, Marvasti has examined the interplay between objective social conditions and practices of social construction; for example, the role that homeless people’s poverty (quantitative data) formed in how they constructed themselves as ‘worthy’ clients of homeless shelters (qualitative data). In the present study, I employed a constructivist viewpoint to better understand how parental experiences were built in the parents’ interviews. Quantitative data, such as parenting stress scores, were utilised to enrich understanding of parental experiences and vice versa. Following Marvasti (2019), it is worth noting that the quantitative research strand could also be perceived as constructive in that the ‘parent’ can be constructed or deconstructed with figures and numbers such as those about parenting stress or SES. Varying, to an extent subjective, constructs could be illustrated as per decisions on the data to be presented. The tendency in research on long-term parent outcomes following EP/VP birth to focus on discussing adverse outcomes instead of robustness in outcomes could be regarded to reflect Marvasti’s position.

In addition to individuals’ social relations and interactions in the world, also research in itself may construct what an entity such as ‘parenthood’ may entail (Giddens, 2013, p. 18-28). Therefore, both impersonal agents such as culture, and personal agents such as a group of researchers may shape and re-define our understandings of the social fact of ‘parenthood’. As I introduced at the beginning of this thesis, and what will become more apparent in the qualitative section of this study, I have taken dominant discursive phenomena such as ‘intensive parenting’ and ‘parental determinism’ to be parts of the current dominant Western understanding of normative parenting behaviour. In this thesis, I will examine how the parents’ descriptions related to these discursive concepts, and how they had appeared in research agendas and interpretations of previous mainly positivistic research findings in the research field of prematurity.

The viewpoint in this study also sprang from my own multi-disciplinary background in both health and social sciences, with a particular interest towards improving women’s and children’s wellbeing and status in society. Throughout this project, when I explored the phenomenon and the concepts under investigation, such as ‘extreme prematurity’, ‘parental determinism’, or ‘parenting stress’, I employed my previous disciplinary knowledge in

Nursing, Epidemiology, and Development Studies. Thus, the knowledge I have gained through my studies in Nursing, Epidemiology, Sociology, Anthropology, and Health Economics are all echoed on the pages of this thesis. I chose to explicitly employ a critical sociological lens to view the research field of prematurity, to not only assess and describe parent outcomes in the chosen population and explore the emerged image in a wider sociocultural context, but also to utilise the critical lens on the research field itself with an ultimate aim to improve knowledge.

Sociology explores the social construction of various forms of knowledge in a given sociocultural context (Bradby, 2012, p. 1) and aims to shed light on the connections between individual struggles and larger societal issues. The term ‘critical’ emphasises sociology’s role as an “argumentative discipline” (Bradby, 2012, p. 3). As well as being an analytical systematic exploration of concepts, attached meanings, and/or understandings, sociology has an advocacy role by exposing “taken-for-granted” norms and “undoing assumptions” (Bradby, 2012, p. 36). Consequently, the discipline makes a unique contribution by bringing forward social debates and controversies (Tierney, 1999), while envisaging more ‘humane’ future societies (Bradby, 2012, p. 3).

Within the social constructionist viewpoint (Marvasti, 2019), sociological investigation may ‘deconstruct’ the social scaffolding around a meaning by breaking down the common understandings of it for critical examination. The approach not only examines the sociocultural framings of a concept, but also considers that power may have been used in its construction (Rosen & Suissa, 2020). For example, Macvarish and Lee (2019) systematically examined how the ‘parent’ was constructed in submissions to a government committee call to assess the evidence-base for early-intervention programmes in the UK. Their critical examination demonstrated that the parent was constructed as gendered (a woman) and primarily solely responsible for their children’s outcomes (Macvarish & Lee, 2019). Framing ‘parenting’ as “an independent mechanism through which negative outcomes for children can be avoided” (Dermott, 2012, p. 2) may, for example, avoid governmental responsibility to support families with generally poorer child outcomes, such as those from low income backgrounds (Dermott, 2012). Additionally, framing of ‘parenting’ as an undertaking that is inherently resource-heavy and, therefore financially expensive, may be viewed as an ‘agenda’ to promote consumerism at present, and to ensure the ‘production’ of a next generation of eager capitalist consumers (Thornton, 2011). Therefore, the concepts under investigation may not only be viewed as influenced by social perspectives, but also as ‘produced’ to support particular ways of thinking, thus deserving critical examination (Bradby, 2012, p. 26-28).

I employed this critical theoretical perspective to explore parental experiences among parents whose children were born extremely prematurely at the time their children were entering adolescence. As discussed in Chapter 1, I recognised that dominant discursive framings, such as parental determinism may have simultaneously influenced thinking in research paradigms as well as in parenting practices. By viewing traditionally positivistic research practice, such as neonatology, from a sociological perspective allowed me to critically examine claims of objectivity. Thus, this study was not only a sociological investigation of parenting experiences following EP birth, but also a critical exploration of concepts and understandings used to study and understand extreme prematurity. The focus of this thesis was on analysing and understanding social framings of a phenomenon and pointing out controversies and conflicts in order to bring about change (Tierney, 1999).

Chapter 4 Parenting Stress and Health-related Quality of Life

In this chapter, I will present results from the quantitative data analysis of parenting stress and parent perceived HRQoL. The objective of this quantitative research strand was to identify sources of parenting stress among primary caregivers of EP YA and their age, gender and school class matched control children's primary caregivers. First, I will discuss literature on parenting stress among preterm born populations over time and explore factors influencing parenting stress scores. This short discussion will complement those presented earlier in the introduction and the review chapters of this thesis. Following the introduction, the methods used, the available data, and the data analyses conducted are described in detail. Then, in the results section, I will describe respondent characteristics and present descriptive results of both the PSI-4-SF and the SF-12v1 questionnaires. I will then present the main results from the multiple linear regression and logistic regression analyses in sections 4.4.3 and 4.4.5, respectively. The findings will be further interpreted and discussed.

4.1 Introduction

Research on parenting stress levels among parents of adolescents has been uncommon (Barroso et al., 2018). Further, parental experiences among parents of EP children have rarely been examined (Wilson & Cook, 2018). Among younger preterm born populations, when parent outcomes have been assessed, Abidin's Parenting Stress Index (2012) has been a popular outcome instrument (Treyvaud, 2014). Overall, only marginally elevated parenting stress levels have been found among parents of preterm born children in comparison with FT parents. Although, the meta-analysis conducted by Schappin et al. (2013), with one exception, included studies of families with children under the age of six years who were born with varying levels of prematurity (<37 weeks' GA). As presented in the introduction to this thesis, later in childhood, parents of VP born children have generally reported only modest elevations in stress levels in comparison with parents of FT born children and the general population (Landsem et al., 2014; Linden et al., 2015; Schappin et al., 2013; Treyvaud et al., 2014).

Parenting stress is a theoretically related concept to parental psychological health and, on occasion, these outcomes have been found to correlate (Linden et al., 2015). Rates of postnatal depression among mothers of preterm born children (<38 weeks' GA) have been found to be higher than among the general population (Vigod, Villegas, Dennis, & Ross, 2010). After the initial response, during the first year following the birth, symptoms of psychological distress among parents of VP born children have been found to decrease (Misund et al., 2013; Singer et al., 1999). During childhood, parental psychological distress, such as depressive symptoms

or/and anxiety, have been suggested to generally decrease among parents of preterm born children (Treyvaud, 2014). As per the finding from my systematic review, in adolescence, parents of preterm and full-term born children reported comparable, generally low, levels of parental psychological ill-health.

Child factors and parenting stress - Somewhat inconsistent findings have been reported on the impact of child age on parenting stress. Some studies among parents of VP born children have found that the initially high parenting stress scores decrease during early years of childhood (Treyvaud, 2014; Treyvaud et al., 2011) but may increase again in middle childhood (Treyvaud et al., 2014), whereas others have reported more stable levels of stress among parents of preterm born children (Gerstein & Poehlmann-Tynan, 2015; Schappin et al., 2013). Yet studies have reported that after a decline in the initially heightened stress levels, more stable or increasing levels of parenting stress in early childhood may occur, particularly among parents of VP born children with medical concerns (Singer et al., 1999). In a RCT of parental sensitising early intervention programme, Landsem et al. (2014) found a trend of decreasing total parenting stress scores during early childhood (till child age of five years) among all parents, except among mothers of preterm born children (BW <2,000 grams) in the non-intervention arm, whose stress levels remained stable or slightly increased over time. Between the child's ages of five and seven years, parents' stress levels in the preterm born intervention arm increased slightly (Landsem et al., 2014).

As the preterm born child gets older, other child factors, such as neurodevelopmental disability and/or behavioural disorders, may become more impactful. Parents of older preterm born children have generally reported higher levels of child-related parenting stress in comparison with parent-related stress (Brummelte et al., 2011; Schappin et al., 2013; Treyvaud et al., 2011; Treyvaud et al., 2014). Among parents of VP born children, from the age of eight months to 18 months, parents of VP girls reported decreasing total parenting stress, whereas parents of boys reported an opposite trend. The increased parenting stress scores among parents of boys were associated with a more prominent decline in child cognitive development (Brummelte et al., 2011). Yet among the same group of parents at child age of seven years, externalising behaviours rather than child IQ were associated with high parenting stress scores. Perhaps influenced by cultural stereotypes, parents of girls found their child's problem behaviour as more stressful than parents of boys (Linden et al., 2015). As sustained high levels of parent-related parenting stress from child ages of two to seven years have been found among mothers of VP born children (Treyvaud et al., 2011; Treyvaud et al., 2014), it may be that those parents who experience parenting stress that relates to the personal parent factors, rather than child behaviour, may be more likely to persist over time.

Among other at-risk populations, specifically children with autism, developmental delay, chronic illness, or with or at risk for behavioural and/or mood disorders, child behavioural problems have been associated with increased levels of parenting stress. This association was not impacted by child age when a majority of the studies included in the meta-analysis had assessed parenting stress levels among parents of children under the age of 12 years (Barroso et al., 2018). The validity of a decrease in parenting stress levels in early childhood is additionally supported by other theoretically related constructs such as parental perception of child vulnerability (Tallandini et al., 2015) and highly protective parenting behaviour (Wightman et al., 2007) demonstrating a similar trend of decrease over time as the child grows. Among a general population sample of British parents, the proportion of parents reporting intensive parenting practices reduced with increasing child age (Yerkes et al., 2019).

A noticeable finding from the systematic review presented earlier in this thesis was that some evidence was found that towards adolescence, parenting stress levels among parents of full-term born children increased, whereas they decreased among parents of children born VP with low medical risk. In addition, some evidence indicated that the parents of preterm born low medical risk children reported lowest levels of parenting stress in adolescence (at the ages of eight and 14 years) in comparison with parents of full-term born adolescents and parents of preterm born high medical risk adolescents (Singer et al., 2010). Yet, it is important to note that the limitations in methods of these early cohorts may have challenged the validity of their findings; for example, as discussed in detail in the review chapter, Singer et al. (2010) reported considerable attrition to their study sample over the years.

Parent and contextual factors and parenting stress - Parent factors (e.g., gender, age, education, income, marital status) may impact parents' ability to cope with stress (Lazarus, 2000). In general, among all parents, factors such as single parenthood (Anderson, 2008), low parental educational attainment (Larkin et al., 2021; Singer et al., 2007; 2010; Zerach et al., 2015), low social support level (Noy et al., 2015) and physical illness (Almogbel et al., 2017; Anderson, 2008) have been found to be associated with increased levels of parenting stress and decreased parental wellbeing. Yet, among a sample of generally well-educated parents of preterm born children, educational level and parenting stress were not associated (Linden et al., 2015), whereas among a sample of VP parents with low educational attainment, level of education was associated with parenting stress (Ong, Chandran, & Boo, 2001). As per my systematic review, preterm birth may impact parental educational attainment (Singer et al., 2007). In parent perceived quality of life (QoL), contextual factors such as support systems and parent factors such as mental health have been found to impact more parents' perceptions of their QoL in comparison with child factors (Amorim, Silva, Kelly-Irving, & Alves, 2018).

In comparison with fathers, mothers have reported higher levels of parenting stress, which may have related to the primary caregiver role that women assume more commonly in comparison to men (Baia et al., 2016; Schuetz Haemmerli et al., 2020). In the NICU context, older maternal age has been associated with lower levels of stress related to the altered parent role and stressfulness of the NICU environment and in later childhood older mothers may have wider social support networks which may help to buffer against parenting stress (Schappin et al., 2013).

When caring for a preterm born child is perceived as demanding (Brummelte et al., 2011), factors such as families' SES, social support level, parents' age or level of education may play a more significant role later, when parents care for their child at home (Baia et al., 2016). Studies have postulated that well-educated parents may have increased capacity to respond to their children's behaviours, but also that they have more financial resources available to utilise in the parenting task, and consequently, they may experience less stress in their roles as parents (Almogbel et al., 2017). Among other clinical groups (children with behavioural disorders), higher levels of parenting stress among low-income families have been suggested to be linked to a lower tolerance of child misbehaviour among parents who experience multiple other stressors in their lives due to the family's low-income status (Barroso et al., 2018). Yet, contrary findings, such as parental educational attainment having no association with parenting stress levels among parents of adolescents with ADHD have been reported (Wiener, Biondic, Grimbos, & Herbert, 2016). Findings like these may suggest that when child behaviours are particularly challenging, factors supporting parental coping, such as education, are not sufficient to buffer against feelings of stress. Yet, it is important to also consider that studies have, in general, used a wide range of variables to indicate SES such as the Hollingshead Four/Two Factor Indexes, and varying composite variables including factors such as ethnicity, parental educational attainment, neighbourhood poverty, marital status and/or number of emergency contacts (Jaekel et al., 2012). Thus, findings may be challenging to compare directly.

4.2 Methods

4.2.1 Study setting

The original EPICure 2 cohort comprised 1041 infants born in England less than 27 weeks of gestation in 2006 who were discharged alive from hospital (Costeloe et al., 2012). Following discharge, the first follow-up took place at the children's median age of 34 months (age corrected for weeks of prematurity) between 2010 and 2011. At that time, all families were invited to participate, of which 576 (55.3%) consented for their children to be clinically

assessed by trained independent study assessors. As a part of the re-assessment, the parents were invited to complete self-administered questionnaires assessing their perceptions of their child's health and abilities, and to collect sociodemographic information (Moore et al., 2012). No parent outcomes were collected. The EPICure 2 cohort and its earlier phases have been described in detail previously (Costeloe et al., 2012; Moore et al., 2012).

EPICure 2 at 11 years of age (EPICure2@11 Study) re-evaluated key physical, psychological, and social outcomes at an important phase in the lives of the participants and their families as they transitioned to adolescence and secondary education, using standardised established measures. The study's main aim was to determine any changes in later life outcomes following EP birth in line with advances in practice since the first EPICure cohort established in 1995. As discussed earlier, EPICure 2 has shown improved survival at all gestations (Costeloe et al., 2012) whilst the proportion of neonatal and early childhood morbidities have remained similar between the cohorts (Costeloe et al., 2012; Moore et al., 2012); an outcome that is in line with other similar birth cohort studies (Ancel et al., 2015; Tommiska et al., 2007). As survival has continued to improve, the outcomes for EP children and the wider factors influencing these outcomes have become a major point of interest. However, few studies have explored the long-term outcomes from the parents' perspectives; a gap in knowledge that was reflected in the aims of this study and further explored in Chapter 5.

The EPICure2@11 Study began on 27 March 2017 when the first recruitment letters were sent out to the families. Due to the large size of the EPICure 2 cohort, a sample of children from two geographical areas in England based on neonatal care networks at birth were included. It was established that the 17 included neonatal clinical networks were representative of the total EPICure 2 cohort as per i) infant survival rate; ii) survival rate of infants born less than 25 weeks of gestation; iii) average Index of Multiple Deprivation (IMD) decile (Ministry of Housing Communities and Local Government, 2019) based on the mother's postcode at the time of delivery; iv) proportion of children alive at 2.5 years, and v) proportion of children assessed at 2.5 years.

Four hundred and eighty-two approximately 11-year-old cohort children (46.3% (482/1041) of the total cohort) and their families were invited to take part in a clinical child assessment taking place at the participants' schools (or homes if preferred). The control participants were recruited amongst the classmates of the EPICure 2 cohort children. All control participants had been born full-term (≥ 37 weeks of gestation). The last set of recruitment letters were sent out on 16 November 2017. The postal, email and telephone contacts to recruit participants

were ceased in October 2018, when the maximum number of contacts were made, and no new families consented.

The day-long clinical child assessment consisted of cardiovascular, respiratory, motor-sensory, and growth-related medical assessments, and of psychological assessments relating to child cognition and academic skills. In addition to this clinical assessment, families were sent self-completion pen-and-paper questionnaires collecting data on parent perception of child health and abilities, sociodemographic information, parenting stress, and parent physical and mental health (i.e., Parent Questionnaire). The survey used standardised validated instruments. The parents were asked to return their completed questionnaires using a free-post envelope.

The clinical data collection took place between 7 June 2017 and 18 December 2018. The last Parent Questionnaire was completed on 24 January 2019 and returned by post. In addition to undertaking the present study, I conducted the medical assessments in Southern England together with a psychology assessor. A second team consisting of a clinician and a psychologist was based at Leicester University and assessed participants in the north of the country.

4.2.2 Participants

Details of inclusion criteria and recruitment of infants into the EPICure 2 cohort in 2006 have been described previously (Costeloe et al., 2012). In short, infant gestational age was determined with either the earliest dating ultrasound scan (97%), certain dates of last menstrual period (0.5%) or clinical estimation (2.5%) according to the then accepted best practice. All maternal units in England contributed data to the study (Costeloe et al., 2012).

The control children were recruited as part of the EPICure2@11 assessment. Parents responded on the written Informed Consent Form (ICF) and indicated that their child was born full-term. One to three control participants were matched for child's school class, age (± 3 months) and gender to ensure comparability of clinical and psychological outcomes. Sampling techniques and recruitment processes as they related to this study are described in detail in the following sections. Data on all parents who consented for their children to take part in the EPICure2@11 Study were included in the quantitative section of this study.

4.2.3 Ethics and consent

The study protocol was approved by the UCL Research Ethics Committee (Ref:10175/001) and by the University of Leicester (Ref:10225). Every parent gave their written informed consent. The children additionally gave their written assent at the beginning of the clinical assessment.

4.2.4 Sampling techniques

The quantitative section employed probability sampling techniques as designed for the EPICure2@11 Study. A sample of EPICure 2 families, based on neonatal care networks at birth, from two geographical areas of England were invited to take part in the study. As the assessments for the main project took place at schools, head teachers were requested to identify three children born full-term (≥ 37 weeks of gestation) of the same age (± 3 months) and gender from the same school class as the EP participant, to act as potential controls. From these, initially one was selected at random to participate and parental consent sought. Due to challenges in recruiting controls, later a protocol amendment was approved, and all three control children were invited to take part. Consequently, some index children had up to three controls. Where a head teacher was unwilling to help select and contact a control child, or when parents requested a home visit instead of the school assessment, the index parents were invited to identify a friend that could be approached. Control children were not recruited for children attending special educational needs (SEN) schools or units.

4.2.5 Recruitment

The EPICure2@11 Study had a research coordinator who was responsible for recruitment. She contacted families and schools via post, email, and telephone. The recruitment was ceased when the maximum number of contacts were made, and no new families consented to take part in the study.

4.2.6 Data collection and instruments

The quantitative data for this study were routinely collected as part of the main project and were mostly derived from the self-completion Parent Questionnaire. Child level of disability was assessed during the clinical school assessment. Each variable's definition, sources of data and methods of assessment are described in detail below.

Primary outcome measure: Parenting Stress

I chose the Parenting Stress Index Short Form 4th Edition (PSI-4-SF) (Abidin, 2012) as the primary outcome instrument. I decided on this tool as it would yield data comparable with previous studies among preterm born populations (Landsem et al., 2014; Polic et al., 2016; Schappin et al., 2013; Singer et al., 2007; 2010; Zerach et al., 2015) and due to its popularity it would be well-accepted, understood and readily available. The PSI-4-SF was included in the Parent Questionnaire.

The Parenting Stress Index (PSI) is a well-validated questionnaire assessing the level of stress in the parent-child system (Abidin, 1992). The PSI-4-SF is a 36-item self-report questionnaire divided into three 12-item domains of Parental Distress (PD), Parent-Child Dysfunctional Interaction (P-CDI) and Difficult Child (DC), measured using a 5-point Likert scale from “strongly agree” to “strongly disagree” with higher scores indicating more stress. The PD domain assesses stressors associated with personal factors directly related to parenting such as depression, satisfaction in partner relationship and/or social support. The P-CDI domain assesses the extent to which the interaction with the child is challenging and the extent to which the parent perceives that their expectations of the child are not met. The DC domain assesses the child’s behavioural factors that affect the parent-child relationship. The three domains are summed up to a Total Stress (TS) score. Additionally, Defensive Responding (DR) is measured to indicate the extent to which the parent attempts to portray a favourable picture of themselves or to minimise problems or stressors (Abidin, 2012). The TS score was used as a primary outcome measure indicating overall parenting stress in the quantitative section of this study.

The PSI-4-SF is an abbreviated version of a full 101-item Parenting Stress Index (PSI). The PSI has a well-established validity and reliability and the two instruments have been shown to correlate highly ($r=0.98$ for TS; $r=0.94$ for PD; $r=0.95$ for DC; $r=0.91$ and 0.82 for P-CDI (derived from 2 subscales) domains, respectively). In the PSI-4-SF all domain specific coefficient alphas have been reported to be near 0.90. Test-retest reliability has been assessed over a 6-month retest interval with test-retest coefficients of 0.84 for TS, 0.85 for PD, 0.68 for P-CDI and 0.78 for DC domains, respectively (Abidin, 2012).

In this study I scored the PSI-4-SF following the standardised scoring. First, the seven items to assess DR were summed up. Then, the domain scores were calculated by summing up the 12 domain items. Scores for each domain were combined to obtain the TS score. Missing items were calculated as an average of the items within the subscale. If more than one item was

missing in one domain, the scores were not calculated. Internal consistency (Cronbach's alpha) was high in each domain and TS by the groups of parents (PD 0.91 and 0.87; P-CDI 0.87 and 0.89; DC 0.92 and 0.88; TS 0.96 and 0.94; DR 0.86 and 0.80 for parents of EP and FT born children, respectively). The raw scores were interpreted by converting them to percentiles provided as normative metrics. As diagnostic criteria, generally the normal range for scores is within the 16th and 84th percentiles. A DR score of 10 or lower indicates defensive responding. Scores at the 91th percentile are considered as clinically significant levels of parenting stress (Abidin, 2012).

Secondary outcome measure: Health-related quality of life

The Short Form Health Survey (SF-12v1) is a 12-item health-related quality of life (HRQoL) questionnaire created by the Medical Outcome Study to assess perceived general physical (PCS) and mental (MCS) health (Ware, Kosinski, & Keller, 1998). The shorter 12-item version was developed based on a longer 36-item questionnaire to produce a more respondent friendly HRQoL measure. The original PCS-36 and MCS-36 scores were derived using principal component analyses on data from the general US population. The standard scoring for the SF-12v1 was regressed from those original PCS-36 and MCS-36 scores and transformed to have a mean of 50 and standard deviation of 10, in the general US population (Hagell, Westergren, & Arestedt, 2017; Ware et al., 1998). The PCS and MCS scores have been found to have a strong correlation between the SF-12v1 and the SF-36 questionnaires, and the results have been successfully reproduced in many countries and study populations (Gandek et al., 1998; Gonzalez-Chica et al., 2017; Hagell et al., 2017; Jenkinson & Layte, 1997).

The SF-12v1 evaluates quality of life in the past four weeks. Both the PCS-12 and MCS-12 composite scores range from 0 to 100; higher scores indicating better HRQoL. The standardised scoring for SF-12v1 allows direct comparisons to general populations, indicating that scores above 50 are above the average, and a 1-point score increase represents a 10th of 1 SD. In this study, the PCS-12 and MCS-12 component scores were derived using the standardised scoring (Ware et al., 1998). Firstly, four questions were re-scored so that in all items higher scores indicated better health. Then indicator variables were created using the responses to each question, and finally they were combined using the proposed coefficients to generate final physical (PCS-12) and mental (MCS-12) health composite scores (Ware et al., 1998). Internal consistency (Cronbach's alpha) by the groups of parents was high (0.87 and 0.78 for parents of EP and FT born children, respectively).

Demographic and child health data collected

Parent characteristics

Parent demographic data collected included: Parent Questionnaire respondent (mother/father/carer, foster parent, grandparent or friend); parent age (continuous); parent ethnicity (White/Mixed or Multiple ethnic groups/Asian or Asian British/Black, African, Caribbean or Black British/Other ethnic group); employment status (employed, self-employed, full-time student/retired, semi-retired, long-term illness/homemaker, carer/unemployed); marital status (married, living with a partner/separated, divorced, widowed/single); living with the father or mother of the study child (yes/no); number of siblings study child had (continuous); multiple birth (yes/no; (data available for index parents only)); highest academic qualification (university degree/some post-secondary education/secondary education or less); receiving benefits (family benefits, income support, tax credits/incapability and disability benefits) and whether the family received more than one type of benefit (yes/no).

The Index of Multiple Deprivation

To provide an additional indication of families' socio-economic status (SES), the Office for National Statistics Index of Multiple Deprivation (IMD) 2019 scores (Ministry of Housing Communities and Local Government, 2019) were obtained for families resident in England using the child's postcode of residence at the time of the clinical assessment. The IMD is a relative small-area measure of deprivation in England, derived from seven domains: income, employment, education, skills and training, health and disability, crime, barriers to housing and services, and living environment. The IMD is indicated as deciles based on the English population, lowest decile (1) being most deprived and the highest decile (10) least deprived (Ministry of Housing Communities and Local Government, 2019).

Child characteristics

Child demographic data collected included: sex (male/female); age (continuous); school type (mainstream/SEN school or unit/home educated).

Child neurodevelopmental disability

Child neurodevelopmental disability (none, mild/moderate/severe) included four domains of IQ, vision, hearing, and gross-motor functioning. A Mental Processing Index (MPI) score (*Mean* 100; *SD* 15) derived from the Kaufman Assessment Battery for Children, 2nd edition (KABC-II) was used to assess child IQ together with clinical assessment of vision, hearing, and motor function. Moderate neurodevelopmental disability was defined as a child having any of the following: MPI score 2 to 3 *SD* below the mean of classmates (MPI 78-67), impaired but appearing to have useful vision, hearing loss corrected with aids or some hearing impairment but loss not corrected with aids, a score of 2 on the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997), and/or the Manual Ability Classification System (MACS) (Eliasson et al., 2006). Neurodevelopmental disability was severe if the child had any of the following: MPI score more than 3 *SD* below the mean of classmates (MPI <67), GMFCS or MACS score ≥ 3 , no useful hearing with aids, and/or no useful vision or sees light or gross movement only. If the child was unable to complete the psychology assessment due to severe cognitive impairment a nominal MPI score of 42 was assigned.

4.2.7 Data analysis

The Total Stress (TS) score indicating overall parenting stress was used as the primary outcome measure. I hypothesised that potential differences in the levels of parenting stress between the groups of parents (or within the EP parents) could largely be attributed to the level of care giving that the child required. The hypothesis was formed based on findings from both the systematic review and impressions from the qualitative interviews. In many of the reviewed studies, increased parenting stress diminished when families with children with neurodevelopmental disabilities were excluded from the analyses. This suggested that the care burden of a child with complex care needs contributed towards the increased parenting stress, rather than the child's birth status per se. The analysis of the qualitative data further strengthened these findings, showing that many of the interviewed parents contributed the potential difficulties they described to the extensive caregiving efforts their children's special needs required.

Descriptive statistics were used to present both parent and child demographic characteristics and health factors based on the child's birth status. Unadjusted median and mean scores, and proportions of parents scoring above cut-offs for each stress domain (cut-offs defined as in the PSI-4-SF instrument manual) by birth status were presented. Medians together with interquartile ranges were used to present the unadjusted PSI results to indicate positive skewness of the data. As per the instrument manual, the SF-12v1 unadjusted data were presented as means (SD). Univariate analyses were performed to assess potential group differences between the EP parents and FT parents using Chi Square Test on categorical variables and Welch's t-Test on continuous variables, as appropriate.

Following the univariate analyses, multiple linear regression analyses for each stress domain, the Total Stress and HRQoL measures (PCS-12 and MCS-12) were performed to assess overall group differences. Group comparisons between EP and FT parents were performed for each outcome measure adjusting for child's birth status, child male sex, child age (categorical), parent age and IMD decile at the time of the clinical assessment. Selection of covariates was guided by previous research presented in the systematic review above. Previous scientific findings have indicated that both parent education level and ethnicity might be associated with parents' experiences of increased stress (Franco, Pottick, & Huang, 2010; Holditch-Davis et al., 2009; Singer et al., 2007; 2010). However, as the IMD decile included a consideration of area level education, and the three variables (ethnicity, education and IMD) were found to have a level of co-linearity between them, parent ethnicity and education level were not considered as covariates. Fewer covariates in the adjusted analyses prevented overfitting the models (Lever, Krzywinski, & Altman, 2016).

In the adjusted analyses, child age was dichotomised and included in the models to indicate whether the child's age was below the median age of all assessed children in the study. Due to the narrow range, the child age as a continuous variable did not function meaningfully in the interaction analyses. All other variables were defined as described above. Group differences were presented as adjusted differences in means with 95% Confidence Intervals (*CI*) and a *p*-value of ≤ 0.05 was considered significant. Bonferroni correction was applied as appropriate. The group differences were tested by excluding the parents with children with neurodevelopmental disability (moderate or severe) and/or multiple births (EP participants only).

To further explore the hypothesis of the potential impact of extensive caregiving efforts on parenting stress among the EP parents, logistic regression analyses were used. A Parenting Stress Index score above the 84th centile was considered high as per standard scoring (Abidin,

2012). Univariate analyses were performed on categorical variables to assess the potential association between high parenting stress and sociodemographic and health factors, using Chi Square Test or Fisher's Exact Test, as appropriate. To avoid small (<10) and zero cell counts, variables were dichotomised when needed (child neurodevelopmental disability: none, mild / moderate, severe; school type: mainstream, home educated / SEN school or unit; parent work status: employed, self-employed, full-time student / retired, semi-retired, long-term illness, homemaker, carer; parent education: below university level / university degree). All other variables were defined as described above. Potential associations between high parenting stress and continuous variables were assessed using Welch's t-Test. Odds ratios together with 95% CI were presented and a *p*-value of ≤ 0.05 was considered significant. In the multivariable analyses, the odds ratios were adjusted for all other variables associated with high stress in the univariate analyses ($p \leq 0.10$).

Descriptive statistics were presented on all parents who had returned their Parent Questionnaires ($n=300$) and whose data were available. Whereas, those parents who had returned their questionnaires, but whose PSI-4-SF Total Stress data were missing or incomplete ($n=9/300$), whose child was not assessed ($n=3/300$) or were resident outside of England at the time of the re-assessment ($n=10/300$) were excluded from the adjusted analyses ($n=21/300$) (1 parent PQ only and resident outside of England)) due to the unavailability of complete data. An analysis was performed to compare the characteristics of those parents whose data were complete and whose data were missing or incomplete.

A further analysis was performed between the families who returned their Parent Questionnaires and were known to be the biological mother of the cohort child ($n=171/175$ (4 index children known to be adopted or in foster care), and the mothers in the remaining cohort whose EPICure 2 cohort children were discharged alive from hospital but who were not invited to take part in the re-assessment at 11 years, who declined to participate or who consented to take part but failed to return their questionnaires ($n=866$)). The proportion of male infants, rates of multiple births, average IMD decile at birth based on maternal postcode, and mother's ethnicity and age at birth were compared.

All the analyses were performed using the statistical software STATA, version 15.1 (StataCorp, Texas, USA).

4.3 Respondents

Two hundred and twenty parents of children in the EPICure 2 cohort consented to take part in the re-assessment at their child's age of 11 years; 45.6% of the 482 families invited to participate, and 21.1% (220/1041) of the total cohort of EP infants discharged alive from hospital in 2006. Of those 220 consented parents, 175 (79.5%) returned their Parent Questionnaires. Two hundred EP children underwent the clinical assessment. Consequently, there were 20 consented parents whose children were not seen, of whom three returned their Parent Questionnaires. Reasons why the 20 consented children were not clinically assessed are presented in Appendix C Supplementary Table 1. For the 220 EP children, 143 control children were recruited and assessed, and 125 (87.4%) of their parents returned their Parent Questionnaires (see Table 4-1 below).

Demographic and unadjusted parenting stress data are presented for all parents who returned their questionnaires regardless of their children's assessment status. Data on parents whose parenting stress data were incomplete or missing from the returned questionnaire, whose children were not assessed or/and who were resident outside of England were excluded from the adjusted analyses. A comparative analysis of demographic characteristics between parents whose data were available and missing or incomplete is presented in Appendix D Supplementary Table 2. In comparison to the parents with complete data, the parents with missing or incomplete variables more often had children with moderate or severe neurodevelopmental disabilities (parents with complete data: 21.5% and parents with incomplete/missing data: 38.9%, respectively), they were more often recipients of incapability or disability benefits (21.9% and 42.9%, respectively), and of more than one type of benefit (15.4% and 42.9%, respectively). They were more commonly either separated, divorced, widowed or single (20.2% and 33.4%, respectively). Other variables were comparable ($p > 0.05$) (Appendix D Supplementary Table 2).

Table 4-1 Parent and child characteristics by child's birth status

	EP	Control	<i>p</i> value ¹
<i>Discharged alive from hospital</i>	1041		
<i>Invited to participate at 11 years</i>	482		
<i>Consented parents</i>	220	143	
<i>Child assessment completed</i>	200	143	
<i>Parent Questionnaires returned</i>	175 (86.2)	125 (87.4)	0.74
<i>Parent questionnaires returned and child assessment completed</i>	172 (86.0)	125 (87.4)	0.71
Parent Characteristics	EP (n=175)	Control (n=125)	<i>p</i> value¹
Respondent (n=292)			
Mother	156 (91.2)	109 (90.1)	
Father	12 (7.0)	11 (9.1)	0.66
Carer/Foster parent/Grandparent/Friend	3 (1.8)	1 (0.8)	
Age, median (IQR) (n=290)	45.9 (42.0, 49.8)	46.6 (42.3, 49.1)	0.74
Ethnicity (n=300)			
White	113 (64.5)	101 (80.8)	
Mixed/Multiple ethnic groups	8 (4.6)	1 (0.8)	
Asian/Asian British	26 (14.9)	14 (11.2)	0.02
Black/African/Caribbean/Black British	23 (13.1)	6 (4.8)	
Other ethnic groups	5 (2.9)	3 (2.4)	
Employment status (n=297)			
Employed/Self-employed/Full-time student	134 (77.0)	101 (82.1)	
Retired/Semi-retired/Long-term illness	4 (2.3)	2 (1.6)	0.61
Homemaker/Carer	25 (14.4)	16 (13.0)	
Unemployed	11 (6.3)	4 (3.3)	
Marital status (n=299)			
Married/Living with a partner	137 (78.7)	99 (79.2)	
Separated/Divorced/Widowed	15 (8.6)	17 (13.6)	0.15
Single	22 (12.6)	9 (7.2)	
Living with the father or mother of the study child (n=299)	128 (73.6)	95 (76.0)	0.63
Multiple birth (n=175)	43 (24.6)	-	-
Highest academic qualification (n=277)			
University degree	70 (43.5)	55 (47.4)	
Some post-secondary education	45 (28.0)	30 (25.9)	0.81
Secondary education or less	46 (28.6)	31 (26.7)	

Table 4-1 (continued)

	EP	Control	<i>p</i> value ¹
Receiving benefits (n=300)			
Family benefits, income support, tax credits	106 (60.6)	60 (48.0)	0.03
Incapability and disability benefits	62 (35.4)	8 (6.4)	<0.001
Receiving more than one type of benefit (n=300)			
	48 (27.4)	4 (3.2)	<0.001
IMD at 11 years, mean (SD) (n=290)²			
	5.6 (2.8)	5.6 (2.9)	0.90
IMD at delivery, mean (SD) (n=173)³			
	4.7 (2.7)	n/a	
Child characteristics⁴			
Male sex (n=297)			
	90 (52.3)	55 (44.0)	0.16
Age at the assessment, mean (SD) (n=297)			
	11.8 (0.6)	11.7 (0.6)	0.26
Gestational age in weeks, median (range) (n=172)			
	25.7 (23.0-26.9)	n/a	
Number of siblings, mean (SD) (n=297)			
	1.2 (0.9)	1.3 (1.0)	0.16
School type (n=296)			
Mainstream	148 (86.0)	124 (100.0)	
SEN school or unit	21 (12.2)		n/a
Home educated	3 (1.7)		
Overall neurodevelopmental disability (n=297)			
None or mild	109 (63.4)	121 (96.8)	
Moderate	33 (19.2)	4 (3.2)	n/a
Severe	30 (17.4)	0 (0)	

Data are presented in n (%) unless otherwise stated

Note: Missing data accounts for differing totals

¹Chi Square Test for categorical variables and Welch's t-Test for continuous variables were used

²Those resident in England at the time of the re-assessment at 11 years (n=169 and n=121, respectively)

³Those who returned Parent Questionnaires at 11 years (2 missing postcodes at birth)

⁴Children who were assessed, and Parent Questionnaires were returned (n=172 and n=125, respectively)

The proportion of male infants ($X^2(1) = 0.50, p = 0.48$) and rates of multiple births ($X^2(1) = 0.10, p = 0.75$) did not differ between the biological parents who completed their Parent Questionnaires at 11 years (n=171) and mothers in the remaining cohort whose EPICure 2 cohort children were discharged alive from hospital, but were not invited to take part in the re-assessment at 11 years, or who consented to take part but failed to complete their questionnaires (n=866). However, the participating parents were of older ages ($t(1035) = 4.70, p < 0.001$) and had on average higher IMD deciles at birth based on maternal postcode at the time of the delivery compared to the non-participants, $t(1017) = 2.14, p = 0.03$. In addition, ethnic backgrounds of participants and non-participants differed; mothers taking part were more often from Asian ethnic backgrounds (16.4% and 11.7%, respectively), whereas non-participants were more often from Black ethnic groups (19.9% and 13.5%, respectively), $X^2(4) = 9.42, p = 0.05$ (Appendix E Supplementary Table 3).

4.3.1 Parent characteristics

A majority of the questionnaire respondents among both groups of parents were mothers (EP parents 91.2% and FT parents 90.1%, respectively). Parents of EP YA and FT YA were of similar ages (*Median*: 45.9 years, (*IQR*: 42.0, 49.8) and *Median*: 46.6 years, (*IQR*: 42.3, 49.1), respectively), the majority of them were married or living with a partner (approximately 79% among both groups) and were either in employment, self-employed or full-time students (77.0% and 82.1%, respectively), although information on hours in paid employment (i.e., full-time/part-time) were not collected. Nearly half of the participating parents had a university degree (45.1%; 125/277). The groups of parents did not differ in the number of other children (study children had a median of one sibling among both groups of parents), and a majority of them were living with the father or mother of the study child (EP parents 73.6% and FT parents 76.0%, respectively). Parents of YA born EP were however from more diverse ethnic backgrounds ($X^2(4) = 12.19, p = 0.02$), and were more commonly receiving one or more type of benefit ($X^2(1) = 29.87, p < 0.001$). The average IMD deciles based on the child's postcode at the time of the re-assessment amongst the groups of parents were comparable, $t(288) = 0.13, p = 0.90$ (Table 4-1).

4.3.2 Child characteristics

YA born EP and FT who underwent the clinical child assessment and whose parents returned their Parent Questionnaires were of similar ages ($M = 11.8, SD = 0.6$ and $M = 11.7, SD = 0.6$, respectively) and similar proportions of them were male (52.3% and 44.0%, respectively). As per recruitment criteria, larger proportions of YA born EP attended SEN schools or units at the time of the assessment and had neurodevelopmental disabilities (Table 4-1).

4.4 Results

4.4.1 Parenting stress by child's birth status

Parents of YA born EP reported higher levels of parenting stress on all stress domains compared to the parents of YA born FT, as presented in Table 4-2 below. The highest median domain scores were reported among the EP parents in the Difficult Child domain, reflecting that the parents of EP YAs found their children's behavioural challenges to have a negative impact on their interaction with their child (Abidin, 2012).

Table 4-2 Median and quartile Parenting Stress Index (PSI-4-SF) scores by child's birth status

Stress Domain	EP	n/N ¹	Control	n/N ¹	<i>p</i> value
Total Stress	71 (53, 92)	167/175	55 (45, 71)	124/125	<0.001
Parental Distress	24 (16, 30)	171/175	21 (16, 25)	124/125	0.04 ²
Dysfunctional Interaction	23 (16, 29)	170/175	17 (13, 23)	124/125	<0.001 ²
Difficult Child	25 (18, 34)	171/175	19 (14, 24)	124/125	<0.001 ²

Data are presented as median (interquartile range)

Note: Total Stress score above 109 (>84th percentile) indicates high stress (Abidin, 2012)

¹Those parents with complete data / all parents who returned their Parent Questionnaires at 11 years

²Mann-Whitney Test with Bonferroni correction

4.4.2 HRQoL by child's birth status

Of the 300 returned Parent Questionnaires, 287 (95.7%) parents had fully completed the SF-12v1 instrument; 168 (96.0%) EP parents and 119 (95.2%) control parents. On average, both groups of parents reported similar levels of physical and mental HRQoL (physical health $t(285) = 1.56$, $p = 0.12$ and mental health $t(285) = 1.40$, $p = 0.16$ (Table 4-3).

Table 4-3 Parent reported health-related quality of life (SF-12v1) by child's birth status

Health Domain	EP	n/N ¹	Control	n/N ¹	<i>p</i> value ²
Physical health (PCS-12)	50.7 (9.3, 49.3-52.1)	168/175	52.3 (8.0, 50.9-53.8)	119/125	0.12
Mental health (MCS-12)	49.2 (9.4, 47.8-50.7)	168/175	50.7 (8.1, 49.2-52.2)	119/125	0.16

Data are presented in mean (SD, 95% Confidence Interval)

¹Those parents with complete data / all parents who returned their Parent Questionnaires at 11 years

²Welch's t-Test

4.4.3 Adjusted mean differences in parenting stress and HRQoL between the groups of parents

4.4.3.1 Total parenting stress

As per Table 4-4 below, the overall picture of parenting stress among the groups of parents was little affected by the effects of the covariates of child age and male sex, parent age, and the family's IMD decile at 11 years. In comparison with parents of FT born YA, on average EP parents reported 14.8 points higher total parenting stress ($t(265) = 5.19, p < 0.001$). Yet average stress scores among both groups of parents were within the normal range of parenting stress (Abidin, 2012). When excluding the parents of YAs with neurodevelopmental impairments and non-singletons, the difference in the overall stress levels, although smaller, remained similar between the groups of parents, $t(188) = 2.49, p = 0.04$ (Table 4-4).

Of the included covariates, child age was found to predict the total parenting stress differently within the groups of parents; among the parents of EP YAs younger child age predicted higher parenting stress ($\beta = -7.63, t(1, 162) = -2.05, p = 0.04, SE = 3.72$), whereas the opposite was true among the parents of FT YAs (although the correlation among the controls did not reach statistical significance, $\beta = 3.49, t(1, 122) = 1.38, p = 0.17, SE = 2.52$). Consequently, a child age and birth status interaction effect was introduced into the adjusted model, improving the fit of the model significantly, $X^2(1) = 5.39, p = 0.02$. Regardless of the increased total parenting stress among the EP parents, when adjusting for the other covariates, those EP parents whose children were older than the median age of all assessed children (*Median* 11.8 years (*IQR*: 11.3, 12.2)) reported on average 13.2 points lower total parenting stress than those EP parents whose children were of younger ages, $\beta = -13.22, t(6, 263) = -2.30, p = 0.02, SE = 5.74$. This suggested that the increased parenting stress among the EP parents was more commonly experienced among the parents of younger children in the study.

Since the age range of children in the study was narrow, I examined whether other factors associated with child age, such as child's transition to secondary education, provoked higher parenting stress among the EP parents. Sixty-five children born EP underwent the clinical assessment when they were in primary school, whereas 99 EP YAs had started secondary education at the time of the assessment. Figure 4-1 below shows a scatter plot of total parenting stress scores by child's age among the groups of EP parents pre and post child's transition to secondary education. The figure suggests that although decreasing parenting stress with increasing child age was an overall trend among EP parents, similar proportions of parents reported high levels of parenting stress pre and post transition.

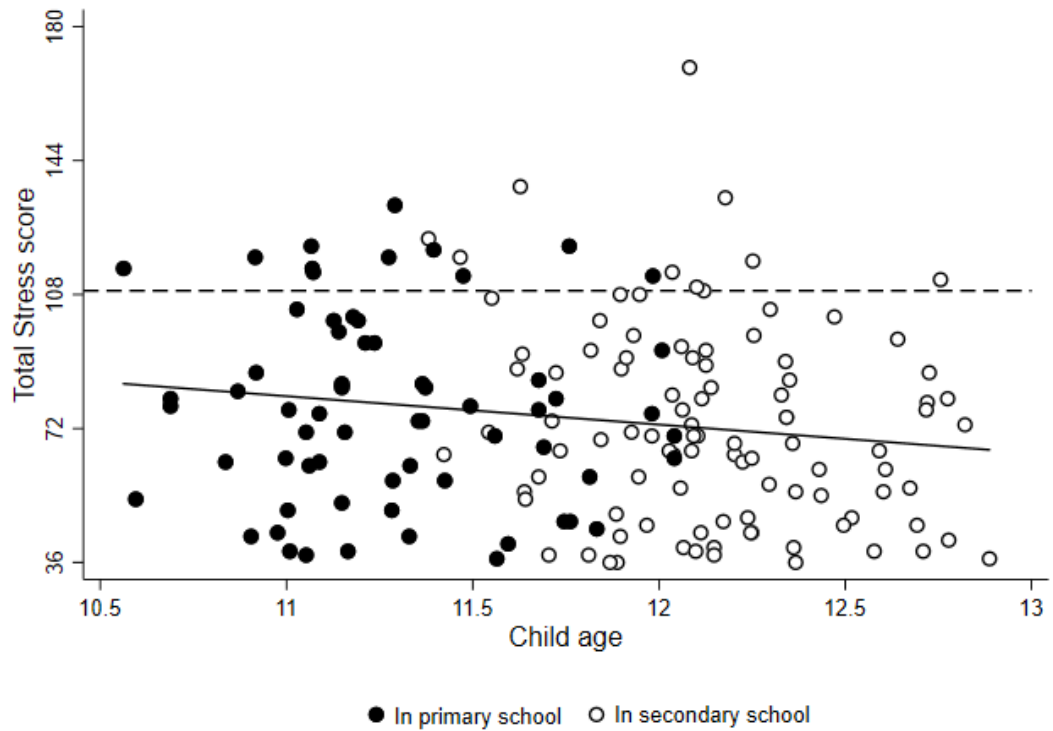


Figure 4-1 Scatter plot comparing PSI-4-SF Total Stress scores by child’s age among EP parents whose children were assessed in primary school (n=65) and those who had started secondary school (n=99)

The black line represents linear fitted values for total parenting stress by child’s age. The dashed line represents the cut-off for high parenting stress (>84th centile).

Table 4-4 Adjusted mean group differences in average PSI-4-SF and SF-12v1 scores between the groups of parents

	n/N ¹	Unadjusted sample mean	(95% CI)	Adjusted ² sample mean	(95% CI)	Adjusted ² difference in means	(95% CI)	p value ³
PSI-4-SF								
Total Stress								
EP	158/175	73.8	(69.6 – 77.9)	74.0	(70.4 – 77.6)	14.8	(9.2 – 20.4)	<0.001
Control	112/125	58.6	(55.4 – 61.8)	59.2	(54.9 – 63.5)			
Parental Distress								
EP	161/175	24.1	(22.6 – 25.6)	24.0	(22.7 – 25.4)	3.0	(0.9 – 5.2)	0.02
Control	112/125	20.9	(19.6 – 22.2)	21.0	(19.4 – 22.6)			
Parent-child Dysfunctional Interaction								
EP	161/175	23.3	(22.0 – 24.7)	23.4	(22.2 – 24.6)	4.8	(2.9 – 6.7)	<0.001
Control	112/125	18.3	(17.2 – 19.4)	18.6	(17.1 – 20.0)			
Difficult Child								
EP	161/175	26.4	(24.7 – 28.0)	26.5	(25.1 – 27.9)	7.0	(4.8 – 9.2)	<0.001
Control	112/125	19.4	(18.2 – 20.6)	19.5	(17.8 – 21.2)			
PSI-4-SF: Restricted to singletons with no/mild impairment								
Total Stress								
EP	81/85	65.9	(60.4 – 71.3)	66.4	(61.8 – 71.0)	7.7	(1.6 – 13.7)	0.04
Control	109/121	58.5	(55.2 – 61.7)	58.7	(54.7 – 62.6)			
Parental Distress								
EP	82/85	21.8	(19.8 – 23.8)	21.9	(20.1 – 23.7)	1.1	(-1.3 – 3.4)	>0.999
Control	109/121	20.9	(19.5 – 22.3)	20.8	(19.3 – 22.4)			

Table 4-4 (continued)

	n/N ¹	Unadjusted sample mean	(95% CI)	Adjusted ² sample mean	(95% CI)	Adjusted ² difference in means	(95% CI)	p value ³
PSI-4-SF: Restricted to singletons with no/mild impairment								
Parent-child Dysfunctional Interaction								
EP	81/85	20.9	(19.0 – 22.8)	21.0	(19.3 – 22.6)	2.5	(0.4 – 4.7)	0.06
Control	109/121	18.3	(17.1 – 19.4)	18.4	(17.0 – 19.8)			
Difficult Child								
EP	82/85	23.3	(21.1 – 25.4)	23.5	(21.8 – 25.3)	4.2	(1.9 – 6.5)	<0.001
Control	109/121	19.3	(18.1 – 20.5)	19.3	(17.8 – 20.8)			
SF-12v1								
Physical Component Summary score (PCS-12)								
EP	158/175	50.7	(49.2 – 52.1)	51.0	(49.4 – 52.1)	-1.5	(-3.6 – 0.6)	0.16
Control	107/125	52.4	(50.8 – 53.9)	52.3	(50.6 – 53.9)			
Mental Component Summary score (MCS-12)								
EP	158/175	49.5	(48.1 – 51.0)	49.4	(48.1 – 50.7)	-1.0	(-3.1 – 1.2)	0.38
Control	107/125	50.7	(49.2 – 52.2)	50.3	(48.7 – 52.0)			
SF-12v1: Restricted to singletons with no/mild impairment								
Physical Component Summary score (PCS-12)								
EP	79/85	50.7	(48.4 – 53.0)	50.5	(48.5 – 52.4)	-2.0	(-4.6 – 0.6)	0.13
Control	104/121	52.3	(50.7 – 53.9)	52.5	(50.8 – 54.2)			
Mental Component Summary score (MCS-12)								
EP	79/85	52.1	(50.4 – 53.8)	51.9	(50.2 – 53.6)	1.6	(-0.7 – 3.8)	0.17
Control	104/121	50.8	(49.2 – 52.3)	50.4	(48.9 – 51.8)			

¹Those parents with complete set of data / all parents who returned their Parent Questionnaires at 11 years

²Adjusted for child age (categorical), child male sex, parent age, the Index of Multiple Deprivation decile at 11 years and interaction of child age and birth status if present

³Multiple linear regression with Bonferroni correction (Total Stress not corrected)

4.4.3.2 Parental distress

After adjusting for the covariates, the average Parental Distress domain scores differed by the groups of parents indicating that the parents of EP YAs experienced more distress than the parents of YAs born FT (Table 4-4). However, when parents of YAs with moderate or severe neurodevelopmental morbidity and multiple births were excluded from the analysis, the difference between the groups of parents diminished, $t(189) = 0.88, p = >0.999$. Contrary to Total Stress, child age did not predict Parental Distress differently among the groups of parents, $X^2(1) = 1.90, p = 0.17$. This suggested that the potential personal distress that the parent may have experienced in their parenting role did not vary due to the child's age or birth status but was influenced by the presence of child disability or/and multiple births. No other interactions were identified.

4.4.3.3 Parent-child dysfunctional interaction

In comparison with parents of FT born YAs, EP parents reported more challenges in the parent-child interaction. After adjusting for covariates, EP parents on average obtained 4.8 points higher scores in this domain than the control parents, $t(271) = 5.03, p < 0.001$. After excluding the parents of YAs with long-term morbidities and multiple status, the difference between the groups of parents was reduced to 2.5 points on average, $t(188) = 2.34, p < 0.06$ (Table 4-4). Similar to Total Stress, the parents of older EP participants reported lower levels of increased parenting stress within the P-CDI scale than the EP parents of younger children, $\beta = -4.66, t(6, 266) = -2.43, p = 0.02, SE = 1.92$.

4.4.3.4 Difficult child

Even after adjusting for the covariates and the interaction effect of birth status and child age, the average difference in reported parenting stress between the EP and control parents was the largest in Difficult Child sub-scale (7.0 points, 95% $CI = 4.8, 9.2, p < 0.001$) (Table 4-4). This suggested that the parents of EP YAs found their child's behavioural factors most stressful in their parenting. When assessed only among the parents of morbidity-free singleton children the difference between the groups of parents persisted, $t(188) = 3.58, p < 0.001$.

4.4.3.5 Mental HRQoL

After considering the effects of covariates and the interaction of birth status and child age, the parents of YAs born EP and FT perceived their mental HRQoL similarly, $t(263) = -0.89, p = 0.38$. When assessed separately and only with the effect of child age (continuous), in line

with the PSI-4-SF, the EP parents perceived their mental HRQoL to improve with increasing child age ($\beta = 3.02$, $t(1, 163) = 2.27$, $p = 0.03$, $SE = 1.33$), whereas parents of FT born YAs reported decreasing mental HRQoL with increasing child age ($\beta = -3.14$, $t(1, 117) = -2.59$, $p = 0.01$, $SE = 1.21$). Interestingly, after excluding the parents of YAs with neurodevelopmental morbidities and/or multiple statuses, the EP parents reported better mental HRQoL compared to the control parents, but this difference did not reach statistical significance, $t(181) = 1.38$, $p = 0.17$ (Table 4-4).

4.4.3.6 Physical HRQoL

As with the mental HRQoL, both groups of parents perceived their physical health similarly, $t(263) = -1.47$, $p = 0.16$. The group ratings remained similar when only parents of singleton YAs with no neurodevelopmental morbidities were included (Table 4-4). In the fully adjusted model ($F(6, 258) = 3.85$, $p = 0.001$) rather than the child characteristics, such as age or birth status, the increased parent age and lower average IMD decile predicted poorer physical health; parent age, $\beta = -0.32$, $t(6, 258) = -3.56$, $p < 0.001$, $SE = 0.09$; IMD decile, $\beta = 0.39$, $t(6, 258) = 2.03$, $p = 0.04$, $SE = 0.19$.

4.4.4 Proportion of parents reporting high parenting stress by child's birth status

Twenty EP parents (12.0%) reported high total parenting stress (>84th centile), compared to none among controls $X^2(1) = 15.95$, $p < 0.001$ (Table 4-5). Similar statistically significant differences were seen in the three subscales after Bonferroni corrections and EP and control parents reported similarly on the Defensive Reporting scale, although there was a trend to lower scores among the FT parents (Table 4-5).

Table 4-5 Proportion of parents reporting high parenting stress (PSI-4-SF) by stress domain and child's birth status

Stress Domain	EP		Control		<i>p</i> value
Total Stress	20/167	(12%)	0/124	(-)	<0.001
Parental Distress	20/171	(12%)	2/124	(1.6%)	0.003¹
Dysfunctional Interaction	23/170	(14%)	3/124	(2%)	0.003¹
Difficult Child	30/171	(18%)	3/124	(2%)	0.003¹
Defensive Responding ²	52/172	(30%)	47/124	(38%)	0.17

Data are presented in n (%)

¹Fisher's Exact Test with Bonferroni correction

²Score ≤ 10 is suggestive of defensive responding (Abidin, 2012)

4.4.5 Factors associated with high parenting stress among EP parents

4.4.5.1 Total parenting stress

On univariate analyses there were no significant associations between high total parenting stress and parent sociodemographic characteristics (age; ethnicity; education level; employment status; marital status; living with the other birth parent; number of other children; average IMD decile at 11 years or at birth). Poorer HRQoL, in terms of physical ($t(157) = 2.23$, $p = 0.04$) and mental health ($t(157) = 4.69$, $p < 0.001$), and receipt of income support ($X^2(1) = 4.10$, $p = 0.04$) were associated with high total parenting stress scores. From the assessed child characteristics, neurodevelopmental impairment, and attendance at Special Educational Needs (SEN) school or unit were both associated with high parenting stress (disability: $X^2(2) = 10.62$, $p = 0.007$ and SEN school: $X^2(2) = 12.42$, $p = 0.006$, respectively),

and a younger child age had a borderline association with high parenting stress ($t(162) = 1.9187, p = 0.07$).

Further investigations were undertaken to assess whether co-morbidities segregated together.

Benefits and child disability - Those EP parents who reported high total parenting stress were also more often receiving incapability and disability benefits ($X^2(1) = 12.48, p < 0.001$) and were recipients of more than one type of benefit ($X^2(1) = 9.75, p = 0.002$). As child neurodevelopmental disability and parent receiving disability benefits were interlinked, an analysis was conducted to assess whether the association between receiving incapability and disability benefits and high parenting stress was driven by child morbidity. A majority of families with children with severe disabilities received disability benefits (93.3%, 28/30). Sixteen families whose children had moderate disabilities did not receive benefits (16/33, 48.5%), but all these parents reported low levels of parenting stress. Of those 17 (15.6%) families whose children were morbidity-free, but received incapability and disability benefits, only two parents reported high levels of stress. Consequently, odds ratios presented in Table 4-6 below were adjusted for child neurodevelopmental disability, but not whether the parent was receiving incapability/disability benefits. Parent receiving income support benefits was included in the adjusted analyses as it was not directly linked to child morbidity.

Parental mental health - Among the EP parents, those respondents who reported high overall parenting stress reported significantly lower levels of mental HRQoL than those parents with lower stress scores ($M = 37.5, SD = 12.4$ and $M = 51.2, SD = 7.4$, respectively, $t(157) = 4.69, p < 0.001$). Consequently, in the multivariate model parent mental health scores may have swamped potential effects of other variables. Therefore, two separate models were built and in Table 4-6 the associated adjusted odds ratios are reported for both models; 1) odds ratios adjusted for all other variables associated with high parenting stress in univariate analyses ($p < 0.10$), except HRQoL and 2) odds ratios adjusted for all other variables associated with high parenting stress in univariate analyses ($p < 0.10$), including HRQoL.

Logistic regression analysis - In model 1 (excluding HRQoL) the associations between high Total Stress scores with both receipt of income support and child moderate/severe impairment diminished ($aOR = 2.5, 95\% CI: 0.7, 8.1, p = 0.14$ and $aOR = 1.6, 95\% CI: 0.5, 5.1, p = 0.43$, respectively) and the child's age became borderline significant ($aOR = 0.4, 95\% CI: 0.1, 1.0, p = 0.06$), whereas the odds of EP parents with children at SEN schools or units reporting high overall parenting stress compared to the parents whose children attended mainstream schools

or were home educated reduced but remained significant ($aOR = 4.1$, 95% CI : 1.1, 15.4, $p = 0.03$) (Table 4-6).

In model 2, the association between high parenting stress and SEN school or unit attendance diminished ($aOR = 2.7$, 95% CI : 0.5, 13.2, $p = 0.22$) but parental mental health was strongly associated with high parenting stress ($aOR = 0.9$, 95% CI : 0.8, 0.9, $p < 0.001$) (Table 4-6). Introducing an interaction effect of physical and mental health into the model did not improve the fit of the model significantly, $X^2(1) = 0.30$, $p = 0.58$. This was expected, as the SF-12v1 instrument has been built on orthogonal rotations, which reduce covariation between the domains (Hagell et al., 2017).

Table 4-6 Demographic and health variables associated with high parenting stress (PSI-4-SF) by stress domain among parents of EP YA: n, row %, unadjusted and multivariate logistic regression

Demographic and Health Variables	Proportion of parents reporting high stress	OR (CI 95%)		p value	1st model ¹		2nd model ²			
					aOR (CI 95%)	p value	aOR (CI 95%)	p value		
Total Stress	20 / 164 (12.2%)									
HRQoL³										
Physical health (PCS-12)	45.2 (11.9) / 51.5 (8.7)	0.9	(0.9 – 1.0)	0.01	–		1.0	(0.9 – 1.0)	0.07	
Mental health (MCS-12)	37.5 (12.4) / 51.2 (7.4)	0.9	(0.8 – 0.9)	<0.001	–		0.9	(0.8 – 0.9)	<0.001	
Receipt of income support benefits										
No	4 / 67 (6.0)	1.0			1.0					
Yes	16 / 97 (16.5)	3.1	(1.0 – 9.8)	0.05	2.5	(0.7 – 8.1)	0.14	1.8	(0.5 – 6.8)	0.40
Child age	11.6 (0.5) / 11.8 (0.5)	0.4	(0.2 – 1.0)	0.06	0.4	(0.1 – 1.0)	0.06	0.5	(0.2 – 1.7)	0.27
School type										
Mainstream or home educated	13 / 145 (9.0)	1.0			1.0					
SEN school or unit	7 / 19 (36.8)	5.9	(2.0 – 17.7)	0.001	4.1	(1.1 – 15.4)	0.03	2.7	(0.5 – 13.2)	0.22
Moderate/severe neurodevelopmental disability										
No	8 / 107 (7.5)	1.0			1.0					
Yes	12 / 57 (21.1)	3.3	(1.3 – 8.6)	0.02	1.6	(0.5 – 5.1)	0.43	0.7	(0.2 – 3.1)	0.65
Parental Distress	20 / 168 (11.9%)									
HRQoL										
Physical health (PCS-12)	42.2 (11.7) / 51.8 (8.4)	0.9	(0.9 – 1.0)	<0.001	–		0.9	(0.9 – 1.0)	0.02	
Mental health (MCS-12)	40.1 (11.2) / 50.6 (8.2)	0.9	(0.9 – 0.9)	<0.001	–		0.9	(0.9 – 1.0)	0.01	
Working status										
Employed/Self-employed/ Full-time student	11 / 129 (8.5)	1.0			1.0			1.0		
Retired/Semi-retired/Long-term illness/Homemaker/Carer	9 / 39 (23.1)	3.2	(1.2 – 8.5)	0.02	3.1	(0.9 – 10.8)	0.07	1.6	(0.3 – 7.3)	0.58
Education										
Below university level	5 / 90 (5.6)	1.0			1.0			1.0		
University degree	12 / 67 (17.9)	3.7	(1.2 – 11.1)	0.02	4.2	(1.2 – 14.7)	0.02	4.1	(1.0 – 17.6)	0.06

Table 4-6 (continued)

Demographic and Health Variables	Proportion of parents reporting high stress	OR (CI 95%)		<i>p</i> value	1st model ¹		<i>p</i> value	2nd model ²		<i>p</i> value
		aOR (CI 95%)	<i>p</i> value		aOR (CI 95%)	<i>p</i> value				
School type										
Mainstream or home educated	12 / 149 (8.1)	1.0			1.0			1.0		
SEN school or unit	8 / 19 (42.1)	8.3	(2.8 – 24.6)	<0.001	4.4	(1.1 – 17.3)	0.04	2.1	(0.4 – 11.1)	0.36
Moderate/severe neurodevelopmental disability										
No	5 / 109 (4.6)	1.0			1.0			1.0		
Yes	15 / 59 (25.4)	7.1	(2.4 – 20.7)	<0.001	5.6	(1.3 – 24.2)	0.02	4.2	(0.8 – 22.8)	0.10
Dysfunctional interaction										
23 / 167 (13.8%)										
HRQoL										
Mental health (MCS-12)	40.8 (13.6) / 50.7 (7.8)	0.9	(0.9 – 1.0)	<0.001	–			0.9	(0.9 – 1.0)	<0.001
Receipt of income support benefits										
No	5 / 67 (7.5)	1.0			1.0			1.0		
Yes	18 / 100 (18.0)	2.7	(1.0 – 7.7)	0.06	2.4	(0.8 – 7.0)	0.1	1.8	(0.6 – 5.8)	0.29
Moderate/severe neurodevelopmental disability										
No	11 / 107 (10.3)	1.0			1.0			1.0		
Yes	12 / 60 (20.0)	2.2	(0.9 – 5.3)	0.09	1.9	(0.8 – 4.6)	0.2	0.8	(0.3 – 2.4)	0.71
Difficult Child										
30 / 168 (17.9%)										
HRQoL										
Physical health (PCS-12)	46.4 (10.8) / 51.5 (8.9)	1.0	(0.9 – 1.0)	0.01	–			1.0	(0.9 – 1.0)	0.05
Mental health (MCS-12)	41.2 (11.5) / 51.2 (7.8)	0.9	(0.9 – 0.9)	<0.001	–			0.9	(0.9 – 1.0)	<0.001
Receipt of income support benefits										
No	5 / 67 (7.5)	1.0			1.0			1.0		
Yes	25 / 10 (24.8)	4.1	(1.5 – 11.3)	0.01	3.7	(1.3 – 10.6)	0.01	2.9	(1.0 – 9.0)	0.06
Child age										
	11.6 (0.5) / 11.8 (0.5)	0.5	(0.2 – 1.0)	0.04	0.4	(0.2 – 0.9)	0.03	0.5	(0.2 – 1.2)	0.13
School type										
Mainstream or home educated	22 / 148 (14.9)	1.0			1.0			1.0		
SEN school or unit	8 / 20 (20.0)	3.8	(1.4 – 10.4)	0.01	3.0	(0.9 – 10.2)	0.07	1.8	(0.5 – 7.1)	0.40
Moderate/severe neurodevelopmental disability										
No	15 / 109 (13.8)	1.0			1.0			1.0		
Yes	15 / 59 (25.4)	2.1	(1.0 – 4.8)	0.06	1.1	(0.4 – 3.0)	0.84	0.7	(0.2 – 2.2)	0.52

Note: High parenting stress defined as PSI-4-SF scores > 84th centile (Abidin, 2012)

¹1st model adjusted for all other variables associated with high parenting stress in univariate analyses (*p* <0.10) except HRQoL

²2nd model adjusted for all other variables associated with high parenting stress in univariate analyses (*p* <0.10) including HRQoL

³Reported as mean (SD) among high stress parents / mean (SD) among low stress parents

4.4.5.2 Parental distress

On univariate analyses, from the assessed parent characteristics, age, ethnicity, marital status, living with the other birth parent, receiving income support benefits, number of other children, average IMD decile at 11 years or at birth, and from the assessed child characteristics, age, sex, gestational age in weeks and multiple status were not associated with high parental distress. Of those parents who were not in employment, self-employed or full-time students, a larger proportion reported high parental distress compared to those who were working or studying outside the home (23.1% and 8.5%, respectively). Parental distress was also associated with university degree education level ($X^2(1) = 6.07, p = 0.01$), child school type ($X^2(1) = 18.63, p < 0.001$) and moderate/severe neurodevelopmental disability ($X^2(1) = 15.9, p < 0.001$). Poorer HRQoL, both physical ($t(161) = 3.54, p < 0.001$) and mental health ($t(161) = 4.07, p < 0.001$) were associated with high parental distress in the univariate analyses.

Following the model 1, parents who were not employed or studying were more likely to experience high parental distress ($aOR = 3.1, 95\% CI: 0.9, 10.8, p = 0.07$). This association had borderline significance. After adjusting for the other variables associated with high parental distress, the association with higher level of education and parental distress increased ($aOR = 4.2, 95\% CI: 1.2, 14.7, p = 0.02$), and the associations between high stress and child's SEN school or unit attendance and moderate/severe neurodevelopmental disability reduced, but remained significant (SEN: $aOR = 4.4, 95\% CI: 1.1, 17.3, p = 0.04$ and disability: $aOR = 5.6, 95\% CI: 1.3, 24.2, p = 0.02$, respectively) (Table 4-6).

When considering that the parental distress domain measured personal stressors in the parent's life that may have directly impacted parenting, such as depression or partner relationship satisfaction, it was unsurprising that the parent's perceived physical and mental health were strongly associated with parental distress (model 2) reducing the effects of the other variables to non-significance. Higher parent education level remained borderline significant ($aOR = 4.1, 95\% CI: 1.0, 17.6, p = 0.06$) (Table 4-6).

4.4.5.3 Parent-child dysfunctional interaction

On univariate analyses mental HRQoL ($t(159) = 3.27, p < 0.001$), parent receiving income support benefits ($X^2(1) = 3.75, p = 0.07$) and child's moderate/severe neurodevelopmental disability ($X^2(1) = 3.06, p = 0.10$) were associated with high parenting stress resulting from parent-child dysfunctional interaction. Following the adjustment for these variables, only the association between lower perceived mental health and experience of dysfunctional interaction persisted ($aOR = 0.9, 95\% CI: 0.9, 1.0, p < 0.001$). (Table 4-6).

4.4.5.4 *Difficult child*

In line with the multiple linear regression analyses presented above, younger child age was associated with high reported parenting stress in the Difficult Child domain ($t(166) = 2.19, p = 0.04$). In addition, the child attending a SEN school or unit ($X^2(1) = 7.59, p = 0.01$) and having a moderate/severe neurodevelopmental disability ($X^2(1) = 3.55, p = 0.05$) were associated with the parental experience of difficult child behaviour negatively impacting their parenting. On univariate analyses there were no significant associations between high parenting stress caused by the child's behavioural factors and parent demographic characteristics (age; ethnicity; education level; employment status; marital status; living with the other birth parent; number of other children; average IMD decile at 11 years or at birth). Poorer HRQoL, both physical health ($t(161) = 2.37, p = 0.01$) and mental health ($t(161) = 4.46, p < 0.001$), and receiving income support benefits ($X^2(1) = 8.21, p = 0.01$) were associated with high stress in the DC domain.

After adjusting for all other variables associated with high parenting stress in the univariate analyses, except perceived HRQoL (model 1), the association between high stress in the DC domain and younger child age ($aOR = 0.4, 95\% CI: 0.2, 0.9, p = 0.03$) and the parent receiving income support benefits ($aOR = 3.7, 95\% CI: 1.3, 10.6, p = 0.01$) remained. The association with the child attending SEN school or unit had a borderline association with high parenting stress in this domain ($aOR = 3.0, 95\% CI: 0.9, 10.2, p = 0.07$), whereas the association with child moderate/severe neurodevelopmental disability diminished ($aOR = 1.1, 95\% CI: 0.4, 3.0, p = 0.84$). After introducing the parent's perception of their HRQoL into the model (model 2), the association between high DC stress and other variables diminished. The association between high stress scores and the parent receiving income support benefits remained borderline significant ($aOR = 2.9, 95\% CI: 1.0, 9.0, p = 0.06$) (Table 4-6).

4.5 Discussion

4.5.1 Summary of key findings

In this quantitative research strand of a convergent MM study, I examined parenting stress and parent reported HRQoL among groups of parents of EP and FT born YA at the time their children transitioned to secondary education. The objective of this study was to identify sources of parenting stress among primary caregivers of EP YA and their age, gender and school class matched control children's primary caregivers. Consistent with findings among parents of younger preterm born children (Schappin et al., 2013), in this study EP parents reported elevated levels of parenting stress in comparison with FT parents. The median Total Stress score among EP parents was considerably high in comparison with the control parents' median stress (*Median: 71, (IQR: 53, 92)* and *Median 55, (IQR: 45, 71)*, respectively). Yet, aligned with previous studies among parents of VP born children (Landsem et al., 2014; Linden et al., 2015; Schappin et al., 2013; Treyvaud et al., 2014) both groups of parents reported average parenting stress scores that were within the normal range of parenting stress (Abidin, 2012). As discussed earlier in this thesis, a level of parenting stress is typical as stressors occur in the parent-child relationship (Abidin, 1992; Deater-Deckard, 1998; Lazarus, 2000) and consequently, although heightened, the level of stress reported by EP parents in this study may have indicated typical levels of stress among parents whose children may have had developmental challenges (Marlow et al., 2021). Consistent with studies among middle school-aged VP born children (Linden et al., 2015; Singer et al., 2007; Treyvaud et al., 2014) parenting stress levels of EP parents were higher in comparison with controls. Twelve percent (20/167) of parents in this study experienced high levels of stress (>84th centile) in their parenting roles approximately 11 years after giving birth EP, whereas none of the parents of FT born YAs reported high levels of parenting stress.

In the fully adjusted linear regression analysis, on average EP parents reported a 14.8 (95% *CI: 9.2, 20.4*) point higher total parenting stress level in comparison with parents of FT born YA. The covariates of child male sex, parent age, and IMD decile at 11 years had little effect on the average stress scores among the groups of parents, suggesting that other factors were influential. In line with this, when factors associated with high parenting stress among EP parents were assessed in the logistic regression analysis, child male sex or sociodemographic characteristics of parents, such as ethnicity, educational level, or the family's IMD decile at birth or at 11 years, were not associated with high total parenting stress. Higher parental educational attainment has been associated with lower levels of parent and child-related stress among parents of 8-year-old VP born children (Singer et al., 2007). Yet contrary findings have been reported among VP populations (Linden et al., 2015) and parents of adolescents with

ADHD (Wiener et al., 2016). Yet none of these studies utilised the PSI-SF instrument which prevents direct comparison of stress scores between the studies. In the present study, the relatively small number of parents reporting high TS and the fact that the families taking part in this study had generally high SES, may have influenced the finding that parent SES was not associated with high TS. Parental distress increased among those EP parents who had a university level education, whereas parents of younger EP children and those who received income support benefits reported high levels of child-related parenting stress. Higher levels of total parenting stress have previously been associated with low income level among VP and FT parents of 8-year-old children (Singer et al., 2007). Among a low-income population, instead of the household income level, low parental educational attainment and high family conflict were associated with increased parenting stress among parents of adolescents (Anderson, 2008) In the present study, parents of EP YA who attended SEN schools or units more commonly reported increased levels of total parenting stress in comparison with those parents whose children were educated in mainstream schools. These findings will be further discussed below.

Consistent with studies among younger preterm born populations (Schappin et al., 2013; Treyvaud et al., 2011; 2014), in the present study, in early adolescence EP parents reported most heightened child-related parenting stress (Difficult Child domain). In addition, among EP parents of younger children, the stress levels were higher than among parents of older children, suggesting that behaviours of younger children were perceived as more challenging, although the age range of EP children in this study was narrow (10.6 to 13.0 years). This finding was in line with the findings reported by Singer et al. (2010) that towards adolescence parenting stress levels among parents of FT born children increased, whereas they decreased among parents of children born VP with low medical risk. Parents of adolescents with cerebral palsy have reported decreasing parenting stress with increasing child age (Guyard et al., 2017; Wiegner & Donders, 2000). A decreasing parenting stress along with increasing child age among EP parents may have related to improved parental coping with child behaviours (Almogbel et al., 2017), which may have related to increased parental as well as dyadic experience in the parent-child relationship (Thomasgard & Metz, 1997). The finding may also reflect a parental experience of relief, as a child who may have initially experienced developmental difficulties was performing comparably with peers (Marlow et al., 2020). In addition, in the present study, EP parents of YA, of whom a majority attended mainstream schools, may have felt that as their children got older they needed less parental involvement (Yerkes et al., 2019) and consequently, parents may have felt less child-related stress in their roles as parents. Increasing parenting stress with increasing child age among the FT parents in this study may have indicated increased parent-child conflict in adolescence, which may have

been less common among adolescents born preterm and their parents (Kajantie et al., 2008), although parents of adolescents from both groups have reported similar low levels of family conflict in other comparable studies among preterm born populations (Burnett et al., 2017; Greenley et al., 2007). As a limitation of the present study, no measures of family functioning were utilised. The proportion of parents reporting high levels of total parenting stress pre and post transition to secondary school among EP parents did not differ, suggesting that the stress reported by EP parents of younger children was not linked to parental concerns about the transition per se.

Overall, the groups of parents perceived their HRQoL comparably. The average HRQoL, both physical and mental, were close to the norm among EP parents. Similar to the PSI-4-SF instrument, in the SF-12v1 analysis parent perceived mental health improved with increasing child age among EP parents, whereas control parents of older children reported decreasing perceived mental HRQoL. As opposed to the Parenting Stress Index, the SF-12v1 questionnaire was not an outcome measure directly related to the parenting role, but rather measured overall perceived HRQoL of the individual. Similarity in the associations with covariates between the instruments, and between the linear and logistic regression analyses, increased the validity of these findings, although, small cell counts and consequently wide confidence intervals in the logistic regression analyses should be noted when interpreting these results. The comparable trend in child age in parent perceived HRQoL and in parenting stress among the EP parents supports previous findings that in addition to parenting stress levels (Landsem et al., 2014), parent outcomes in general (e.g., partner relationship, social relations, emotional health, life satisfaction) among EP parents may improve over time (Saigal et al., 2010) which may relate to parent factors such as reduced social isolation among older parents (Schappin et al., 2013). Yet, in the present study, increased parent age and lower average IMD decile predicted poorer physical HRQoL among all parents.

In the logistic regression analysis, SEN schooling appeared to have a stronger association with high TS in comparison with child disability. The odds of EP parents with children at SEN schools or units reporting high overall parenting stress compared to the parents whose children attended mainstream schools or were home educated reduced but remained significant after adjusting for all other variables associated with high parenting stress, except parental mental HRQoL. The higher impact of SEN schooling in comparison to child disability variable could reflect the earlier discussion that child behavioural difficulties and mental health disorders may be more stressful for parents than physical/functional child disability (Barroso et al., 2018). The child disability variable in the EPICure2@11 Study did not include behavioural or

psychiatric disorders. Unfortunately, data on these child outcomes (e.g., psychiatric disorders) were not available at the time of the present analyses.

4.5.2 Parenting stress scores in the context of previous research

As presented in the introduction to this chapter, a trend of initially heightened parenting stress scores that decrease in early childhood has been reported among parents of preterm born children in some studies (Singer et al., 2010), whereas others have found more stable levels of stress (Gerstein & Poehlmann-Tynan, 2015). Yet, in the present study, the EP parents reported comparable levels of total parenting stress with levels that parents of preterm born infants have reported. A meta-analysis of parenting stress among parents of preterm born (mean GA ~32 weeks) children reported a pooled average Total Stress score of 71.6 (95% CI: 68.3, 74.8) from five studies utilising the PSI-SF instrument at 0-18 months after the birth (Schappin et al., 2013). The pooled average TS was a similar level of stress in comparison with the average stress that EP parents in this study reported approximately 11 years after giving birth. Similar to the present study, parents of preterm born infants experienced stress within the normal range. When examining several parenting stress instruments together, the authors found that maternal age was a significant moderator of the effect sizes, indicating that at a younger child age older mothers experienced less stress (Schappin et al., 2013). In the present study, the overall picture of parenting stress among the groups of parents was little affected by the effects of the covariates of parent age, the family's IMD decile at 11 years and child factors. Similarly, high parenting stress was not associated with parent age in the univariate analyses. Factors other than those assessed in the present study may predict parenting stress in adolescence, such as child behavioural disorders and family functioning (Guyard et al., 2017; Wiegner & Donders, 2000).

Studies have found stress levels comparable to parents in this study among parents of preterm born older children. In a randomised controlled trial of an early intervention programme, 61 Norwegian mothers of 9-year-old children born with BW <2000 grams reported an average Total Stress score (PSI-SF) of 67.0 in the non-intervention arm, whereas the average Total Stress score of the comparator FT parents was 54.7. Like the EP parents in this study, child-related parenting stress was the most heightened stress domain among the Norwegian parents of preterm born children (Landsem et al., 2014), supporting the impression that child behavioural factors might be impactful.

A retrospective single hospital-based Croatian cohort study compared parenting stress among three groups of mothers; 1) mothers of late preterm born (34-36 weeks' gestation) infants

requiring intensive care; 2) mothers of full-term born infants admitted for intensive care and 3) mothers of late preterm born children who were not admitted for intensive care. The authors discovered that mothers of preterm born children, regardless of the experience of intensive care, reported higher levels of total parenting stress utilising the PSI-SF instrument, six to 12 years after giving birth in comparison with parents of FT born children. The average stress levels were 1) 88.0, 2) 58.0 and 3) 87.0 among the groups of parents, respectively. Similar to other studies, mothers found the characteristics of their child (Difficult Child domain) to primarily affect the levels of maternal stress. It was noticeable that the parents of late preterm born children reported a nearly 10 point higher average total stress score than the EP parents in the present study. Children in their study were born at older gestations, perhaps at lower medical risk level, although early parental distress may not directly correlate with infant medical risk (Brummelte et al., 2011; Caporali et al., 2020), but higher medical risk has been associated with later developmental concerns that have been linked to parenting stress (Brummelte et al., 2011). In addition, approximately two-thirds of parents of preterm born children in their study reported clinically significant levels of parenting stress (>90th centile), whereas 10% of parents of full-term born children experienced high levels of parenting stress (Polic et al., 2016). None of the control parents in the present study reported parenting stress on high levels. The authors examined covariates together for all parents and found that lower education and older child age were associated with higher total parenting stress scores, whereas family income had no effect (Polic et al., 2016).

To support the hypothesis that at older child ages behavioural difficulties may have a particular impact on parenting stress, in a study of parental perceptions of child characteristics and parenting stress, parents of two to 18-year-old British children diagnosed with ASD (mean age 7.7 years) or ADHD (mean age 8.5 years) reported average total stress scores (PSI-SF) of 105 among both groups of parents, whereas the average parenting stress score among parents of typically developing children (mean age 5.9 years) was 62. The authors found that among all parents in their study, older child age, male sex, and lower parental education predicted higher total stress scores (Larkin et al., 2021). In the present study, the EP parent group included parents of both typically developing children and children with long-term disabilities and thus formed a more heterogenic group of parents.

Even though EP parents in the present study reported statistically significantly higher levels of stress in comparison with control parents, the stress levels of EP parents, and proportions of parents reporting high stress, were generally modest and somewhat comparable to other clinical and general populations. Parents of approximately 9-year-old children with cerebral palsy (45% were born preterm; mean GA 31 weeks) reported an average total stress score

(PSI-SF) of 66.7, although 44% of parents in their study reported high parenting stress (>85th centile) (Majnemer et al., 2007). Carers of nine to 14-year-old Northern Irish adolescents in placement reported 78.3 average points of total parenting stress among carers of boys and 70.3 average points among carers of girls (PSI-SF). A third of these parents reported 'abnormal' levels of parenting stress (the authors did not specify the exact cut-off used) (McSherry et al., 2019). Similarly, a study assessing consistency in parenting stress levels among a self-selected general population sample of 111 White American mothers found an average Total Stress score of 74.1 at children's age of 10 years and 76.3 at the age of 14 years. As among control parents in the present study, parenting stress levels increased with child age (Putnick et al., 2010). Finally, a self-selected sample of parents from Houston, Texas whose three to 18-year-old children (53% were above the age of 12) with neurodevelopmental disorders attending SEN schools reported an average total parenting stress score of 108; 71% of the parents experienced high parenting stress (>80th centile) (Almogbel et al., 2017).

As shown above, varying studies among high-risk populations of parents of older children have generally used the cut-off for high parenting stress (>84th centile) as determined by Abidin (2012). However, Barroso et al. (2016) examined the psychometric properties of the PSI-SF among a high-risk (defined as high levels of child behavioural difficulties) sample of mothers and their young children (up to the age of 15 months). The authors concluded that among high-risk populations, such as in their study, a lower clinical cut-off of 73rd to 77th percentile scores could be used. However, as discussed throughout this thesis, and demonstrated in this chapter, the children in the present study were not necessarily a homogeneous group of 'at-risk' YA. A large proportion of EP children in this cohort had no long-term morbidities and the participating families had generally high SES. In addition, the origin of stress among parents of young adolescents may be different, for example, worries about child social functioning versus worries of challenging parent-child interaction with a toddler with behavioural difficulties. As discussed earlier, the birth experience and adjustment to a life with a new child may still contribute towards parental stress levels among parents of toddlers, whereas among parents of adolescents, regardless of initial 'at-risk' status, the stress levels may be less and of different origin.

The increased level of parental distress among EP parents in comparison with controls diminished when families with children with severe/moderate impairments and multiple births were excluded. Child age did not impact the groups of parents differently for this outcome. This suggested that the potential personal distress that the parent may have experienced in their parenting role did not vary due to the child's age or birth status but was influenced by the presence of child disability or/and multiple births. Aligned with this finding, Treyvaud et al.

(2014) found that high levels of parental distress among both VP and FT parents persisted from child age of two years to the age of seven years. In the present study, the groups of parents perceived their HRQoL similarly in all group comparisons. Those EP parents who reported high parental distress, however, perceived their mental HRQoL as strikingly poor in comparison to those who reported low levels of stress (high stress: *Mean* =40.1, *SD* =11.2 and low stress: *Mean* =50.6, *SD* =8.2, respectively). Even though the cross-sectional design of the present study prevented inferring causality between low parental mental health and increased parenting stress, the association might have suggested that the increased psychological ill-health experienced among EP parents with children with neurodevelopmental impairments was related to their parenting roles. A correlation between parental psychological ill-health and increased parenting stress have been reported before (Linden et al., 2015). In the present study, many of the parents who were not employed or studying reported being homemakers or carers for their children, and more commonly experienced high levels of parental distress. This may have suggested that caring for a young adolescent with long-term disabilities at home was personally stressful. Not being in a paid employment may have indicated the family's low-SES which has been associated with increased parenting stress (Singer et al., 2007). However, since the IMD at birth or at 11 years, or receiving income support benefits were not associated with high parental distress in this study, low SES was a less likely contributor to parental distress among the EP parents in this study. The way in which EP infants were screened and recruited to the main cohort had been robust (Costeloe et al., 2012). Yet maternal mental health outcomes before the present study had not been examined. Therefore, as was the case in several studies discussed in the review chapter, maternal mental health concerns among some parents could have pre-existed the birth and carried on over the years.

It was unfortunate that due to disparities in outcome measures (e.g., PSI full form versus PSI short form), I was unable to directly compare parenting stress scores from this study to those presented in the systematic review. A particular point of interest might have been to examine whether evidence existed that parenting stress levels were increasing over time, in the context of changing parenting culture and increased sociocultural focus on parenting practices (Smith, 2010). This is particularly as the previous cohort studies had commonly been established in the 1980s and early 1990s. Some evidence of overall decreasing parental stress levels in modern birth cohorts in comparison with older studies have been presented (Schappin et al., 2013). A majority of the respondents in this study were white female parents from higher socio-economic backgrounds; the sociodemographic group that has been suggested to most commonly internalise intensive parenting practices in the parenting culture literature (Lee et al., 2014, p. 31-33; Shirani et al., 2011). Nevertheless, as both EP and control parents in this

study were from similar sociocultural environments, this was unlikely to contribute towards the differing parenting stress levels among the groups of parents.

As presented in the systematic review, the effects of SES on parent outcomes among parents of preterm born children vary depending on the measure of SES and the type of family impact assessed. Indredavik et al. (2005) found that among parents of VP born 14-year-old Norwegian adolescents higher parent SES promoted higher parental distress. Contrary to this, when their adolescent children born with BW less than 750 grams in Cleveland, Ohio were aged between 11 and 14 years, the parents with low SES reported higher parental distress compared to parents with high SES (Moore et al., 2006). Both Moore et al. (2006) and Indredavik et al. (2005) included parent education level in their measures; they used the Hollingshead Four and Two Factor Indices, respectively. However, when Moore et al. (2006) assessed life stressors and social resources of the families, such as intergenerational relations or employment, but not parent education level, they found that families with high resources and children born with BW <750 grams reported higher family burden at all ages.

4.5.3 Reflections on limitations

A limitation of this study was that, overall, a relatively modest proportion of families from the original EPICure 2 cohort participated in this re-assessment at 11 years. The quantitative sample constituted only approximately a fifth of the families in the original EPICure 2 cohort. In total, 1041 children in the EPICure 2 cohort were discharged alive from hospital. Overall, one hundred and seventy-five parents reported parenting stress data. The low follow-up rate was due to varying factors. Firstly, only 46.3% (482/1041) of families in the EPICure 2 cohort were invited to participate in the re-assessment at 11 years based on their neonatal care network at birth. Thus, a majority of the families in the original cohort were not given an opportunity to take part. Of those families who were invited to participate, 45.6% (220/482) consented to take part, some of whom failed to provide parenting stress data. Thus, even though the response rate among those who had an opportunity to participate in the re-assessment (i.e., invited families) was relatively satisfying, due to the design of the sampling process in the main study, the proportion of assessed families from the total cohort was modest. Challenges in participant recruitment resulted in a sample size of approximately half of the initially invited parents. The original target sample size was 300 index families, and eventually 220 were successfully recruited. The limited size of the sample may challenge the generalisability of the findings of this study. In total, 125 control parents returned their Parent Questionnaires. Since recruitment of control families was done by head teachers at schools, data on number of families that were approached but declined participation was not available. In the present

study, every effort was made to conduct a comprehensive comparison of sociodemographic characteristics between participants and non-participants. Yet it remained a speculation how those parents who did not take part would have rated their parenting stress levels. However, since no previous data on parenting stress or HRQoL among parents of YA born EP in England existed, the contribution of the present study was valuable.

When examining sociodemographic characteristics of those EP parents who contributed data to this study some evidence existed that they were from higher socio-economic backgrounds in comparison with the remaining families in the EPICure 2 cohort. The parents who returned their Parent Questionnaires at 11 years were older and had on average higher IMD deciles at delivery than non-participating parents. In addition, fewer parents from Black ethnic minorities took part in the re-assessment. Previous studies have suggested that parent outcomes among Black ethnic groups may be poorer (Holditch-Davis et al., 2009). Non-response bias resulting from lost to follow-up of families from higher sociodemographic risk groups has been a frequent challenge of longitudinal cohort studies (Gerstein & Poehlmann-Tynan, 2015; Johnson et al., 2014). However, as discussed earlier, the direction of impact of SES on parenting stress remains somewhat ambiguous.

In this study, EP and control parents had comparable sociodemographic characteristics, except for parent ethnicity and family benefits. Control parents were commonly White, whereas EP parents were from more varied ethnic backgrounds. Previous studies among general population have suggested that minority group parents may report higher levels of parenting stress (Franco et al., 2010). Thus, a proportion of the differing stress levels among the groups of parents found in the present study could have been impacted by a difference in the ethnic backgrounds of EP and FT parents. EP families were more often recipients of income support and incapability/disability benefits. Yet, the differences in the proportions of families receiving disability and income support benefits were expected as EP families more often had children with morbidities. The interview data, that will be discussed in detail in the next chapter, suggested that in a comparison with FT parents, EP parents were more often either full-time or part-time carers for their children and worked reduced hours in paid employment. Consequently, they were more likely to be eligible for benefits. Unfortunately, data on hours of paid work of all parents were not collected and therefore it was not possible to assess whether the trend was similar in the EPICure2@11 Study population as a whole (i.e., in the Parent Questionnaire, the parent could have reported being in employment even if they worked only limited hours). The similarity in background characteristics between EP and control parents in this study suggested that, to an extent, the increased parenting stress was linked to the status of being a parent of an EP young adolescent.

Only a small proportion of parents who took part failed to fully complete their questionnaires (<10%). Participants whose PSI-4-SF was incomplete, whose children were not assessed or who were resident outside of England, when IMD deciles could not be obtained, more commonly were parents of children with severe morbidities and were recipients of disability benefits. These parents were also more commonly single. Considering these familial factors, the parents perhaps had less time to contribute towards the study activities, which resulted in missing data. The characteristics of parents with complete and incomplete data were otherwise similar.

The relatively small quantitative sample size limited statistical analyses. Particularly, the relatively small number of EP parents reporting high levels of parenting stress limited the applicability of logistic regression analyses. This was demonstrated by wide confidence intervals. Yet, with such small numbers, several comparisons showed strong statistical significance. The well-matched control parents provided an excellent comparison group and, together with data triangulation, improved the validity of findings.

Lastly, since this study was both cross-sectional and exploratory in its nature, it allowed me to describe parenting stress levels and examine their associations with other variables in this study population, but it did not permit me to examine causality. Thus, the findings from this study provided a ‘snapshot’ into the lives of its participants.

4.5.4 Suggestions for future research

As presented in the introductory chapter of this thesis, in the research field of prematurity, the event of premature birth has generally been regarded as a negative parental exposure, whereas in popular discourse preterm birth may be framed in a more positive light (Isaacson, 2002). Undoubtedly, uncertainty surrounding child’s survival and later life health status increases parental stress. As discussed in the review chapter, however, as the child reaches adolescence a wider range of parental experiences may emerge. Mothers of ELBW young adults with neurosensory impairments reported feeling significantly better about themselves for managing their child’s health compared to mothers of ELBW young adults with no impairments (Saigal et al., 2010). In addition, families with adolescents born preterm reported fewer stressful life events (Singer et al., 2007). Families with ELBW young adults with disabilities reported improved family functioning in comparison with families with healthy ELBW children (Saigal et al., 2010). The factors influencing positive family outcomes have not been systematically examined, but they may relate to factors such as; a) parents of prematurely born children with disabilities have experienced more uncertainty around their child’s physical and functional

development and have therefore adapted differently; b) parents may have had a prolonged stay at NICU with more extensive support (such as parental bonding and sensitivity) compared to parents whose infants were discharged earlier and therefore cope better with uncertainty; c) due to their child's medical needs they may have developed closer connections with healthcare professionals and other support services over the years strengthening the parental support networks. Simultaneously, parents of children born preterm have reported increased marital dispute and fewer subsequent children after the EP birth (Alenius et al., 2018; Saigal et al., 2010). In the present study, EP parents of singletons with mild/no disabilities reported better mental health than FT parents, although this association did not reach statistical significance. Future research could explore the emergence of positive parental experiences or/and robustness of those experiences over time and factors associated with them.

Studies assessing parenting stress among parents of preterm born children do not need to aim to demonstrate that EP parents or their children are at increased risk of poor outcomes in comparison with families of FT born children. Rather, studies may aim to understand better the types of risks that EP parents may experience, and whether risk factors differ to those of the parents of term born children. Frameworks to assess the initial responses of parents to traumatically stressful events such as diagnosis of child disability, have initially been viewed as progressive linear models in which the vocabulary, terms such as 'coping', 'acceptance' or 'denial', have been provided by the researchers (Landsman, 1998). A main focus of these models have been to reduce parent stress and promote adjustment, and many of them have overlooked possible transformative aspects of traumatic events, such as positive life changes (Scorgie et al., 2004). I will return to the concept of parental adjustment in the next chapter. Linear frameworks have been criticised as they may suggest that those parents who do not manage stressors and thus do not 'cope', may themselves become a 'risk' in their child's life due to 'sub-optimal' parenting behaviour (Hoffman, 2010). Alternative ways to conceptualise parental responses have been presented, such as those allowing parents to define and re-define their experiences continuously (Landsman, 1998).

Treyvaud et al. (2014) demonstrated that the experience of stress in parents of preterm born children may fluctuate. In addition, instead of a linear progress from shock to acceptance in stages, a qualitative study found that parental adaptation to child disability may be a back and forth movement when events occur in life and in the family (Roll-Pettersson, 2001). Several longitudinal cohort studies have explored parental stress as a trend, either decreasing or increasing over time. However, fluctuation might signal changing adaptations and re-emergence at different points in life, and thus, call for frequent and long timeframes of analyses. Future research could further explore these processes among parents of EP children.

Key findings:

- 1) Parents of EP YA reported higher levels of parenting stress in comparison with controls, that was independent of the presence of child disability
- 2) A proportion of EP parents reported high levels of parenting stress and considerably low levels of mental HRQoL approximately 11 years after giving birth
- 3) Parenting stress level decreased, and parent perceived mental health improved with increasing child age among EP parents, whereas control parents of older children reported increasing parenting stress levels and decreasing perceived mental HRQoL
- 4) Covariates of child male sex, parent age, and IMD decile at 11 years had little effect on the average stress scores among both groups of parents
- 5) Among EP parents, child attendance at an SEN school or unit was associated with high total parenting stress scores, a university level education was associated with high parental distress scores and receiving income support benefits and younger child age were associated with high levels of child-related parenting stress

Chapter 5 Parental Experiences

In this chapter I will present findings from the qualitative telephone interviews. As a context within which to consider the interviews, I will first discuss targeted literature on parental experiences of parents of preterm born children. I choose to discuss recent qualitative studies among families with children born preterm and among adolescents with long-term morbidities as these investigations provide relevant empirical findings to contextualise my interview data. I then re-visit my positionality discussed earlier in Chapter 3. I further discuss the theoretical lenses I have utilised in this thesis which shaped how I understood the interviews, and how I approached the interview data. I deepen the discussion presented in the introductory chapter of this thesis. The methods used to collect and analyse the data are then described in detail. Following this, the main results are presented together with direct quotes from the parents' interviews. The chapter will culminate in a discussion of findings.

5.1 Background

Parental experiences over time - As discussed earlier in this thesis, EP parents' experiences may be marked by the EP birth and prolonged infant hospitalisation that has often lasted for months (Baia et al., 2016; Holditch-Davis & Miles, 2000; Wraight et al., 2015). Parents of infants admitted for neonatal intensive care have, for example, reported feeling stress, anxiety, fear and guilt in relation to their infant's survival and later life health (Wraight et al., 2015). Swiss parents of two to 3-year-old VP born children recalled a feeling of 'loss' of being able to feel 'trusting' of their infant's health status during the NICU stay (Schuetz Haemmerli et al., 2020). In addition, family functioning such as financial coping or/and sibling care may become interrupted (Kantrowitz-Gordon, 2013; Kantrowitz-Gordon et al., 2016). The EP birth may have been an unexpected, potentially life-threatening event to the mother or may have followed from a lengthy stressful time under the risk of preterm labour while the woman may have been subjected to bed rest and intensive medical monitoring impacting psychological health and family functioning (MacKinnon, 2006).

The time in the NICU is characterised by distance between the infant and parents, perhaps challenging the development of the parent-infant relationship (Kantrowitz-Gordon, 2013; Schuetz Haemmerli et al., 2020). In a qualitative study of parental experiences when their infants were admitted for neonatal intensive care and were mechanically ventilated, but not facing end-of-life decisions, 10 parents from the US expressed concerns about 'bonding' with their child. They were pleased when they were able to assist in their infant's care and expressed feelings of guilt and self-blame for the hospitalisation. The parents referred to their infants as

‘fighters’ and did not want to ‘give up’ on their children (Wraight et al., 2015). In this context, the transition to parenthood may become ‘disrupted’ (Bury, 2001) as parents of EP children may be required to begin to reformulate their perceptions of themselves as parents of a medically ill child or child with disabilities instead of a parent of a healthy infant (Kantrowitz-Gordon et al., 2016; Landsman, 1998; Widding & Farooqi, 2016)

Following discharge from hospital, many parental pressures, both psychological as well as those relating to the family life, may continue. Particularly mothers may still experience mental health problems relating to the adverse birth experience such as post-traumatic stress disorder (Zerach et al., 2015). In the NICU the infant care had been supported by medical professionals, yet at home parents may feel overwhelmed by the responsibility (Jackson et al., 2003). A marked feature of being a parent of an EP young child may be the uncertainty about child development and future health (Marlow et al., 2020). EP parents have described prolonged feelings of worry relating to their children’s outcomes (Kantrowitz-Gordon et al., 2016). Yet parents may feel a sense of relief when they realise that despite the adverse birth, their child is developing comparably with their peers (Marlow et al., 2020). In a study of 10 mothers of children with learning disabilities, the mothers described the process of becoming a parent of a child with disabilities as a slow process, but which had a ‘point of realisation’ (Valle, 2017).

In a study from New Zealand, nine mothers of four to 7-year-old children born EP (≤ 28 weeks’ gestation) were retrospectively interviewed about their neonatal intensive care experiences and current quality of life. The interviewees’ measured their current quality of life against their NICU experiences. The mothers described the first year following birth as a nadir. Due to the frequent contacts with healthcare professionals, they felt as if they were raising a child in a “fishbowl” (Wilson & Cook, 2018, p. e1634). Strong partner relationship preceding the birth had assisted the mothers to persist through the early years of childhood. The study recruited their participants by advertising in a follow-up care clinic and online. Thus, it may be that the parents who decided to take part felt particularly strongly about their early parental experiences (Wilson & Cook, 2018).

There is a lack of research on experiences of parents of adolescents born preterm, particularly among parents of children born at the lowest gestations (Wilson & Cook, 2018). As discussed in the preceding chapters, children born EP may have multiple simultaneous developmental and behavioural concerns that can impact the parent-child relationship (Drotar et al., 2006). In addition, by adolescence, the parents may have experienced increased strains relating to the parent role, poor psychological health, and parenting stress for a long period of time. Parents

with mental health concerns have reported regretting becoming a parent due to a concern that their own poor psychological health prevents them from being a ‘good parent’ (Moore & Abetz, 2018). Adolescence is a particular time when becoming an adult with an independent future may become more topical, and the importance of peer relationships is heightened (Casey, 2015). Simultaneously, disorders among adolescents themselves, such as ADHD symptoms, may worsen (Hallberg et al., 2009). In this context, parents may experience new challenges while they may become increasingly aware of their child’s future independence. In a Swedish study of parental concerns, mothers of adolescent girls diagnosed with ADHD reported feeling anxious about being continuously solely responsible for their daughters’ well-being while the teenagers’ behaviours were particularly concerning (e.g., threats of suicide and violence). A majority of the 12 mothers had developed stress-related conditions such as chronic pain (Hallberg et al., 2009).

Attachment and early childhood discourses in parental experiences - In his early theory on child competence, discussed earlier in Chapter 1, Ogbu (1981) critiqued the psychoanalytic framework in which early experiences were regarded to form the basis for adult competence. In psychoanalytic theory, parents were perceived as ‘teachers’ (Maccoby, 1992) carrying out a “hidden curriculum at home” (Ogbu, 1981, p. 415). Following from this framework, groups perceived as ‘at-risk’, such as children born EP, may have lacked in early experiences and consequently, together with other risk factors related to the early birth, such as altered brain development, child development may become compromised. This may be evident later as limited adult competence, such as academic achievements (Ogbu, 1981; Wolke, Jaekel, et al., 2013). Aligned with this rationale, it has been suggested that a strong parent-child relationship is particularly important for preterm born children’s development (Widding & Farooqi, 2016) and the parent-infant relationship and attachment experiences have received particular attention in the NICU context (Kantrowitz-Gordon, 2013). The logic suggests that a secure attachment experience among parents will lead to “involved parenthood” (Widding & Farooqi, 2016, p. 3) and consequently improve child outcomes. Yet according to Ogbu (1981), for example, those culturally valued skills required for adult competence may be acquired throughout life when contexts change and new skills are needed. Among families with children with disabilities, healthcare professionals may overestimate the importance of early experiences as those are perceived as ‘interrupted’, and underestimate the potential influences of later life experiences (Roll-Pettersson, 2001).

Widding and Farooqi (2016) examined negative feelings in ‘birth stories’ of four mothers of children born EP, 10 to 12 years after the birth. Their investigation emerged from the field of discursive psychology, understanding that parental experiences were partially about the

dominant discursive framings but also that the parents interpreted, responded, and altered the dominant understandings in their stories. Within the framework of attachment theory and the importance of the early parent-child relationship, parents of EP children discussed their negative feelings of becoming a mother which were experienced as a dilemma against the discourse of 'involved parenthood'. The mothers utilised the concept of attachment to justify varying viewpoints. They perceived 'natural' not to 'bond' with their infant when the child was perceived as ugly (e.g., described as a chick or alien) or sick, and when the mother's 'natural hormones' rejected attachment. Yet the mothers also discussed their attachment experiences as being built over time following repeated caregiving efforts. Thus, for these mothers 'secure attachment' was a process rather than an 'instant bond'. Widding and Farooqi (2016) stated that "motherly feelings can eventually grow into something 'incredibly strong'" (Widding & Farooqi, 2016, p. 12). In a comparative mixed methods study of parental experiences among parents of two to 3-year-old preterm born children, the respondents described the experience of becoming a parent as a deepening feeling of attachment growing over time (Schuetz Haemmerli et al., 2020). Similarly, in a study of the impact of music therapy on parent-child relationship among 11 families with children with ASD aged three to six years, the music therapy impacted mothers' perceptions of their children, their response to the child and therefore, their relationship with the child. The parents described feeling more hopeful about the future. The authors concluded that building a strong parent-child relationship could be regarded as a target rather than a vehicle (Thompson & McFerran, 2015).

Parental adjustment over time - Child chronic condition/s may demand parents to adapt to the altered parenting context. A qualitative study among 46 parents of seven to 16-year-old Swedish adolescents with cognitive disabilities assessed a psychoanalytic framework of sequential stages of adaption-mourning in parental experiences. A few parents experienced progressing from shock to acceptance in stages, but for many of the parents the adaptation was a bidirectional fluctuating process as new events occurred in life and in the family. Parental experiences were related more to a feeling of disappointment at particular points in life, such as at the onset of puberty, in comparison to a feeling of chronic sorrow. Parental reactions were influenced by the level of impact of child disability on the family (Roll-Pettersson, 2001).

In a longitudinal ethnographic multi-method investigation about changes in parental roles of 68 parents of children with complex care needs aged six months to 26 years, being a parent was described as a labour-intensive undertaking that had put stresses on the parents' relationship, social life, and work. The parents described continuously increasing parental responsibilities due to a lack of progress in their child's development. Among the parents, increased caregiving responsibilities were linked to psychological and physical health

problems such as increased stress, anxiety, pains, and lack of sleep. It is important to note that although the study had a longitudinal design, which was further strengthened by multi-method data collection, such as interviews and photovoice, only 20 parents participated in all three interview sessions (Woodgate et al., 2015).

Nevertheless, Woodgate et al. (2015) found that for the participants in their study, being a 'good parent' was synonymous with their child having a 'good life'. The parents constituted 'good parenting' as their child "never going without" what they needed (Woodgate et al., 2015, p. 5) which meant that the parents constantly strove to ensure that all their child's needs were met. In general, parental perceptions of what a good child's life may entail are contextually varied, for example in terms of parent educational attainment. Childrearing goals of parents with lower levels of education have been found to entail more 'traditional values' in comparison with highly educated parents (Tulviste et al., 2007). In a study about responsive and stimulating parenting practices and long-term goals and values for their children, among a general population sample of 120 French and 84 European American parents of children up to the age of six years, the parents from both cultures valued self-reliance and self-assurance, tolerance of others and happiness in their children. Yet, for French parents, stimulating parenting practices (e.g., child learning) were more important than emotional responsiveness (e.g., hugging or touching), whereas American parents placed lesser importance on stimulating parenting practices (Suizzo, 2016).

Finally, in a case study, Minoli, Castillo, Ginsberg, and Muraskas (2013) described the long-term outcomes for three 'micropremies' (a combined birthweight of 1100 grams). Their report illustrated the varying contexts for being a parent of EP child well and demonstrated what Ogbu's "making it" (1981, p. 420) in the society may constitute for those born at the threshold of viability:

"All three girls are functioning well. Baby V after having worked as a cook is now working as housekeeper and is very content with her new job. She uses bicycle but she does not drive. She has a boyfriend but at the moment she does not wish to get married. Baby O experienced difficulties at school as hard time memorizing and problems in mathematics. She suffers from depression and anxiety and dependence on others (not drugs). She suffers from hypothyroidism and obesity. Baby D is attending competitive college, majoring in Psychology. She drives, she is an honors student and despite her short stature has had active social life at college" (Minoli et al., 2013, p. 79)

To conclude this introductory discussion, in adolescence parental experiences may still be marked by the EP birth experience and the experience of parenting an EP child over time. Yet secure attachment experiences and the parent-child relationship may develop over time. A diagnosis of child disability, which could occur or change in adolescence, may place

increasing stresses on the family. Cultural framings of parenthood may influence childrearing goals and parents' perceptions of themselves as parents.

5.2 Understanding the Interviews

Throughout this thesis, I have attempted to make my philosophical, theoretical, and personal assumptions of this research project explicit (Braun & Clarke, 2006; Nowell et al., 2017). My personal position was shaped by the facts that I am not English nor an EP parent. Thus, during the interviews, the parents were in the expert position. A point I will return to later in this chapter. Earlier in Chapter 3, as a philosophical stance, I referred to pragmatism and that for this research project the most important consideration in understanding parental experiences was 'workability'; how parents explained or understood the social realities of their 'parenting'. Thus, in a theoretical sense, in these interviews, I was most interested in the social construction of 'parenting' that occurred in the parents' accounts of their experiences when they discussed their parenting roles, actions, and decisions. Thus, my positionality directed my analytical approach to realising the aims of the study and what I wished to learn from the data (Braun & Clarke, 2006).

In the individual interview, a parent (mother) and a non-expert female researcher came together to talk about 'parental experiences' in the context of a study about survival and later health following EP birth. In Giddens' (2013) terms, I regarded the interview as an event in which the parent (together with me and guided by my questions) constructed a version of themselves as parents by reflecting against shared sociocultural framings of 'parenthood'. In his theory of identity, instead of the 'self' being a sum of life events or traits, Giddens (2013) views self-identity as a reflexive retrospective story of self that can be continuously revised. Since the identity is not given (e.g., based on a role, gender, or age), individuals are invited to continuously build their self-identities by reflecting against surrounding social systems, available choices, and possible risks. Following from the constructive nature of self, internal consistency and continuity are central to it; the identity holds within the continuity of the retrospective story of self (Giddens, 2013, p. 3-5; Jordan, Redley, & James, 1994, p. 23-24).

If an individual interview account is built in a particular sociocultural context, a collective set of interviews can be understood as a reflection of that particular sociocultural context. Thus, individual interview accounts are unique but also similarities between them appear (Jordan et al., 1994, p. 24). Dominant cultural understandings may provide constructs, meanings, and framings that parents can link to their own personal accounts (Eerola et al., 2021; Sevón, 2011; Widding & Farooqi, 2016). Parents may adjust their behaviour to fit a culturally dominant

framing of parenthood, cultural framings may generate role satisfaction among parents, or/and provoke feelings of guilt. Central is that parents are positioned to evaluate/reflect their own behaviours, beliefs, and values (i.e., 'self as a parent') against the dominant sociocultural framings (Eerola et al., 2021; Sevón, 2011). For example, when individuals reflect themselves against dominant discourses, they may perceive themselves to hold characteristics of a 'good parent' which then grants them access to the cultural group of 'good parents' (i.e., role satisfaction) (Widding & Farooqi, 2016). In parental experiences, it is influential how the parent perceives the surrounding context (Valle, 2017), as experiences are not created in a vacuum. Parental experiences may also be linked to what is 'socially acceptable' to experience; for example, it may be a cultural taboo for parents to express negative feelings towards their children (Moore & Abetz, 2018) and negative parental feelings may be labelled as an illness or a condition, such as postnatal depression (Widding & Farooqi, 2016). It may be, for example, culturally normative to perceive children born preterm as 'fragile', and in need of particularly caring parenting behaviour (Tallandini et al., 2015).

Parents execute their roles in the social context of modern life, where interaction with the world is a necessity. Consequently, through this constant interaction and reflection, parenthood becomes an ever-changing socially constructed institution rather than a merely natural phenomenon (Pashkin, 2001, p. 60-61; Valle, 2017). In their interviews with me, the parents aimed to construct their individual identities as parents by reflecting against dominant cultural schemas or framings of acceptable/good parenting behaviour (Jordan et al., 1994, p. 23-24; Lee et al., 2014, p. 31-32; Ogbu, 1981). Eerola et al. (2021) stated that in their study about couples' negotiations of parenting responsibilities, parents reflected dominant cultural expectations of parenthood by 'doing parenting' in their interviews. Subsequently, an 'ideal' abstract interview script could be seen to exist; how modern English parents of EP children would describe their familial composition, day-to-day tasks, their children's characteristics or what they ought to be 'experiencing' at the time their children transitioned to secondary education. However, no story is 'perfect', and the parents discussed varying 'nuances' or 'mishaps' in their accounts, of which the EP birth was apparent. The point of analysis, therefore, was the reflections in the individual interviews between parental experiences and of the social construction of parenthood as illustrated in Figure 5-1 below.

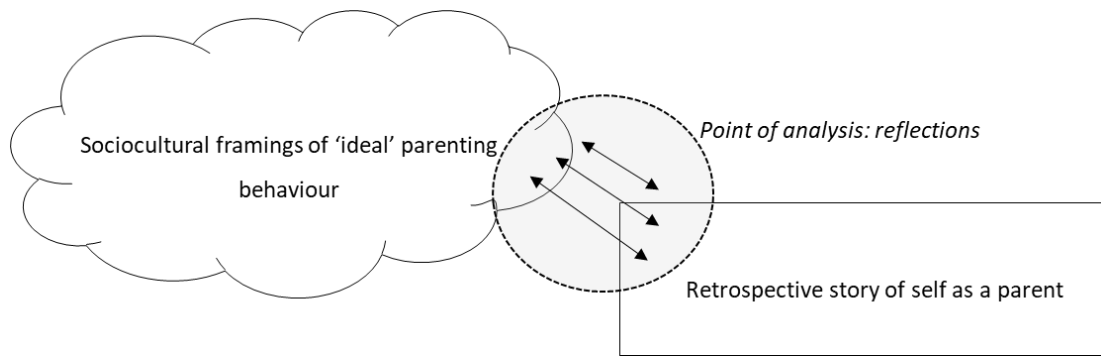


Figure 5-1 Point of analysis in the interviews with parents of EP YA

The rectangle represents a parent interview. The cloud represents sociocultural understandings of parenting behaviour. The circle represents the point of analysis and arrows inside it the reflections in individual parent interviews against sociocultural understandings of parenting behaviour

5.3 Methods

5.3.1 Interviews

The qualitative telephone interviews were conducted between 19 October 2017 and 1 May 2018, in parallel with the main study (EPICure2@11) described in detail in the preceding chapter. I called the parents at a time and date chosen by them, either from a private location at work, or at home. Beside the participant and I, no one else was present during the interview. For the qualitative research strand of this study, parents were eligible if they had consented to take part in the EPICure2@11 Study and agreed to be contacted about the telephone interview, self-identified as the primary caregiver of the child who took part in the main study, and therefore had completed the Parent Questionnaire.

5.3.2 Ethics and consent

The study protocol was approved by the UCL Research Ethics Committee (Ref:10175/001) and by the University of Leicester (Ref:10225). Every parent gave their written informed consent, and their consent was verbally confirmed at the beginning of the telephone interview.

As it was anticipated that the interview topic may be sensitive to some parents, the Participant Information Sheet (PIS) (Appendix F) provided parents with contact details for available support services. The protocol for acting on safeguarding issues disclosed during the interview were detailed in the PIS. I ensured that those parents who expressed hardships during the interview were linked to professional services, and I confirmed that they felt that they received sufficient support. Following disclosure of information on sensitive issues, two interviews

were referred to the EPICure 2 Chief Investigator (NM). In both cases, it was decided that no further actions were needed.

All interviewees were identified with an interview number and were given pseudonyms. The PIS detailed that it was permissible for direct quotes from the interviewees to be used when publishing study findings, but that the interviewees could not be identified from the quotes. To ensure this, and that deductive disclosure would not occur, child sex and parent ethnicity were not presented in the table describing interviewee characteristics.

5.3.3 Design of the purposive sample

The semi-structured telephone interviews aimed at exploring a varied range of experiences of parenting a YA born EP. Since the aim of the analysis was to explore to what extent the quantitative and qualitative results converged; whether the quantitative outcome measure, parenting stress, related to parental experiences, the sampling process aimed to recruit parents with varying experiences. Therefore, the case selection process focused on a range of characteristics of interest that might have signified or encapsulated varying parental experiences in order to gain a maximum variation sample (Teddlie & Yu, 2007). The characteristics chosen were:

- Child sex
- Child gestational age
- Child disability
- Family's socio-economic status
- Parent's relationship status
- Ethnicity
- Parents of multiples
- Parenting stress level (PSI-4-SF)

These characteristics were based on a review of literature, according to which, the level of prematurity or disability may have a gradient effect on family impact (Taylor et al., 2001). Parents from varying socio-economic backgrounds have reported varying levels of family burden (Moore et al., 2006; Taylor et al., 2001). Quantitative studies assessing parent outcomes have focused on the comparability of the groups of parents of preterm and full-term born children, therefore commonly excluding single parents, parents from ethnic minorities, parents of children with severe morbidities and parents of multiples (Treyvaud, 2014). As a

methodological advantage of this study, parents from these groups were purposefully sampled in the qualitative research strand (Teddlie & Yu, 2007).

The aim of the sampling was not to achieve a representative sample or to establish causality, but rather to be able to describe a range of experiences. Therefore, the sample size was aimed to be large enough to achieve a relevant range, but not to represent the population directly. In total I aimed to recruit 20 interviewees, to include:

- Equal proportions of parents of girls and boys
- A sample of parents of children with mild, moderate, or severe disabilities
- A sample of parents of children from lower gestational ages (≤ 24 weeks)
- A sample of parents from varying socio-economic backgrounds
- A sample of single parents
- A sample of parents from ethnic minorities
- A sample of parents of multiples
- A sample of parents with high parenting stress scores

For the purposes of sampling the PSI-4-SF Total Stress, Parental Distress, Parent-Child Dysfunctional Interaction and/or Difficult Child domain scores above 91st centile were considered as high levels of stress, otherwise the stress was of low level. In the categorisation of child's mild, moderate, or severe disability, cognitive performance, vision, hearing, and cross-motor functioning were considered. These morbidities were chosen as they were in accordance with the previous EPICure assessments, as presented earlier (Moore et al., 2012).

It is important to note that the sample design did not assume that these variables would determine the kind of experiences that the parents would have, but rather aimed to elicit a range of experiences. By sampling more parents with similar characteristics, I aimed not only to capture complexity of experiences, but also illuminate varying contextuality (Walsh & Downe, 2006). The case selection was designed as an interactive process, in which topics emerging from the interviews were able to modify the selection process. Yet only one such topic emerged, which was the interviewees' working status (full time, part-time, homemaker/carer/unemployed). In the original range of characteristics for sampling, I had not considered the parents' working status. As a few interviews were completed, it became apparent that this factor was relevant in parental experiences in terms of the time spent in the family context. An advisory panel was consulted regarding the sample design (NM, AL).

5.3.4 Sampling techniques

As discussed in detail in Chapter 3, sampling occurred sequentially for the quantitative and qualitative sections of this study; sampling for the telephone interviews followed the quantitative sample and was a subset of it. To *begin* the sampling, the selection of parents to be invited to take part in the telephone interview employed an element of random sampling to ensure that more parents had an opportunity to participate. From a list of eligible parents, every fifth parent was approached. A recruitment log was kept throughout the data collection and was cross-checked with the sampling target list. Following interviews, the log was updated accordingly, which enabled close monitoring of the recruitment process. Monitoring and modifying the sampling process (i.e., adding parents' working status) helped to minimise under or over coverage (Teddlie & Yu, 2007).

5.3.5 Recruitment process

The parents were recruited to the telephone interviews in a following manner:

- I obtained a list of contact details from the EPICure2@11 Study coordinator for the parents who had given a permission to be contacted about the interview and had returned their Parent Questionnaires.
- I used the above-described sampling techniques to identify the parents to be contacted. I called the selected parents and introduced the interview. I explained the PIS (Appendix B) and answered questions.
- If the parent, in principle, agreed to take part, a preliminary interview date and time were agreed. I explained that the parent would be able to cancel or decline the interview at any time without giving a reason.
- The parent was provided with a copy of the PIS via email.
- I called the parent on the preliminary interview day and confirmed that the parent had had an opportunity to read the PIS, and that they agreed with the content of the PIS. I answered any questions that were raised in relation to this.
- If the parent was present during the child's clinical assessment and I had an opportunity to introduce the interview, it was done then.

5.3.6 Design of the semi-structured telephone interview

This study set out to better understand parental experiences of parents of children born EP in transition to adolescence at around 11 years of age. Parenting stress may have formed an aspect of parental experiences, but other elements were presumed to exist. The qualitative telephone

interview explored those elements. The interview followed a pre-designed topic guide themed around the context of family life, child characteristics, parenting experiences, parents' experiences of the child's transition to secondary school, and parental support. The topic guide is presented in Appendix G in a format it was used in the interviews, including possible prompts and reminder notes for me to utilise during the interview. Table 5-1 below presents the interview topics with key example questions. The topics covered and terms used in the interview were based on review of literature about experiences of parents of children born preterm or/and of children with long-term morbidities, however, the interview was not limited to these topics. Some parents discussed matters freely following the first question relating to their family context and covered questions in the subsequent topics. In other parents' interviews questions were asked in succession. At the end of each interview, I confirmed whether the parent wished to mention something else.

Table 5-1 Semi-structured interview topics and example questions in the interviews with parents of EP YA

Topic	Example questions
Family context	Could you tell me about your family / 'make up' of your family / family's daily life?
Child characteristics	If you were to think about (child's name) how would you describe her/him as a child? If you were to think about (child's name) as a child, is there something that has impacted your parenting?
Transition to secondary school / adolescence	(Child's name) has started secondary school / will start soon, is that something you think about? Do you have any particular ambitions in mind for (child's name)? Are there any challenges in reaching those ambitions? How do you support (child's name) in reaching those goals?
Parental experiences	Are there things that particularly influence your parenting? Do you have priorities as a parent? - something that has always been important for you in your role as a parent? What would you need to achieve these goals / priorities? Is there something that you find challenging in your parenting?
Parental support	What do you feel you need as a parent to assist you through the transition years? Do you receive support in your role as a parent? Do you share responsibilities with your partner? Do you see that you and your partner have similar roles in parenting?

Similar directive semi-structured interview questions have been utilised in previous studies of parental experiences. In their qualitative research component of an RCT of impact of music therapy on parent-child relationship among parents of three to 6-year-old children with ASD, Thompson and McFerran (2015) pre-determined the experience under investigation to be the parent-child relationship, and directed their questions in a semi-structured face-to-face

interview towards parental perceptions of change in the relationship following the music therapy intervention. Hetherington et al. (2010) examined lived experiences of parents of adolescents with disabilities during postschool transition using pre-defined open-ended questions in focus groups with parents. The study aimed to “describe the experiences of adolescents with disabilities and their parents during the transition from high-school to postsecondary education or employment” (Hetherington et al., 2010, p. 165). Predefined interview questions allowed the authors to direct the focus to the particular time of transition (Hetherington et al., 2010). Finally, Scorgie et al. (1996; 2004) were interested in ‘parental transformation’ among parents of children with disabilities and examined this in interviews with parents by using directive questions such as “How has raising your child changed you?” or “What has your child taught you that you might not have otherwise learned?” (Scorgie et al., 1996; Scorgie et al., 2004, p. 87). The authors went on to develop and test a theoretical model of parental transformation among parents of children with disabilities (Scorgie et al., 2004).

When finalising the topic guide, I approached members of the EPICure Parent Group via email to invite their feedback and comments on the PIS and the topic guide. This Patient and Public Involvement (PPI) group was approached as they were parents of young adults born EP and therefore had recently had the experience of parenting a child at the time of transitioning to adolescence. Any comments were invited, but the parents were especially encouraged to comment on the following aspects: 1) when thinking about the experiences as a parent of an EP child, were these the topics most important to discuss; 2) were there any other issues that might be particularly important in parenting a young adolescent born EP and 3) would the parent be comfortable in discussing these topics in the order presented or would there be a particular topic that they felt most comfortable to start. The PPI group parents requested a change to some of the wording in the topic guide to be more accommodating, and to add questions relating to potential sibling impact (under the topics of child characteristics and transition to secondary school, please see full topic guide in Appendix G).

Following the PPI consultation, I piloted the interview with three parents of full-term born children of similar ages as the EPICure2@11 Study participants. Although the topic guide could have been modified during the data collection phase based on experiences gained from the interviews, it remained largely unchanged throughout the interview phase.

5.3.7 Data collection

I conducted all the interviews using my previous experience and training in qualitative interviewing gained during my master's studies as guidance. The interviewees knew that I was also a clinical assessor in the EPICure2@11 Study. I chose the method of telephone interviewing for logistical reasons; to be able to reach parents from the wide geographical area that participants lived. The method of telephone interviewing has been successfully utilised in similar studies among nation-wide samples of parents of preterm born children (Widding et al., 2020). The interviews took place at the time and date chosen by the participants. They lasted 14-65 minutes (excluding study introduction and closing remarks) and were audio-recorded using a password protected digital recorder together with a telephone pick-up microphone. All interviews were transcribed verbatim. Later, parents' quotes were edited to improve clarity. I transcribed the first two interviews, and thereafter a professional transcription service was used. After transcription, if requested, the parents were sent a copy of their interview transcripts. All parents who wished to obtain copies were offered an opportunity to further discuss the interview and to add or change information to enable respondent validation (Lewis & Ritchie, 2003, p. 276; Nowell et al., 2017). Seven parents requested a copy of their interview transcript, but none of them wished to discuss the interview further. The participants did not provide feedback on the findings of this study.

5.3.8 Data analysis

I conducted a thematic analysis of the interview data using an interpretative approach (Snape & Spencer, 2003, p. 17) to identify themes as patterns within the data to analyse and report them (Braun & Clarke, 2006; Nowell et al., 2017). I attempted to "theorize the significance of the patterns and their broader meanings and implications" (Braun & Clarke, 2006, p. 84) in relation to previous literature. This analytic method ensured that the results were (largely) derived inductively from the data through a close engagement with the interview transcripts. I describe my stepwise process below. As stated in comparable qualitative studies of parental experiences among mothers under a threat of preterm labour (MacKinnon, 2006) and among mothers of children with learning disabilities (Valle, 2017), I did not aim to give a complete account of parental experiences of parents of EP YA, but rather, to deepen the current understanding. Interpretations of parents' sense making were more central to my analytic approach than frequencies of descriptions (Moore & Abetz, 2018). As per the qualitative research question of this MM study, the following question was asked from the data: *How parents described experiences of parenting their child born EP during the transition to adolescence at around 11 years of age?*

The thematic analysis was conducted in steps as follows; first, after completing an interview, I noted my immediate thoughts about it, which were usually two to three sentences long. I listened through all the recordings a minimum of once, most commonly two to three times and, while actively listening to search for meanings and patterns (Nowell et al., 2017), I made corrections to the manuscripts if needed. During the re-listening/s, I wrote further notes for ideas for coding, common topics, and reflections against theoretical and empirical literature (Nowell et al., 2017). Both the immediate thoughts and the notes were merged into a document, which was uploaded to NVivo 12 Pro and read repeatedly to support the analysis (Braun & Clarke, 2006; Nowell et al., 2017).

After the above described prolonged engagement with the data, I had familiarised myself with all the interviews (Nowell et al., 2017). I had established a comprehensive sense of what was in the data and what interested me about it (Braun & Clarke, 2006). The manuscripts were then uploaded to NVivo 12 Pro for systematic coding of the entire data set giving equal attention to each data item (Nowell et al., 2017). I identified and grouped interesting features of the data into initial codes of broad descriptions of what was said such as ‘roles of mothers and fathers’ or parental ambitions (i.e., ‘to be or ambitions’, please see Figure 5-2 below). In the MM study of impact of music therapy on parent-child relationship among families with children with ASD, Thompson and McFerran (2015) described a comparable approach to their thematic analysis. While listening and transcribing the interviews, the authors highlighted everything that related to the pre-determined experience under investigation (parent-child relationship) while excluding all other data. Contrary to their approach directed by a specific topic of interest, in the initial coding step, I did not exclude any topic, but sectioned the data to broad initial codes, of which some emerged directly from the topic guide such as ‘descriptions of the child’ (i.e., ‘erm, he or she is...’) while others such as ‘preemie’ followed less directly from the questions asked. The extracts were coded inclusively to retain the context in addition to what was said (Braun & Clarke, 2006). This process ensured that in the next step of generating themes, the whole set of data was considered. Overall, I created 22 initial broad higher-level codes (Nowell et al., 2017). Figure 5-2 below shows the 22 initial codes and Figure 5-3 shows an example of a lower-level coding for an initial code of ‘disability’.

Name	Files	References
about family	22	22
disability	10	20
erm, he or she is...	20	29
fighting for services	7	10
influences	16	22
memories	5	6
our relationship	2	5
outside world	3	4
personal life	14	20
personal parenting	11	15
preemie	10	31
reflections on the study	4	5
role as a parent and change	17	22
roles of mothers and fathers	10	18
sibling	5	8
special needs	12	35
support	19	36
to be or ambitions	20	24
transitioning to secondary school	22	47
you adapt to the circumstances	9	15
your child was different	3	3
work	13	24

Figure 5-2 Initial broad higher-level codes (n=22) in the thematic analysis of interviews with parents of EP YA

Name	Files	References
disability	10	20
challenges	2	2
diagnoses	2	2
early years	2	3
intentional	2	2
limitations in family life	2	2
logistics	1	1
physio programme	1	1
violent behaviour	3	5
worry	2	2

Figure 5-3 Initial lower-order codes for an initial code of ‘disability’ in the thematic analysis of interviews with parents of EP YA

In the next step to search for themes (Nowell et al., 2017), I read and re-read the initial codes to consider how they related to each other and to generate and refine themes from groups of codes (Braun & Clarke, 2006). The broad initial codes assisted me to interrogate the data set more closely (Pickersgill, 2021, p. 604). As an example, extracts from several initial codes such as descriptions of the child, disability, and transition to secondary school fed into preliminary themes initially referred to as ‘effortless / trusting’, ‘explaining different / persevering’ and ‘acknowledging different / resigned’, but which were then during the analysis process further refined to a major theme of ‘parental perceptions of child ability’ (please see Figure 5-5 below for the final coding structure in the thematic analysis of interviews with parents of EP YA). This major theme will be discussed in the results section of this chapter. While generating themes, to aid the analysis, I utilised ‘mind-mapping’ to diagram connections between themes and I repeatedly returned to my notes about the interviews, as discussed above. A reflective journal was kept (Nowell et al., 2017)

Following from the previous steps to further review themes, in a document, I described the emergent themes, such as differing parental behavioural responses, using extensive quotes from the interviews and existing literature to support my interpretations of associations between parents’ experiences and behaviours. Similar to my analytic approach, Wilson and Cook (2018) used both inductive and theoretical approaches (Braun & Clarke, 2006) in their thematic analysis of parental experiences of parents of EP four to 7-year-old children from New Zealand. The authors stated that due to the lack of previous research in the topic area, following initial coding (inductive), they read extensive literature surrounding the subject area, which resulted in varying theoretical concepts to be explored in the interview data (deductive), such as biographical disruption (Bury, 2001) and post-traumatic growth (Tedeschi & Calhoun, 1996). In the present study, the interview data directed my targeted engagement with the literature which then further guided refining of the main themes (Pickersgill, 2021, p. 604).

The emergent themes were validated continuously by discussing them with three senior researchers (AL, NM, ZG) on three occasions, and always returning to the theme review and interpretation process in between. During this process, the relevant data extracts were collated under each theme (Braun & Clarke, 2006). For example, in the theme of parental behavioural responses, sub-themes such as ‘extensive caregiving efforts’ and ‘pushing for normality’ became specific. Finally, the manuscripts were re-read in the NVivo project to confirm that the established key themes and sub-themes indeed fitted the parents’ full interviews, and no large sections of data were left out.

Across the data set of 22 interviews, I identified three interdependent major themes in which the parents reported ambitions for their children and goals for their parenting (parental ambitions / childrearing goals), discussed their children’s characteristics (parental perceptions of child ability), and how they had supported their children to reach those ambitions (parental behavioural responses), in a way in which commonalities within those descriptions emerged. These major themes were distinct but interdependent as parental perceptions of child ability and parental ambitions influenced the way in which parents talked about their parenting behaviour (i.e., parental behavioural responses). This interdependence will be further demonstrated in the results section of this chapter. Figure 5-5 below presents the final coding structure and Figure 5-4 below presents the three key themes and illustrates their interdependence.

Due to the semi-structured interview design, the process of identifying themes was not entirely inductive. The themes of 1) parental ambitions / childrearing goals and 2) parental perceptions of child ability emerged, to an extent, directly from the interview questions. I will return to this potential analytic limitation in the discussion section of this chapter. The third major theme of varying parental behavioural responses emerged inductively from the data. I considered these themes as most important in relation to the research question and my analytic interest, since they captured the overall patterns in the interviews ensuring that the whole coding process was brought into the results (Braun & Clarke, 2006). Thus, they built an understanding of the phenomenon under investigation (Nowell et al., 2017).

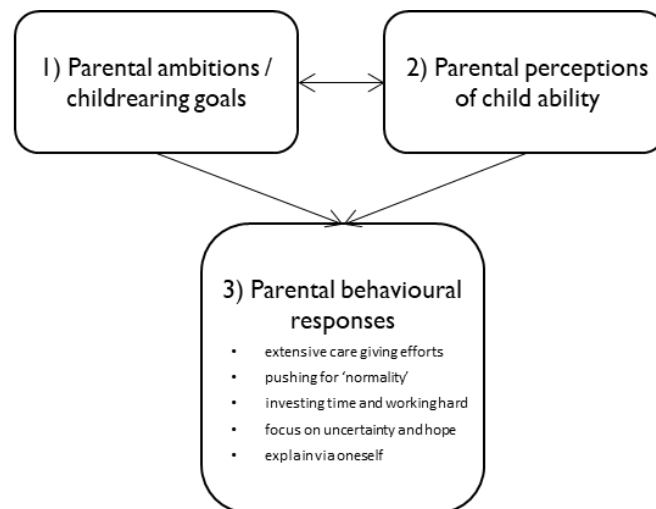


Figure 5-4 Key themes in the thematic analysis of interviews with parents of EP YA

The arrows linking the main themes represent the interdependence between them



Figure 5-5 The final coding structure in the thematic analysis of interviews with parents of EP YA

To achieve conceptual acceptability, an external qualitative researcher (KG) with experience in research among prematurity read four (~20%) interview manuscripts (interview identifiers no. 10, no. 17, no. 20, and no. 29) and validated them against the themes and sub-themes. I purposefully selected these interviews to ensure that the peer reviewed transcripts were 1) conducted throughout the interview phase and 2) because I regarded them as typical examples of the interviews in the study, and 3) as some of them entailed features that I wanted to discuss further with a more experienced qualitative researcher. The findings were discussed and compared with the reviewer.

The external reviewer confirmed that the three major themes were sufficiently clear and comprehensive (Nowell et al., 2017) and that they had an interdependence between them, as I had interpreted. The reviewer described parental ambitions for the child as a *'huge factor'* in justifications of parental behavioural responses. During my discussions with the peer reviewer, parental behavioural responses were deliberated in length. For example, the behavioural response 'explaining via oneself', somewhat a variant sub-theme, was a key discussion point. The reviewer brought up a concept of parental control in this group which I had not initially conceptualised. The analytic result of this discussion and the 'exceptions' in this theme in relation to the other behavioural responses are discussed in detail in section 5.4.2.3.5. At the end of the analysis process, I was satisfied that the main themes and sub-themes represented the interview data as a whole and told a coherent, fitting story (Braun & Clarke, 2006; Nowell et al., 2017).

5.3.9 Reflexivity and considerations of validity

As will become apparent later in this chapter, the findings from the interviews were reflected against the framework of parental determinism as a dominant cultural determinant of parenting behaviour. Like Wilson and Cook (2018), following the initial coding process, I read extensively around the subject area. During this phase, the theme of varying parental behavioural responses appeared to relate to the framework of parental determinism. As discussed earlier in relation to the research methodology, and further detailed in the introduction to this chapter, a social constructionist viewpoint was utilised to understand the interview data. Parents were thought to execute their roles in a social world surrounded by dominant discourses such as attachment theory, importance of the parent-child relationship, intensive parenting practices, and parental determinism. Even though the analysis was not readily structured and coded to a given framework, the findings were reflected against the concepts of parental determinism and the critique of intensive parenting culture. The overall aim of the study was to place findings in lived experiences, and therefore, the rationale was to explore them using concepts from dominant discourses, similarly as Widding and Farooqi (2016) explored the discourses of attachment and 'involved parenthood' in the retrospective birth stories of mothers of EP children. A limitation was that this study and the topic guide in itself, were also a part of the 'dominant discourse'. For example, the topic guide portrayed parenting as 'doing' and not 'being' and assumed that the daily life as a family would include evening activities and family events. Therefore, when interpreting the findings of this investigation, it should be considered that to an extent, the parents were invited to reflect their experiences against the dominant discursive understanding of modern Western parenting, and

the fact that these reflections related to the dominant discourses did not emerge purely inductively from the data.

Overall, in this study about parental experiences, I was studying the unfamiliar. A researcher's personal 'insights' into the topic under investigation, or a sense of being 'part of the group', may be regarded as beneficial when designing the study, forming research questions and/or approaching participants (Berger, 2013). While conducting this study, I had no personal experience of 'parenting', nor had I been 'parented' within the English sociocultural context as I was born and raised in another country. Consequently, I was unable to claim being a member of the group of 'English parents' or 'parents of EP children' or 'parents of children with disabilities', and the parents in this study were left with the expert position.

Nevertheless, the position of an 'outsider' may have been beneficial. As I was unable to assume knowledge of 'parenting', I perceived myself as more likely to ask for clarifications during the interview (prompts such as "what do you mean by that" or "could you give me an example") and I rarely assumed shared understanding. Similarly, owing to my experience of being a 'foreigner', I had been accustomed to the practice of 'dismantling' concepts to learn/understand their meanings. I utilised this learned practice more systematically in this study in regard to the sociocultural concepts discussed. The relatively open research question to explore parental experiences among parents of children born EP during the transition to adolescence, facilitated this approach.

My disciplinary knowledge in Nursing and my previous research experience in the field of prematurity helped me to understand the health conditions relating to prematurity and the healthcare context that the parents sometimes referred to during their interviews. Perhaps due to the longitudinal nature of EPICure 2, on occasions, the parents assumed that I had previous knowledge of their child's health and development. For example, referring to previous diagnoses as "as you probably know [child name] was diagnosed with [diagnosis] three years ago...". In these instances, I corrected the parent, explaining that I did not have previous individual-level information about any of the children in the study.

In addition to being the parents' stories, the interview accounts also reflected me. As discussed earlier, the lens I used to filter the information given by the participants was affected by my own multi-disciplinary background. It influenced the way I chose to interpret the parents' accounts; what elements in them interested me and what ideas I chose to prompt. Thus, ultimately the meanings presented in this study were co-constructed (Kohler Riessman, 1993, p. 5). For example, during the interviews I tended to ask questions relating to work outside the

home and division of labour within the family, as these were areas of social life that have particularly interested me. However, as per the research question, I did not report these discussions in detail in this thesis, as they were not the primary focus of the study. Another interview study among parents of EP and VP born children in Sweden explored parental experiences relating to the birth, NICU experience and childhood, resulting in several smaller analyses with varying theoretical focuses (Widding & Farooqi, 2016; Widding et al., 2020; Widding, Hagglof, & Farooqi, 2019). Similarly, I intend to maximise the output of this study by describing the main findings in this thesis, but, at a later date, further explore the data with a particular focus such as the division of labour within the family.

Due to my positionality, as discussed throughout this thesis, how the interview accounts were constructed interested me as much as what was said. Thus, the choices made in the thematic analysis also reflected my interests. Another investigator might have focused on a different aspect of the data. With the analysis, I have presented the overall construct of parental experiences in the interviews and drilled into varying parental behavioural responses.

Overall, doctoral degree is a lengthy process. As time passed, I felt that I moved more and more towards the expert position and I was able to assess my previous actions and decisions more critically, such as those relating to the topic guide and the sampling process for this qualitative research strand. When employing a critical sociological approach to examine a topic, it is important to remain critical to one's own thinking. I was not revealing an 'absolute truth' behind the previous 'misconceptions', but that this study, in itself, was a part of the phenomenon under the investigation. Similarly, it is important not to 'distort' the emergent image for the purposes of advocacy or argumentation. During the analysis and interpretation of the findings, I was careful not to overemphasise parental challenges, and not to place critique where it did not appear. I believe that comparison with quantitative data, reflective discussions with senior researchers, and lengthy direct interview quotes all supported the validation of the findings of this study (Nowell et al., 2017).

5.4 Results

The analysis I present here, reveals the social construction of 'parenting' that occurred in the parents' accounts of their experiences when they discussed their parenting roles, actions, and decisions. Hence, it will give a presentation of what is from a sociocultural point of view constituted as 'parenting' in modern English society in general, and among parents of EP YA in particular. As discussed above, this presentation is in itself a version of many possible presentations and additionally reflects my own background as a young Western female

researcher with an interest in critical sociology. When direct quotes are used, I presented short quotes in text *'in italics between apostrophes'* to indicate that the used terms were chosen by the parents. I placed longer quotes in their own clearly distinct paragraphs together with interviewees' pseudonyms. Following each distinct direct quote, the interview pseudonym and identifier (a number), child disability level (none/mild; moderate; severe) and the type of school (junior; secondary; special educational needs (SEN) school) are presented in brackets.

Although the semi-structured telephone interview covered several topic areas, generally four interview questions yielded most of the discussion; 1) how the parent perceived the child, 2) what ambitions the parent had for the child's future, 3) how the parent had supported the child to reach those ambitions, and 4) potential challenges parents may have experienced in reaching their ambitions. A majority of the analysis I present here focused on these four questions.

5.4.1 Interviewees

Overall, I approached 28 parents. I was unable to get in contact with six after three attempts. None of the contacted parents declined to take part in the interview. Thus, 22 parents completed the interview. To include parents from varying backgrounds, I interviewed two more parents than the planned pre-defined sample size of 20 interviews. All the parents who were contacted and consented to take part were women. They were approached as they had completed the Parent Questionnaire and therefore provided the parenting stress data. In addition to the 22 mothers, one father wanted to take part in the interview together with his partner. The interviewees were parents of 23 EP YA (one set of twins).

The interviewees' characteristics are presented in Table 5-2 below. A majority (14/22) of the women were in their forties, four mothers were above the age of 50, and three below the age of 40. One parent did not report their age. The interviewees were mothers of 12 children with severe or moderate neurodevelopmental disabilities of which three attended SEN schools. Two children were born less than 24 weeks' gestation. Most parents (15/20) had one to two other children to care for in the family. A majority (14/22) of the interviewees were either married or living with a partner. Ten parents had university level education, five had some post-secondary education, five had completed secondary education, and the remaining two did not report their education. In most of the families (13/22), the highest income in the household was either high or intermediate. Nonetheless, 16 interviewees either worked part-time in paid employment or were full-time carers or homemakers, and therefore not in paid employment.

Table 5-2 Interviewee characteristics by interview ID in a study of parental experiences among parents of EP YA

Interview ID	Pseudonym	Age	Relationship status	Highest academic qualification	Working status	Highest SES in the household ¹	GA in weeks	Child disability	School type	Siblings	Total Stress centile ²
1	Ms Beryl	≥40	married	post-secondary	carer/ homemaker	not in employment	≥24	severe	SEN	none	68
2	Ms Turquoise	≥40	married	university degree	carer/ homemaker	high	≥24	severe	SEN	1	94
3	Ms Chalcedony	≥40	married	secondary education	part-time	high	≥24	none/mild	secondary	none	≤1
5	Ms Sillimanite	≥40	married	post-secondary	part-time	high	≥24	none/mild	secondary	2	33
6	Ms Iolite	≥40	married	secondary education	part-time	high	≥24	severe	junior	1	96
7	Ms Kyanite	≥40	single	university degree	carer/ homemaker	not in employment	≥24	none/mild	junior	1	94
9	Ms Garnet	≥40	married	university degree	carer/ homemaker	intermediate	≥24	moderate	junior	1	80
10	Ms Quartz	≥50	living with a partner	secondary education	part-time	low	≥24	none/mild; moderate	junior	1	65
11	Ms Fire Agate	≥40	single	university degree	part-time	low	<24	severe	junior	2	.
13	Ms Ammolite	≥40	married	university degree	part-time	intermediate	≥24	severe	SEN	2	96
16	Ms Amethyst	.	married	.	full-time	.	≥24	none/mild	junior	2	.
17	Ms Amazonite	≥40	separated/divorced	post-secondary	full-time	high	≥24	none/mild	secondary	none	19
18	Ms Onyx	≥40	single	secondary education	carer/ homemaker	not in employment	≥24	moderate	junior	1	78
19	Ms Agate	≥40	separated/divorced	university degree	part-time	intermediate	≥24	moderate	secondary	none	77
20	Ms Spinel	≥40	separated/divorced	secondary education	full-time	intermediate	≥24	none/mild	SEN	1	83
21	Ms Dioptase	≥30	married	university degree	part-time	high	≥24	none/mild	junior	1	10
23	Ms Sphalerite	≥50	living with a partner	university degree	full-time	high	≥24	none/mild	secondary	none	46
24	Ms Rhyolite	≥40	single	university degree	full-time	self-employed (no other details)	<24	moderate	secondary	1	42
25	Ms Jasper	≥50	living with a partner	post-secondary	full-time	high	≥24	none/mild	secondary	none	68
27	Ms Drusy	≥30	married	.	part-time	low	≥24	none/mild	secondary	3	.
28	Ms Aquamarine	≥30	married	post-secondary	carer/ homemaker	low	≥24	severe	junior	1	77
29	Ms and Mr Jade	≥50	married	university degree	carer/ homemaker	high	≥24	moderate	junior	1	22

¹ Derived using the National Statistics Socio-economic Classification (NS-SEC) simplified method: High = Higher managerial, administrative, and professional occupations; Intermediate = Intermediate occupations; Low = Routine and manual occupations (Office for National Statistics, 2016); ² PSI-4-SF; derived using Abidin (2012); the 84th centile and above is considered as high parenting stress

5.4.2 Overall construct of the interviews and central themes

In this results section, I will present the overall construct of the interviews demonstrating how parental ambitions and the parents' perceptions of their child's ability to reach their ambitions guided how they built their interview accounts. I will then further explore how the parents described varying parental behavioural responses to support their children to reach their parental ambitions. I labelled these behavioural responses as a) extensive caregiving efforts, b) investing time and working hard, c) pushing for normality, d) focus on uncertainty and hope, and e) explain via oneself.

5.4.2.1 Theme 1: Parents' childrearing goals and ambitions for their children

Although heterogeneity and complexity in parents' goals for their children appeared, universally all interviewed parents wanted their children to be happy. Their ambitions as parents were for their children to be sociable, independent individuals who are *'happy and carefree'*. The concept of a 'goal' is intrinsically not yet achieved. Even though growing up and changes towards more or less independence were topics present in all parents' interviews, what their children would be was not yet achieved. Their goals existed as hopes of what might come. The parents felt responsible (parental responsibility) and accountable for (felt important to account their actions/decisions) their children to reach their goals and during my interviews with them, described how they had supported their children.

For a majority of the parents their ambitions comprised their children being physically and emotionally healthy, showing confidence, and being social. They saw few limits in goals for their children and stressed the importance of supporting their children in *'whatever they want to be'*. A sense of 'trusting' in what the future will bring for their children allowed parents to 'take a step back', and they commonly emphasised that their children were *'making their own choices'*. Although children's academic skills were a point of discussion, it was important that parents supported their children and did not *'expect anything'* from them as specific achievements. Having control over or 'pushing' for their children to achieve particular goals were perceived as undesirable

...I suppose for both of them to have a real sense of belonging and to feel really loved and accepted. No matter what choices they make as they grow up, I think that really matters to me. I think for them to be healthy physically and mentally, to me, that's more important than anything. So, I suppose I want her to do well at school, but more than that I want her to be happy. So, I would try to avoid putting too much pressure on. (no. 23, Ms Sphalerite, none/mild, secondary)

...whatever she chooses to do I will encourage and support her. (no. 5, Ms Sillimanite, none/mild, secondary)

...I hope that academically she is able to go and do whatever she wants to do in life and that she gets right qualifications to be able to do that and that she is happy really which she is... (no. 3, Ms Chalcedony, none/mild, secondary)

The descriptions of parents' ambitions for their children became more extensive when the parents saw their children having challenges in reaching independence. Despite these parents did not foresee their children becoming independent, for example being able to live on their own, their ambitions still were for their children to be *'happy and carefree'*, and they placed extensive efforts into caregiving. Although referring to the same framework of wanting their children to be happy and sociable, the parents additionally focused on discussing *'independence'*, *'functioning'* and helping their children to *'achieve their best across the board'*. The descriptions of independence and *'functioning'* focused on particular tasks such as communication and self-care

So, my wish for her going forward is for her to be able to communicate and also if she's able to communicate then to be able to see to her personal care. (n. 13, Ms Ammolite, severe, SEN)

The goal kind of is that she achieves that she is capable of achieving kind of across the board and that she is happy, if she is happy and healthy then I am happy [laugh]. (n. 1, Ms Beryl, severe, SEN)

..just for him to be independent. He still needs simple instructions for doing things and forgets things and you need to tell him one thing at a time. (n. 20, Ms Spinel, none/mild, SEN)

...and hopefully one day, he'll be of a level where you know, he can live independently. That's what I'd like. (n. 19, Ms Agate, moderate, secondary)

Some parents whose children experienced challenges presently but who perceived their children could reach independence in future had additionally set particular academic achievements as goals, such as *'getting into university'* or going to college, which they perceived would showcase *'competence'* (please also consider Ms Kyanite's discussion further below on page 200)

...when it comes to going to college, I think that's the way that I'd push him. (n. 6, Ms Iolite, severe, junior)

5.4.2.2 Theme 2: Parental perceptions of child ability

Parents who foresaw no challenges in their children becoming sociable, independent, and happy adults described their children's characteristics and how they had supported their children effortlessly. In these effortless descriptions the children had special interests such as music or arts, good friendships were a strong focus, and parents described aspects of growing up, such as becoming more independent or taking part in activities with friends such as football and cooking and baking

...we tend to have a lot of her friends over, it is kind of quite nice to do girly stuff, they bake, or they make creative things [...] I suppose I do spend quite a bit time with her and her friends here. (no. 5, Ms Chalcedony, none/mild, secondary)

[child name] does football after school on a Wednesday and archery and fencing which is on a Thursday after school. (no. 18, Ms Onyx, moderate, junior)

...she's very sociable, she really likes to be with her friends. (no. 23, Ms Sphalerite, none/mild, secondary)

...she's very independent. She goes to school, she comes home. She's almost 12. She's very confident to go and come back by herself. (no. 17, Ms Amazonite, none/mild, secondary)

The effortlessness of the interview accounts of some parents did not, however, follow from the effortlessness of being a parent per se, as these parents also discussed hurdles in their roles as parents. Nevertheless, they believed that despite potential challenges their children would grow up to be independent and happy adults. Ms Garnet and Ms Onyx both had children with moderate neurodevelopmental morbidities. Both parents reported heightened, but not high levels (<84th centile) of parenting stress. In their interview accounts, they expressed concerns about limited parental support and felt that they had to 'push for support' for their children which they would not receive without their parental efforts

I wish I have that cousin, friend as parent, we have some friend through supplementary school but, sometimes I am worried about because she's, her body is not so strong, because she's not eating very well. And she's using hearing aid sometime make me so nervous when I think about it... (no. 9, Ms Garnet, moderate, junior)

...it always seems to be me doing all the pushing and shoving and I don't think there's much out there for us, as a parent [...] I think if we didn't push for things, it's only because I'm constantly battling all the time. (no. 18, Ms Onyx, moderate, junior)

Regardless of experiencing challenges, neither Ms Garnet nor Ms Onyx doubted the future competence of their children and trusted that their children would grow up to be happy, social, and independent adults. Neither of them perceived their parental efforts to be the key factor in guaranteeing their children's future competence, but rather that the 'successes' would originate from their children

...he does try his best, he really does try his best and he will say, when I get bigger, I'm going to stay on at school an extra year, because my nephew, he stayed on an extra year, and he's going to university, and I think with help and support, I think if that's really what he wanted to do, I think if he puts his mind to it, he'll be able to do it. (no. 18, Ms Onyx, moderate, junior)

...and I always said [child name] I feel you will be successful, and you'll be a great person. And she said, even if I am working as a cashier, I said, yes, take five steps, you be a manager ((laughs)) (no. 9, Ms Garnet, moderate, junior)

As seen in the examples of Ms Onyx and Ms Garnet, rather than following from the ease of being a parent, the effortlessness of the interview accounts of some parents rather followed from the general ease to describe behaviours and events that the parents perceived as 'typical'. In these effortless accounts parents believed they were discussing matters that every parent would discuss, and they wanted to assure me that what they described was nothing '*out of the ordinary*'. I will discuss this concept of being 'typical' later in relation to the parental behavioural response of 'pushing for normality'.

Some parents who foresaw their children becoming independent adults volunteered information about their child having been born EP, even though this was not an interview question. Among these parents, an initial sense of *worry* following from the *unexpected* EP birth had shifted to a sense of relief as their children had met their developmental '*milestones*'. These parents described feeling *lucky*, *glad*, *grateful*, and *fortunate* in relation to their children's outcomes and did not, for example, attribute these to their own parenting efforts

I think in terms of birth and how unexpected it was and how poorly she was obviously that was really traumatic experience, and we didn't know what to expect, what was going

to be the outcome short term or long term [...] when she was about two and a half probably, we kind of could see that she was meeting all of her milestones, so the pressure was kind of off. Till that time, I was very concerned, she did not walk until she was nearly twenty months and thinking is this going to happen at times. As all of those things were kind of ticked off if you like then we knew that all was okay. I feel like from then on everything has just been as a normal family and parenting is I don't really feel like we have had any extra pressures. (no. 3, Ms Chalcedony, none/mild, secondary)

...we have been very fortunate you know I guess prematurity just hasn't been a problem for her, she went through all the milestones (no. 21, Ms Dioptase, none/mild, junior)

...we kept an eye on him when he was younger, to make sure that he wasn't getting out of breath and that he could keep up, but he's perfect in fact and we're just so lucky that [child name] has walked away with nothing... (no. 25, Ms Jasper, none/mild, secondary)

I always think I'm so glad she's growing up big and strong when I think how tiny she was ((laughs)) (no. 23, Ms Sphalerite, none/mild, secondary)

Yet parents described that regardless of knowing that '*all was okay*', they were still conscious of what their child had *gone through* in relation to being born EP, which, against their best efforts, had on occasions altered their parenting behaviour

...as a preemie she grew up normally, but you always have at back of your head she's that preemie child. I think that's why sometimes I feel a bit soft with her. Although I'm trying to be really hard, but at the same time I'm like I can't be too hard ((laughs)). (no. 17, Ms Amazonite, none/mild, secondary)

...we were both very aware not to think that or make her special and sort of being over parenting her which I think we were both very aware of... (no. 3, Ms Chalcedony, none/mild, secondary)

As a few parents' quotes above have indicated, the parents' general focus was the running of their children's day-to-day lives; they facilitated their children partaking in various activities and supported them in maintaining friendships. However, when asked about their children's characteristics, parents who did not foresee their children becoming independent placed more focus on describing their children's behaviours and abilities rather than their personality features or peer relations. The descriptions included both positive and negative behaviours.

Their children growing up was commonly seen as a challenge, as behaviours became harder to 'control' and the children deviated further away from the socially expected behaviour. Ms Spinel described personal sacrifice in a challenging family context. Her child had mild disabilities (a diagnosis of cerebral palsy and cognitive deficits) following EP birth. When asked about her child, she spoke openly about her perceptions of his '*demanding*' character and occasionally aggressive behaviour

The first word that came into my head was demanding. He is very, when he's in a good mood, affectionate. He is very loving. When he is not and he's having one of his meltdowns, he can be - in fact, he is very clingy anyway and he always needs reassurance. So, he is very, I don't know what word to describe it. Very needy. He is very demanding. When he's not in a good mood, and he's having a meltdown, or something is not quite right, then, he's very angry, aggressive, not very nice really [...] But he has got probably worse as he's got older, and obviously the bigger he's got, the worse it gets for me and his brother, because he's bigger and stronger. (n. 20, Ms Spinel, none/mild, SEN)

Following from her son's challenging behaviours, Ms Spinel's personal life had become limited

[child name] can't be left at home on his own. So, he will have to come everywhere with me, which then brings on more tantrums and more strops. The older he gets, hopefully, with a few life skills he will get more independent, and I might get a bit more time, but I honestly don't know. At this moment in time now, I see him being at home forever. (n. 20, Ms Spinel, none/mild, SEN)

Regardless of the challenging family situation and perceiving her parenting role to continue indefinitely, she concluded

I mean it's like any child I'm sure, he's got ongoing, long-term disabilities. So, it's juggling hospital appointments and things like that- it's difficult and trying to factor it in with everything else, [...] He's challenging but he's worth it, on a good day ((laughs)). (n. 20, Ms Spinel, none/mild, SEN)

5.4.2.3 Theme 3: Parental behavioural responses

The aim of raising a sociable, independent, and happy child and the parents' perceptions of their child's ability had influenced their parenting behaviour. The interviewed parents described varying parental behavioural responses to support their children. Five overlapping behavioural responses emerged which I labelled as a) extensive caregiving efforts, b) investing

time and working hard, c) pushing for normality, d) focus on uncertainty and hope, and e) explain via oneself, following from the behaviours that the parents described.

5.4.2.3.1 Extensive caregiving efforts

Some parents responded to their children's challenges by placing more time and efforts into supporting their children. Extensive parental contributions towards their children were most striking when the mothers did not foresee their children becoming independent. Regardless of limited expectations for the future, the mothers sought to find 'extra' activities for their children. Thus, the extensive caregiving efforts were not merely a response to more complex care needs of these children. Ms Turquoise's child had severe disabilities, needing fulltime one-to-one assistance in all her self-care needs. She was her daughter's only carer and described her tasks in a following manner

...I think with a child like this you get stuck in time ((laughs)) so for the last eleven years I have been changing diapers and I don't foresee that I would stop doing that anytime in the future, so sometimes I just seem like I am a new parent, I spend the same sleepless nights and same around the clock things ((laughs))... (n. 2, Ms Turquoise, severe, SEN)

Despite a seemingly hopeless description, a central focus in her interview was the varying activities that she had organised for her daughter, such as special needs swimming lessons and horseback riding and adult-assisted cycling. She described herself as a 'Google expert' after researching extensively online for varying activities. Additionally, as a family, they would have been 'quite happy to move', in order for her daughter to get into the 'right kind' of secondary school. When asked about her ambitions for her daughter's future Ms Turquoise continued

...I think our only ambition at the moment is can she come slightly more independent, although that said it is very difficult to foresee and I guess if you try to forecast something you get worked on by the possibilities so no ((laughs)) however ruthless it may sound no I don't have any ambitions for her I just want her to be happy and care free, that's it. ((n. 2, Ms Turquoise, severe, SEN)

Parents of children with limited independence were not attributing their challenges to 'causes', such as the preterm birth. Instead, their accounts featured a sense of 'going along with it', which appeared as an awareness that their extensive efforts would always be required. Their

own efforts or their child's characteristics, however, would not ensure future competence. Ms Spinel described her son's transition to secondary school in the following manner

I felt like if I hadn't have pushed as much as I did - if I'd have been a parent that had just gone along with it, then I wouldn't have got where I am now and I constantly had to fight and push for everything with them, with both schools, the one he was leaving and the one he was going to. But then that's been the case all his life to be fair. I feel like I've had to stay on top of everything and pester. (n. 20, Ms Spinel, none/mild, SEN)

As Ms Spinel's previous quotes have demonstrated, regardless of her 'pushing for support' for her son, she did not believe her efforts would result in his future accomplishments. A similar sense of 'complying' appeared in Ms Aquamarine's interview. She was a mother of two. Her older EP child had severe physical and mental challenges and she described how they 'fell' further behind in development compared to her younger child born FT. Ms Aquamarine found it somewhat challenging to describe how she supported her child's daily activities

I don't know 'cause I do it every day. It's just everyday life. It's just having to keep telling her to get dressed or remind her to clean her teeth and constantly, just have to keep telling her before she does it, but I just do that every day so I'm kind of used to it [...] She seems to have got harder work as she's got older. But I suppose we've always done it, so I suppose it's no different. (n. 28, Ms Aquamarine, severe, junior)

Even though focusing on supporting her daughter, she concluded on a sense of limited ambitions for her daughter's future

Just to be - really try and become independent. Maybe not rely on us quite so much, but we'll see. It'll be depending on what she wants though. She just wants to work at Sainsbury's though... (n. 28, Ms Aquamarine, severe, junior)

As the extracts above have demonstrated, the challenges that the parents with children with limited independence described were commonly linked to the extensive caregiving they provided, and to the thoughts of their parental responsibilities continuing 'indefinitely'. A similar 'extensive caregiving efforts' theme was also present in parents' accounts whose children experienced challenges with independence presently but who anticipated that they would succeed in the future. Yet these parents perceived their efforts as temporary. Due to the parents' belief in reaching their ambitions for their children, they expected their parenting efforts, although extensive, to eventually cease. Consequently, although expressing a sense of

'giving up' at present, Ms Kyanite, for example, foresaw a possibility to return to work in future

I think this year and next year it could be a little bit of a - more of a storm. But then if the outcome of it is that she gets the support and help then actually maybe, just maybe, I'll be able to go to work. (n. 7, Ms Kyanite, none/mild, junior)

5.4.2.3.2 *Investing time and working hard*

Parents who perceived their children having challenges in being happy, sociable, and independent, but who believed they would reach independence in future, used the most rhetorical speech work to describe their children, justify their ambitions for them, and the choices and actions they had taken to support their children in reaching their goals. All parents in these descriptions focused on explaining to me how their child, and therefore the context for parenthood were different from the 'norm', as opposed to ensuring me of 'typicality'. Some parents had responded by 'working hard'. Some parents' stories were themed around uncertainty and hope of what might come in the future. Some parents linked their children's challenges into their own identities or had created alternative identities for their children, such as 'an artist' or 'a loner' to explain why they were not *'like the other children'*. Some parents described 'pushing for normality'. All these parental behavioural responses will be further discussed below. Even when describing similar extensive caregiving efforts, contrary to Ms Turquoise, Ms Spinel, and Ms Aquamarine above, these parents believed that with their extensive caregiving efforts their children would grow up to be independent individuals. They perceived their parenting efforts as necessary for their children to reach adult competence.

For many of these parents the EP birth had merely been an 'unlucky' event that had set them back in the route to raising a happy sociable independent adult, which now meant that they had to work 'extra hard' to guarantee their children's success. Ms Kyanite was a single mum of two. She had relocated her family and given up a fast-paced, high responsibility job to stay at home fulltime to ensure that her child would be able to *'function'* and *'manage school'*

...but a large part of my job, a large part of my life really is spent ensuring [child name] is okay. That's the majority of the time, in terms of the reason that I have to do what I do. [...] All the routines have to kind of be in place to make sure that she's able to function, but it is really kind of about her functioning. Just to get her to a point where she's sort of well. (n. 7, Ms Kyanite, none/mild, junior)

How '*functioning*' would be reached was by investing time and 'putting the work in'. Ms and Mr Jade had adopted similar behavioural responses to Ms Kyanite

Mum: We've worked very hard to almost train her if you like, with regards to the autism, to behave in a socially acceptable way. She understands that. We've invested a lot of time and we've been committed as a family to that, and I think she's benefitted enormously from that [...] I suppose I want her to prove everybody wrong. I think when people meet her, they're quite amazed by her, and what she is able to do because considering the tragic start she had to her life. (n. 29, Mr and Ms Jade, moderate, junior)

A contrast between Mr and Ms Jade's and Ms Kyanite's accounts was the utilisation of the concept of prematurity. How Mr and Ms Jade described their daughter to me, her being born preterm was a central theme. They perceived prematurity as a '*tragic start*', incontestably causing challenges in future life. However, by being '*committed as a family*' and investing time they were able to change the course of prematurity and '*prove everybody wrong*' (including themselves). Although employing similar approaches, contrary to Mr and Ms Jade's interview, to Ms Kyanite '*prematurity*' was not a central theme. Only in relation to her daughter's better than average school success, she mentioned

...you know, which considering all her difficulties is pretty amazing. Considering how early she was born and all the different diagnoses that she's got, it's pretty good that she's got to that point... (n. 7, Ms Kyanite, none/mild, junior)

However, without employing the term '*prematurity*', a thought of '*difference*' was present in Ms Kyanite's descriptions. Ms Kyanite perceived her daughter to be something that she was not '*supposed to be*' and by investing her time now, similarly to Mr and Ms Jade, she was able to correct the situation and eventually '*no one will know*'. She hoped for her daughter to go to university

I: ...is there any particular ways that you try to kind of support her to go to university?

That's what I'm doing. I've given up my career to enable her to be able to go to university. Basically, she has to learn everything. It takes a lot longer to learn everything. It has to be very repetitive, and it has to be very structured and there has to be a lot of explanation. And that is what I'm doing, it's not just about university but it's about her functioning on her own in the community. I see myself as doing this until she's eighteen, and when she's eighteen, do you know what, she'll be all right. No one will know. She'll

function okay, and she'll have the basics [...] if I put this work in now for the next seven years, she's gonna have a life. Where she can actually be the person that she's supposed to be. (n. 7, Ms Kyanite, none/mild, junior)

The concept of 'supposed to be' might have referred to Ms Kyanite's loss of what she had hoped her child to be (Landsman, 1998), as well as to a reference to what a 'normal' average child 'should be' (Lee et al., 2014, p. 41-44; Ogbu, 1981) as well as to parental efforts to 'repair' the effects of the risk exposure of EP birth (Kantrowitz-Gordon et al., 2016).

As discussed at the beginning of this results section, children's acceptable social behaviour and success in social relations also featured as parental ambitions. Parents referred to their children's ability to '*get along with peers*' or have '*good quality friends*'. However, many parents described increased challenges in their children's social behaviour and acceptance by peers

I: You mentioned that she's very bubbly.

Mum: Yeah, very. Doesn't stop talking any time.

I: Is it with the family or also with friends and other children?

Mum: The problem she has with other children is that she doesn't understand her peers now. When they talk about things, it goes totally over her head. At school I can't really say there's anybody close that she's really friends with... (n. 28, Ms Aquamarine, severe, junior)

Although both Ms Kyanite and the Jade's perceived 'investing time and working hard' as the main means to achieve their ambitions for their children, they expressed concerns about factors that might challenge their efforts, such as their children's relationships with their peers as they approached adolescence. The parents felt that acceptance by peers and friendships were the areas where they were least able to influence. Ms and Mr Jade acknowledged that regardless of the time that they had invested in their autistic daughter's future, challenges with friendships could eventually prevent her from reaching what they had hoped for her

Mum: We invested a lot of time in [child name]. So, we hope that she will - I don't know. All we want is for her to be happy at school, to have some friends, and be able to do most of the things that children of her age group do. And to support her as best we can with that.

I: Are there any particular challenges in reaching those ambitions, anything that you worry about?

Father: Friendship side of things is a big deal. As they grow up for all autistic children, it's their understanding of other people

Mum: The social interaction.

Father: Yes, that, and as she has gone through primary school, it's become more of a worry to us and we know that as she gets into secondary school, that will become more of an issue. Children will be less tolerant of somebody different as they get older. They are less likely to potentially invest the time in [child name]. That is a constant worry [...] We do understand that there are plenty of things in the way, not just academically. There are plenty of things in the way that mean [child name] may not reach the potential that she possibly could. (n. 29, Ms & Mr Jade, moderate, junior)

Yet the challenges experienced in social interactions with peers were not always perceived to originate from the child characteristics, but the parents saw that external barriers could also make their efforts in vain. Ms Agate, a single mother of a child with cerebral palsy, felt *'little heart-broken'* that regardless of her supporting her son to develop his social skills, he lacked friendships because of hesitation from other parents. In a way to mitigate the harm, Ms Agate ensured me that her son's life was *'full enough'* without friends

...I've always worked on the basis that if you're not going to be the smartest kid in the class, you can be the nicest. He makes friends easily, but they're not what I would call best friends, or close friends. He seems to know a lot of people, but he doesn't like bringing them around and he doesn't get invited to their houses and parties and things like that. I think that's purely because he's got issues and the parents are more afraid of it. Then we've got a close-knit family, so it doesn't really affect him in that respect, but you think sometimes, it would be nice for him to go to somebody's party [...] I mean, sometimes it can be a little bit difficult and a little bit heart-breaking. Particularly when he's formed a friendship with someone at school, and his parents have sort of like, not stopped it, but not encouraged it. So, you just think, well fair enough, we'll get on with it. We're not bothered. We do enough and his life is full enough that he doesn't need it. (n. 19, Ms Agate, moderate, secondary)

5.4.2.3.3 *Pushing for normality*

As discussed earlier, in some parents' interview accounts the concept of 'typicality' appeared as the parents told me that what they had experienced was 'typical'. For example, a parent concluded her description of their family's daily life as '*yes, normal family*' or described her child as '*just your typical 12-year-old*'. Nevertheless, the concept of 'normality' also had another meaning. It was closely linked to the parental experience of 'pushing' for something they hoped for, for their child (e.g., 'pushing for support') and thus, related to parenting behaviour. Parents described how they did not want to '*give up*' on normality and '*pushed*' for it with an aim of making their child '*as normal as possible*'. How normality was determined was by visible external indicators (Rhodes, Bernays, & Houmoller, 2010), such as the child having friends at school, having special interests or hobbies, and succeeding in her/his school work. Ms Iolite's interview followed the latter stream. She felt that because her son, despite his challenges, was '*quite bright*', if she did not '*teach him to act like an average child*' she would be '*giving up*' on his potential

I suppose challenging trying to ((sighs)), I know it sounds weird, but make him as normal as possible. Teaching him how to act like an average child. I don't like saying normal. Teaching him to, I suppose, be his age. He's an eleven-year-old who, most of the time, acts like an eight-year-old. His interests are very young. I'm constantly trying to push him to grow up a little bit. So that's a challenge ((laughs)). That's hard [cos...] he likes what he likes, and he is the way he is. I suppose I don't want to give up on the fact that he is an eleven-year-old, he should be acting like an eleven-year-old. I'm a bit mean sometimes, I have to say, but I don't go, "Aw, okay, you've got cerebral palsy. I'll just forgive you [...] Or, "You're behaving like a, a seven-year-old," you know. And I also say to him "I don't care if you're disabled" "you're gonna do this. I don't care" "if you've got cerebral palsy. You're going to be like this". I just want him to understand how to, not understand, but I'd like him to grow up in the same way, as much as the other children in his year are and have the same interests, because I just don't want him to always be so far behind. I know it's a difficult one because he has got mental problems. It is hard. I'm probably trying to do something impossible. (n. 6, Ms Iolite, severe, junior)

Ms Iolite's contemplation demonstrates well the intersections between child characteristics, parent's perceptions of child ability, and parental ambitions. Similar to Ms Kyanite and Ms and Mr Jade discussed above, Ms Iolite seemed to have a strong understanding of her role as a parent. Simultaneously, despite these parents' children experiencing challenges, the parents perceived them as being 'nearly like others', which further provoked them to 'work harder'. Commonly these parents compared their EP children's abilities to those of healthy siblings or

classmates (as Ms Iolite above), not adjusting expectations due to the preterm birth. This comparison further provoked their parental efforts. As Ms Iolite also tied her own personality and experiences from her childhood into her understanding of a 'good parent', the possibility of 'giving up' would not only have been detrimental to her son's future, but also at odds with her own self-identity

I grew up in a very relaxed household where I was kind of left to do what I wanted to do. My parents weren't strict at all. So, it's really hard for me cos I want to kind of not make the same mistakes as my parents made which was just not to think about my kids' future. They didn't really think about what we were gonna do in the future. They just let us get on with life. Whereas I would've liked my parents to have gone, "Oh look, she's interested in [activity], so let's pursue that." But they never did. Whereas I think the things that influence me as a parent are looking at my children and looking at what they like. And pursuing that. I think that makes me a good parent in the way that I want my kids to have every opportunity that I didn't and follow their dreams. With [child name], it's more difficult because I don't really know what he wants. But with my [other child], their interests are what pushes me as a parent [...] I always just want to do the best for my children. (n. 6, Ms Iolite, severe, junior)

In the extracts above, Ms Iolite described a tension between her wish to support her children's interests and her son's developmental delays. Her son's limited abilities challenged her own ability to be what she perceived as a 'good parent'. Along with accepting child disability, these parents were also required to adjust their own perceptions of themselves as parents. Ms Agate reflected on her changed parenthood

...when I found out I was pregnant, I was going to be this wonderful parent, that had time for this, time for that, time for the other. We were going to do colouring and all the rest of it, but life didn't work out like that. And you adapt to the circumstances that you've got. So, if there are days where you know, he's a bit unsteady and it's raining and we can't get out so much, then we don't and we have our buffet out days and things. I'm still giving him as much fun as I want it to be. Like I managed to get him to Disneyland in Paris. (n. 19, Ms Agate, moderate, secondary)

5.4.2.3.4 *Focusing on uncertainty and hope at the time of transition*

Parents discussed their current or past concerns in relation to their child's transition to secondary school, for example expressed feeling a 'bit apprehensive'. Parents discussed their children's characteristics referring to them being 'ready' based on physical and/or mental characteristics. Choosing and being accepted to the 'right school', being able to visit the new

school, commute to the school, and whether their children would ‘find their way’ in the new environment, and make friends were also discussed. Parents described supporting their children by ensuring that they had their books with them, practised the new route to the school, drafted timetables, and homework schedules with their children, and followed their new timetables on mobile apps

They just don't look right, because they are big lads, and maybe in stature they're ready. But I'm not sure if they're ready mentally, I don't know, but it will be a big shock. But I can't do anything about that. They have to go and that's it. I just have to worry about it ((laughs)) (no. 10, Ms Quartz, moderate & none/mild, junior)

...we do have to travel a bit further. But now that we've had our place and we've spoken to other parents, the parents of children that are going are the parents that are willing to travel that extra mile... (no. 16, Ms Amethyst, none/mild, junior)

...I must admit, I get her to pack in the night before and when she is not looking I do have a little check to make sure that she has everything... (no. 3, Ms Chalcedony, none/mild, secondary)

Concerns were similar but experiences differed depending on whether the children had started secondary school at the time of the interview, commonly expressing a sense of relief.

It's been brilliant. The school has been absolutely fantastic. They've been totally supportive of him. They've got a great TA programme in place, and he's really settled. I thought we were going to have a really bad transition, but he was just so ready to go. (n. 19, Ms Agate, moderate, secondary)

As discussed earlier, parents of children who experienced challenges with independence also discussed concerns of securing appropriate support for their child in the new school. Generally, the transition process was more labour-intensive for parents with children with special needs. Yet children transitioning to secondary school was commonly perceived as an opportunity for gaining more independence. In some instances, parents linked their expectations for the secondary school into the interlinked ‘uncertainty’ and ‘hope’ that they had experienced in relation to their children’s outcomes after being born EP. Ms Fire Agate’s son had severe cognitive deficits. He also suffered from medical conditions, which meant that he needed constant supervision from an experienced person in case of emergency. He was one of her

three children that she cared for alone. Throughout the interview without apprehension, she discussed her son's challenges with independence

... I think it's a wait and see. I mean, he's growing up, but he is not as independent as I hoped he would have been at this age [...] I have to be with him all the time because of his medical needs, because of his development. I mean, at eleven years old, my eldest [child], [they were] walking to school alone. [child name], he still needs an adult with him all the time. So that level of dependency from him is more or less the same. I haven't seen it changing. Although now he's able to tell you if he's in pain. (n. 11, Ms Fire Agate, severe, junior)

Even though witnessing little change in her son's level of dependency, when asked about her ambitions for her son's future, Ms Fire Agate continued hopefully

...I'd like him to work on something of his interest [...] I mean, secondary school is very important for him because he will find out his interests in the future. He will align himself in something of his interest, either hand crafting, [word], computers, something that will be good for him in the future. So, I'm looking forward to secondary for many reasons... (n. 11, Ms Fire Agate, severe, junior)

For Ms Fire Agate the uncertainty of her son's developmental outcomes had nurtured '*optimism*' and hope for a '*turning point*', after which she could return to her own life

...and now he's going to secondary they've advised for him to be diagnosed, to be known if there's any underlying thing apart from the normal. Because now he's not being linked to anything [...] I think it's reached a time now; I just want to know. I want to plan for my future. Will he need me or is he going to have a turning point, or will he need me for much longer than how it is now, so that I don't have this optimism. I have this optimism within me that I will catch up with whatever I was pursuing at some point. (n. 11, Ms Fire Agate, severe, junior)

Similar to Ms Fire Agate, Ms Rhyolite's interview followed the themes around uncertainty of future outcomes and concerns relating to independence. Different to Ms Fire Agate, Ms Rhyolite did not focus on a '*turning point*', but rather saw her son as having an alternative identity, being '*different from the other children*', which had given her a reason for an optimistic future. She simultaneously acknowledged his challenges

I can see the difference really with his peers and that he does struggle at school still [...] he says I can't think properly, I'm not like the other children, and I'm like, you

actually think differently... (n. 24, Ms Rhyolite, moderate, secondary)

She portrayed a conflicting image of her son when she discussed his extraordinary interest in arts

He's very forward thinking and he's quite focused on what he wants, and he's taken part in quite a number of projects [...] He's really focused. He's kind of driving me with that. (n. 24, Ms Rhyolite, moderate, secondary)

With this alternative identity and evident challenges at school as driving forces, Ms Rhyolite saw her primary task to be to ensure that her son got appropriate support for secondary school. She had '*mass referred*' him to a variety of specialists resulting in five different diagnoses in the past year

I'm having to direct a lot of that. So, it is more worrying, and it's also more time consuming. I just thought, thank god, I work for myself, because the amount of paperwork and because off all the documents and then getting advice from SENs and solicitors and other people [...] I feel like I'm constantly like his caseworker. ((laughs)) (n. 24, Ms Rhyolite, moderate, secondary)

After internalising responsibility and functioning as her son's '*caseworker*', she felt that if she failed to get the appropriate support for him, he would '*miss out on things*'

... I think the thing that I'm trying to avoid with SEN plan an all this sort of stuff, I don't want him to miss out on things, because he was born early and because things are not being put in place, I want him to have the same chances as every other child... (n. 24, Ms Rhyolite, moderate, secondary)

5.4.2.3.5 *Explaining via oneself*

Accounts of parents who focused on discussing their children's challenges via their own characteristics differed from the other, rather practical, parental behavioural responses presented above. Rather than focusing on describing extensive caregiving efforts, parents' interpretations of past life events and how they saw their own upbringing having influenced their parenting behaviour were interwoven into their aim of reaching their childrearing goals. How parents perceived their children and what ambitions they had for them were linked to the parents' own identity, which in some cases, served as an explanation for their children's potential challenges

She's shy. She's very shy, but she's very bright at the same time [...] You talk to her, and she will take time to acknowledge you or answer you. I don't know if she was like that with you, but it's her personality. I think she took that from me and her dad. We're kind of like both on the shy side, so she's also shy... (no. 17, Ms Amazonite, none/mild, secondary)

In the extracts presented above, while discussing their children's future competence, Ms Fire Agate, Ms Rhyolite, and Ms Amazonite all acknowledged their children's challenges and approached these in varying ways. Ms Fire Agate and Ms Rhyolite felt that parenting their children had altered their own personalities, making them more *'assertive'* or *'patient'*. Contrary to these women's approaches, Ms Quartz, a mother of twins, focused in her interview primarily on how her own upbringing and personality had made her the parent she was; her perceptions originated strongly from her own identity instead of her children's characteristics

My dad brought me up on his own. He was a single parent, along with my brothers ((sighs)). He was very calm. [...] But when I was their age [referring to her children], I kind of looked back how he was and draw on that a little bit, but because I don't have a mum now, I've never really had that kind of guidance from that kind of person to say, well do it this way, do it that way. I've had to find my own way, and people would tell me, you know what, you're brilliant. You're so good with them. (no. 10, Ms Quartz, moderate & none/mild, junior)

Having brought up her EP twins virtually alone had given her a sense of achievement. She perceived it as a success that she interwove into her own upbringing, her self-identity, and how she perceived them as a family. When asked about potential challenges in her parenting, she continued

Maybe if you asked me when they were born, when they came home, what on earth, how am I supposed to do this? I've got no parents, partner is at work. I've got no brothers or sisters. I've got friends but they're at work. They've never had twins and they've never had prem twins, I wasn't frightened of failing or something because I wouldn't have done that. That's not in me, that's not how I've been brought up, but I wasn't scared of [children's names]. I was aware the situation was real, and this is a massive deal, and there's only me to do it. But looking back now, I did it so well. I did. Probably at my own, what's the word I'm looking for? Without considering myself sometimes, even now, the way I feel or if I'm upset or wound up or angry, but it's tough. You've got to get on with it, every single day. And that is how we are. (no. 10, Ms Quartz, moderate & none/mild, junior)

Parents, such as Ms Kyanite or Ms Rhyolite presented above, had adopted behavioural responses to their children's challenges that had given them an element of control. By investing time, working hard, or by promoting their child's interests, the parents were able to work towards achieving the goals for their children and to ensure that their children would *'fulfil their potential'* and not *'miss out on'* anything in comparison to children with no special needs. As a contrast to the parents presented at the very beginning of this results section, such as Ms Sillimanite (page 192), parents such as Ms Kyanite or Ms Rhyolite with increased parental control rarely discussed their children *'making their own choices'*. Their parental ambitions for their children, despite being born EP, were seemed to have been set high such as a university education or a driven career as an artist. Contrary to this, although holding on to the same sense of control, Ms Quartz' benchmark for 'success' was different. When discussing her ambitions for her children's future she gave an impression of adjusted goals

I think out of universities and things, to me personally, I think you're best off getting a job. [...] Get a bit of a job, do something, and have some money for yourself. (no. 10, Ms Quartz, moderate & none/mild, junior)

As an additional difference, she perceived her parenting role to continue 'indefinitely', as did parents of children with limited independence. However, contrary to the other parents, Ms Quartz approached it via her own identity instead of her children's characteristics. She portrayed an image of 'adjusted independence' for her children, which would be her personal 'ideal'

...I'll say, you'll always have a place - you don't have to leave home. I will look after you, I'll support you, but obviously you have to have your role in life and have independence for yourself. Provided you're doing that, then you don't have to leave home. I will look after you as your parent. (no. 10, Ms Quartz, moderate & none/mild, junior)

This adjusted goal might have been influenced by her children's developmental characteristics. Even though, throughout her interview, she highlighted how proud she was of their remarkable growth after being born so *'tiny'*, when discussing school support, at the very end of her interview, she mentioned

You cannot be born that prem, have these issues and problems and be tip top. It's going to leave you with something, and like I say now, if you met [children's names], they are big, strapping lads, they are 11. However, when you talk to them, their speech isn't clear. [...] There is a difference in them and another 11-year-old, little things like they can't

tie a shoelace... (no. 10, Ms Quartz, moderate & none/mild, junior)

By considering her children's potential challenges with independence, Ms Quartz had interwoven her own upbringing and identity as a parent to support the adjusted ambitions she had for her children. Even though hoping her children to 'never leave home', bearing their abilities in mind, her behavioural response, and the adjusted independence that she perceived as 'ideal', might be supportive to her children's future competence as social adults. Ms Quartz saw herself as an 'ideal' parent. Similar to Ms and Mr Jade presented above, despite placing extensive caregiving efforts towards their children, and facing frequent challenges, the sense of being in control and having achieved what they themselves perceived as 'ideal' parenting, might have in part explained the low levels of parenting stress that these parents reported (please see Table 5-2 above).

5.4.2.3.6 *Internalised sole parental responsibility*

As the discussion and the extracts presented above have demonstrated, the interviewed parents had generally accepted their self-defined parenting roles. Regardless of how they perceived their children or the context for their parenthood, in their interviews the mothers rarely questioned their responsibilities for childcare and housework

...in general, my life revolves around her if she's got something on whatever I've got on doesn't happen. (n. 1, Ms Beryl, severe, SEN)

As much as I would like to work full time, with the amount of hospital appointments and things that we go to, it's not possible for me to do that [...] I had a career, and obviously when [child name] came along, I had to give that up to care for [child name]. (n. 19, Ms Agate, moderate, secondary)

The interviewees' internalised responsibility for their children was strongly gendered. With a single exception of a two-parent interview where the father had presumed the primary caregiver's role, the main carers participating in the interview were women. A majority (16/22) of the interviewed women worked part-time (n=9/22) or were full-time homemakers or carers (n=7/22). Six parents worked full-time of whom one had a child with moderate disabilities and one whose child attended a SEN school. The parents who were not in paid employment or worked part-time often highlighted their partners' financial responsibilities. Ms Iolite described her partner's role

It's mainly financial. He deals with the – cos it's keeping us all alive ((laughs)). Bringing the food in. (n. 6, Ms Iolite, severe, junior)

Consequently, for those mothers with children with disabilities, assuming sole responsibility for caregiving was more challenging. Consider how Ms Iolite described her daily life

Most of my time is taken up with work and looking after [child name]. He has a lot of therapies and a lot of appointments that I take him to. So that takes up a lot of my time as well. And obviously looking after the house ((laughs)). Washing and cleaning, shopping, and cooking. ((Laughs)) The usual stuff for a mum. My husband isn't working at the moment. He's off work and sometimes he works out of the country. I'm just kind of left on my own a lot of the time [...] It's not easy but you just get on with it. Get through it. (n. 6, Ms Iolite, severe, junior)

In extreme instances the gendered responsibility for the child's wellbeing continued beyond death

...She'll always need an adult to care for her. So, my fear is that I'm not alive to give [child name] that care. Because I'm her primary carer, even though dad does help as well when I'm not available, I'm still her primary carer, so that's my major fear. That if I'm not there what will happen to [child name]. Who will take care of her? Because I seem to know [child name] best and she seems to obey me more, do things for me more than she does for her dad. (n. 13, Ms Ammolite, severe, SEN)

5.4.2.3.7 *Recall of EP birth and the hospital period*

The semi-structured interview schedule did not include explicit questions about the EP birth. Instead of setting the birth as a starting point for the interview, the topic guide gave an opportunity for 'prematurity' to emerge spontaneously from the parents' accounts. As seen in the previous quotes throughout this results section, 'prematurity' featured in parents' interviews in varying manners. In addition, a few parents recalled the EP birth and hospital experience describing it as *unexpected* and *traumatic*, a '*lifetime experience*' that had given them a '*very different perspective on life*'. This related to the earlier descriptions of '*softer*' parenting behaviour following EP birth. For example, a parent recalled her response to a teacher's feedback of her child's slow progress: '*we just think, well he's here. He's happy*'. A parent remembered how she had a decade later, declined an offer to function as a peer support person for new NICU parents: '*I can't go back there, because it's still a scary place*'. Parents described the care that their surviving, *thriving* child had received as *excellent* and *incredible*.

Yet their personal experiences were perceived as separate from their children's experiences, that often had been challenging

...yes, they looked after the babies, yes they were medically supported and everything else, but there was no support as such, for parents. (no. 10, Ms Quartz, moderate & none/mild, junior)

...it wasn't my experience; it was more, well it was my experience as well, but they looked more after the baby [...] in terms of helping her to blossom it was good. (no. 17, Ms Amazonite, none/mild, secondary)

At the end of the interview, when I asked if there was something else the parent wished to mention, a few parents discussed the experience of giving birth EP. Ms Amazonite, a single mother of one, described her birth story in length. It had started unexpectedly with pains that she recalled the medical personnel had '*undermined*'. Later she had experienced a feeling of being in labour which was dismissed by the staff caring for her, as '*it was not possible*'. In her recollection, the severity of the situation had changed in a '*split second*'

I wasn't thinking, "I'm having a baby," I was like, "I'm losing a baby." [...] it broke me, and I said, "I'm not having any more babies." I just - yeah, the whole experience was just so bad. (no. 17, Ms Amazonite, none/mild, secondary)

When asked when the feeling of 'losing a baby' had changed for her she continued

...when I carried her for the first time I knew I had a baby [...] and once I started ventilating her with that little tube myself, that's when that feeling started to grow into I'm a mum rather than being a woman without a child... (no. 17, Ms Amazonite, none/mild, secondary)

A central feature in her adverse birth experience had been the feeling of uncertainty of her child's survival. She discussed how she had not felt '*safe*' until after the discharge from hospital

I never felt safe at all. Because you are always looking around and I seen a baby much bigger than [child name] and the next day she wasn't there. And you're thinking, "How can that one that's like twice smaller survive?" (no. 17, Ms Amazonite, none/mild, secondary)

Similar to Ms Amazonite, a few other parents referred to the hospital period recalling their concerns with 'bonding' with their EP children

...there is a group of us who became friends through the neonatal [hospitalisation] and years later, I bumped in to [child name] mum and she said, she had counselling and I can understand where she was coming from, because it was difficult because you don't have that, straight away, touchy feely with a baby, do you? I mean it must have been three weeks before we were allowed to touch him. (no. 25, Ms Jasper, none/mild, secondary)

...Right back in the beginning when they were born tiny you never think you're going to be able to hold their hand, to cuddle them (no. 10, Ms Quartz, moderate & none/mild, junior)

5.5 Discussion

In this qualitative research strand of a convergent MM study, from the interviews with 22 parents of YA born EP three main themes were identified with respect to parents' experiences at the time their children transitioned to secondary education: 1) Parental ambitions / childrearing goals, 2) Parental perceptions of child ability, and 3) Parental behavioural responses. The findings from this study demonstrated that parental ambitions and the parents' perceptions of their child's ability to reach adult independence guided how they described their parenting behaviour. The analysis also showed that parents who believed their children would become happy, sociable, and independent adults were generally trusting about their children's future and did not perceive extensive parental involvement in their children's lives as desirable. Parents who anticipated their children to have challenges with future independence described varying parental behavioural responses to support their child's development. These parental behavioural responses featured elements from the framework of parental determinism. The findings from this study also indicated that, although parents discussed their children being born EP, the birth was not a central feature in parental accounts at the time their children had reached early adolescence. Aligned with the dominant discursive framing (Lee et al., 2014, p. 51-101), overall in the interviews, 'parenting' was described as a set of actions (i.e., doing rather than 'being'), and the interviewees had internalised the sole parental responsibility for their children. For parents with children with long-term morbidities, having sole parental responsibility was demanding.

5.5.1 'Parenting' to achieve future adult competence

My analysis demonstrated that parental ambitions and the parents' perceptions of their child's ability to reach adult independence guided how they described their parenting behaviour. This meant that the interviewees accounted for their 'parenting' by describing how they had supported their children (and would support) to reach their goals (Jordan et al., 1994, p. 23-24). The interviewed parents described similar childrearing goals; parents wanted their children to grow up to be happy, sociable, and independent individuals. Comparable childrearing goals have been reported in previous studies (Suizzo, 2016). The parents' childrearing goals could be regarded as descriptions of a culturally valued adult competence; what these parents in Ogbu's (1981) terms perceived as "making it" (p. 420) in a modern English society. Arising from the parents' belief of their child's ability to 'make it', three groups of parents emerged; 1) those parents who believed their children would become happy, sociable, and independent individuals, 2) those parents who did not foresee their children becoming independent adults, and 3) parents who perceived that although their children were experiencing challenges presently, with extensive parental efforts they would reach happy,

sociable, and independent adult life. The importance of the child ‘making it’ explained parental actions, and thus, gave consistency and continuity to the interviewees’ accounts of themselves as parents (Giddens, 2013, p. 3-5; Jordan et al., 1994, p. 23-26).

In their interviews, parents discussed their children’s future skills and their own parenting behaviour to support their children to achieve those skills. This was aligned with the general framing of ‘parenting’, discussed throughout this thesis, as a set of actions to socialise the child to reach adult competence (Maccoby, 1992). Following from this framework, the hypothetical ‘future’ can, paradoxically, be regarded as present. As was evident in the parents’ accounts, the particular future adult skills that were valued were influential in parenting behaviour in the present (Ogbu, 1981). Commonly, aligned with parental determinism, parents are portrayed to hold a unique direct stake over ‘the future’ through their actions as parents and as goals for their children (Rosen & Suissa, 2020). For an individual parent, this may appear as a sense of opportunity of what might be in ‘the future’, as was the general framing in these interviews as parents perceived that what their children would be was not yet achieved. For example, Ms Fire Agate hoped for a ‘*turning point*’. Yet the future may also pose a risk of materialisation/realisation of poor child outcomes. This may challenge parents’ perceptions of the successfulness of their ‘parenting’, particularly if they perceive that their children will not achieve those culturally valued competencies and be in a “futureless” position (Rosen & Suissa, 2020, p. 131). The interviewed parents were concerned that, as their children approached adolescence, new developmental demands jeopardised what had already been achieved, such as in relation to increased complexity of peer relations described by Ms and Mr Jade and Ms Aquamarine (pages 202 and 201, respectively). As discussed in the introductory chapter of this thesis, parenting behaviour can be a response to the presence of risks in that parents direct their actions towards ‘preventing’, ‘managing’ or ‘mitigating’ risks in their children’s lives (Lee et al., 2014, p. 10-15). For example, Ms Quartz’ had turned to adjusted independence (please see page 209). I perceived that, in addition to the parent, child and family factors, this interplay between the socioculturally influenced parental ambitions and the demands of ‘the future’ was the context to which the parents responded to with varying parenting behaviours.

5.5.2 Parental behavioural responses in the context of parental determinism

Findings from this study showed that parents who believed their children would become happy, sociable, and independent adults were generally trusting about their children’s future that they attributed to their child’s characteristics rather than to their own parenting efforts (e.g., ‘*if he puts his mind to it, he’ll be able to do it*’). This finding suggested that, contrary to

the intensive parenting culture that presumes that intensive parental involvement in the child's life is necessary to maximise cognitive and socio-emotional outcomes (Schiffrin et al., 2014), these parents did not perceive their parental actions as 'essential' in ensuring their children's future success as adults. Rather, they described their children '*making their own choices*'. In comparison with parents of FT adolescents, VLBW parents have reported more intensive parental involvement (Pyhala et al., 2011). Yet, similar to the findings of the present study, in a quantitative study of intensive parenting practices among a general population sample, 181 self-selected mothers of children below the age of five who responded to an anonymous online survey of intensive parenting practices and mental health, agreed least with statements about their role as a parent being 'essential' in their child's life. Respondents who perceived that they were the 'essential' caregiver reported more stress and less life satisfaction (Rizzo, Schiffrin, & Liss, 2012). In the present study, the children's older age might have further contributed towards parents' 'trusting' experiences. As the children were approaching adolescence, the parents may have felt it developmentally appropriate to be less involved. Aligned with this, Yerkes et al. (2019) reported that among British families, the prevalence of parents who reported intensive parenting practices reduced when children grew older. Yet, regardless that the parents in the present study did not perceive their roles as 'essential' in ensuring their child's future success as an adult, in general, the interviewed parents described child-centred parenting practices in that they were intensively involved in their children's lives by facilitating their children's friendships and managing their schedules for school and hobbies. Contextual factors, such as an increased hesitance to allow even older children to play without adult supervision, may have provoked parents' involvement in their children's social lives (Dodd & Lester, 2021; Füreidi, 2008, p. 8).

The analysis demonstrated that parents who anticipated their children to have challenges with future independence described varying and somewhat overlapping parental behavioural responses to support their child's development. These parental behavioural responses echoed elements of parental determinism. For example, parents who described 'investing time and working hard' perceived their parenting behaviour as a means to promote their child's outcomes long term (Kanieski, 2010). Ms Kyanite stated that her main task was to ensure her child's '*functioning*', whereas Ms and Mr Jade discussed being '*committed as a family*' and to '*almost train*' their child to behave in a socially acceptable manner. In both interviews the abstract goal of the child 'making it' was influential (Ogbu, 1981; Rosen & Suissa, 2020) and the parents described a belief that with their parenting efforts they would be able to ensure that, despite challenges, their children would become sociable, independent, and happy individuals. These parents perceived their parental behaviour to have direct causal impact on future success for their child (Dermott, 2012; Lee et al., 2014, p. 2). These perceptions were

aligned with previous findings reported among families with preterm born children. In a qualitative study of psychological and social responses of parents of younger VP born children (≤ 7 years), Kantrowitz-Gordon et al. (2016) found that parents in their study went to great lengths to support their children's outcomes (e.g., financial sacrifices). The parents aimed to compensate for and 'repair' their children's developmental challenges and described that their intensive parental efforts were aimed for their child to achieve 'normality' in comparison with aiming to achieve particular developmental goals. In their study, 'parenting' had caused the participants prolonged personal distress. Findings from the present study suggested that comparable intensive parental efforts among EP parents may continue till early adolescence.

Parents' behavioural responses to their children's developmental challenges may have also reflected the consequent implication of parental determinism that with 'enough' (i.e., more) parental efforts, the child's future competence could be guaranteed (Shirani et al., 2011). For example, Ms Kyanite's child would secure a place at university. Intensive parenting behaviour allowed these parents to mitigate perceived risks posed by EP birth (i.e., the child would become what they were '*supposed to be*'). Consequently, these parents were able to frame 'the future' as an opportunity rather than as a risk in relation to their children's outcomes (Rosen & Suissa, 2020), as discussed earlier. Although a general hypothesis in intensive parenting culture has suggested that more (and earlier) parental efforts would correlate with improved child outcomes (Gillies, 2020), as discussed earlier in the introduction to this thesis, this has not been demonstrated (Schiffrin et al., 2014; Widding, 2014; Yerkes et al., 2019). On the contrary, among younger children, intensive parenting practices have not resulted in improved cognitive child outcomes in comparison with less intensive parenting behaviour (Schiffrin et al., 2014), and among adolescents, highly involved parenting behaviour has been associated with poor adolescent psychological health (Schiffrin et al., 2013). Among the parents themselves, intensive parenting practices have been associated with poor mental health outcomes (Rizzo et al., 2012), while increased child care-dependency has been linked with reduced parental HRQoL (Hatzmann, Maurice-Stam, Heymans, & Grootenhuis, 2009). These multiple risk factors, together with parental perception of a necessity to 'work hard', may in part explain high parenting stress scores reported by some of the interviewed parents.

By 'investing time and working hard' these parents may have gained a sense of control over their children's outcomes (Yerkes et al., 2019). Yet internalising sole parental responsibility may have also posed a risk to the parent's 'self-identity' which may have further promoted or sustained the need for intensive parental efforts. An interdisciplinary qualitative study among 36 prosperous British couples explored how the partners accounted for their decisions regarding parental roles and responsibilities in the family. In line with Giddens' (2013)

presentation of self-identity as a constructed retrospective story of the self, the couples described their decisions as individual ‘choices’ they had made to ‘prioritise’ the family. Since the self-identity was sustained by the continuity and internal consistency of the account, these ‘choices’ were needed to be perceived as ‘worth it’ which was demonstrated by the wealthy status of the family (i.e., successful family) (Jordan et al., 1994, p. 22-26). Similarly, in the parents’ interviews, decisions such as to leave paid employment to focus on ‘parenting’ may be understood as a ‘choice’. Ms Kyanite explicitly stated that she had ‘*given up*’ her job to ensure that her child would get to university. She hoped to return to work after this goal had been achieved. In addition to Ms Kyanite’s wish to ensure her child’s future competence by securing them a place at university, her parental efforts may have been further promoted or sustained by the sense of them needing to be ‘worth it’ (i.e., child to ‘make it’) as alternative opportunities had been ‘*given up*’.

Contrary to the parents who did not anticipate their children to reach adult independence, parental perception of child characteristics as ‘nearly like others’ may have further provoked parental efforts to ‘push for normality’ (i.e., ‘*push*’ their child to achieve age-appropriate skills) as future adult competence was perceived as achievable (Valle, 2017) (please consider Ms Iolite’s example on page 203). Parents who experienced uncertainty in relation to their child’s abilities may have found it challenging to adjust to the altered parenting context, as their perceptions of themselves as parents and their perceptions of their children may have remained ambiguous (Scorgie et al., 2004). For these parents, the ‘point of realisation’ (or ‘*turning point*’) had not yet occurred (Valle, 2017). Contrary to studies in which parents of children with disabilities have described ‘rebuilding’ their identities as parents of children with morbidities (Landsman, 1998), some parents in the present study were still expecting a change in their child’s abilities and for them to achieve ‘normality’.

To further reflect the sub-theme of ‘pushing for normality’ against previous reports, as discussed earlier in the review chapter of this thesis, previous quantitative studies assessing long-term family burden following VP birth have indicated that families with VP born children with subtle developmental or social difficulties may experience more challenges when their children approach adolescence in comparison to families with children with severe disabilities (Moore et al., 2006). Parents of ELBW adolescents with neurosensory impairments have reported their families to function more favourably in comparison to parents of ELBW adolescents without impairments (Saigal et al., 2010). Findings like these may reflect the experiences of parents in this study who perceived their children’s characteristics as ‘nearly like others’, and thus, responded by ‘pushing for normality’, which may have resulted in a more challenged parenting context. Yet evidence has also indicated that parents of preterm

born children may perceive their children's health and behavioural characteristics more critically in comparison with teacher or adolescent reports (Wolke, Chernova, et al., 2013). Thus, the parental perceptions in this study may have also reflected critical comparisons as well as actual child characteristics. Nonetheless, as indicated by the findings of this study, parental perception of child ability is an important factor as it can impact parenting behaviour. Parental perceptions of child ability have previously been found to impact parents' own emotional health (Indredavik et al., 2005), their parenting behaviour and the parent-child relationship (Thompson & McFerran, 2015) as well as levels of reported parenting stress (Larkin et al., 2021).

As discussed earlier in this thesis, due to the reciprocal nature of the parent-child relationship, parent and child outcomes are interlinked. Despite the extent of parental efforts, child factors, such as cognitive or behavioural difficulties, may prevent the parent from achieving their goals in the dyad (Maccoby, 1992). Some interviewed parents experienced that their child's characteristics hindered them from being what they perceived as a *'good parent'*. Ms Iolite was unable to support her child to pursue their dreams due to their cognitive status, and Ms Aquamarine expressed on behalf of her child what she with her caveat of *'just'*, perceived as relatively low expectations for her child. These experiences can be related to the earlier discussion of how parental perception of *'futureless'* child position may position parents to examine successfulness of their parenting (Rosen & Suissa, 2020). These parents' perceptions of themselves as parents and the actions they perceived that were available to them were limited by child factors (Scorgie et al., 2004). Yet, despite limiting child factors, parents may have turned to intensive parenting practices as they perceived them as *'good parenting'* (Wall, 2021). Ms Iolite for example acknowledged that she perhaps attempted something *'impossible'* when she *'constantly pushed'* her child to behave age-appropriately. Aligned with the rationale of parental determinism, studies of intensive parenting practices, such as Wall (2010) exploring mothers' internalised responsibility for their child's *'brain optimisation'*, have found that parents may internalise responsibility for factors that they, in reality, have little or no control over, such as challenging adolescent behaviour (Barnes & Power, 2012). Increased parenting efforts may impact parent outcomes such as stress and anxiety (Rizzo et al., 2012; Wall, 2010).

5.5.3 EP birth and altered parenting behaviour

The findings from this study indicated that although parents discussed their children being born EP, the adverse birth experience and potentially disrupted early parent-child relationship (e.g., concerns about *'bonding'*) were not common features in these parents' descriptions at

the time their children had reached early adolescence. The research hypothesis that EP parents may perceive their children as vulnerable long term following from the high-risk birth and uncertainty around infant survival (Miles et al., 1998), was less evident in these parents' interviews. This finding may have been a result of several factors. Contrary to the present study, studies on parental experiences following preterm birth have generally set the birth and the hospitalisation as a starting point for their investigations. These studies have invited the parents to recount their birth stories (Kantrowitz-Gordon et al., 2016; Widding et al., 2020; Wilson & Cook, 2018). Wilson et al. (2018) reported that four to seven years after the birth, parents of EP children measured their current quality of life against the NICU period. Yet, the children in their study were younger and the parents described that their life was still "approaching normal" (Wilson & Cook, 2018, p. e1632). In the present study, by the time of early adolescence, some parents expressed that their parental experiences were 'typical' (e.g., *'nothing out of the ordinary'*). Consequently, these parents may have perceived less relevant to recall their early experiences. Nonetheless, some interviewed parents did describe altered parenting behaviours related to the early child medical risk (e.g., being bit *'soft'* as a parent). Yet, contrary to Miles' and Holditch-Davis' (1997) model discussed in Chapter 1, as a whole, the interviewed parents linked their parenting behaviour to the parental perception of child ability to reach adult independence, describing factors such as child neurodevelopmental disability, behavioural problems, and/or concerns with social competence, rather than factors relating to the early experiences.

Similar to previous studies (Widding et al., 2020; 2019), those parents who volunteered their birth stories, recalled their experiences in detail (please consider Ms Amazonite's example on page 212), suggesting that the experiences were still, over a decade later, relevant. Aligned with previous quantitative findings (Alenius et al., 2018), Ms Amazonite described altered reproductive behaviour following the EP birth. In their study about birth experiences of parents of EP and moderately preterm born adolescents, Widding et al. (2020) suggested that those parents whose children had long-term morbidities following prematurity recalled the birth and hospital period more critically in comparison with parents of morbidity-free preterm born children. This was not evident among the interviewed parents in the present study. Within the psychoanalytic framework, previous studies have discussed the attachment and early parent-infant experiences to carry over to the adolescent-parent relationship (Hallin et al., 2012). Although, in the present study, a few birth stories and recollections of the hospital period confirmed previous research findings in that parents described it important to be able to participate in their child's cares, and recalled concerns about 'bonding' during the hospitalisation (Wraight et al., 2015), none of the interviewed parents contemplated that these early experiences had potentially impacted their relationship with their adolescent child

presently. Within the longitudinal birth cohort studies that are conducted among EP and VP born populations internationally, future research could utilise the method of longitudinal qualitative research to explore how parental experiences of attachment, ‘bonding’, and the parent-child relationship may evolve, fluctuate, or remain consistent over time.

Nevertheless, among the interviewed parents, parental perception of child vulnerability may have shifted over time (Tallandini et al., 2015; Thomasgard & Metz, 1997) as a few parents did describe having experienced ‘relief’ when their children had met all their developmental *milestones* together with their peers, which constituted the parents presenting as ‘*lucky*’. Similar experiences of initial uncertainty resulting in a sense of relief following typical child development have been reported by parents of preterm born children (Marlow et al., 2020). As discussed in the introduction to this chapter, parents of children with disabilities have described a ‘point of realisation’ when they have recognised that they were not parents of a typically developing child (Valle, 2017). In the present study, the realisation that ‘*all was okay*’ described by some parents may have reflected a similar ‘point of realisation’.

5.5.4 Implication of internalised parental sole responsibility

Overall, the interviewed parents described sole parental responsibility and experiences of ‘fighting for support’ for their children. The interviewees had internalised their self-defined parental sole responsibility for their children and the parents rarely questioned the responsibility (e.g., ‘*It’s not easy but you just get on with it. Get through it*’, ‘*...with my own expense*’, ‘*..and obviously I had to give up my career to care for...*’). Sociocultural expectations of parental role may promote intensive parenting practices, but also justify them as ‘normative’ and thus, parents may be less likely to question their efforts regardless of the fact that they may provoke personal distress (Eerola et al., 2021). Studies among parents of adolescents with disabilities have reported similar parental feelings of acceptance as ‘going along with it’ (Roll-Pettersson, 2001), experiences of sole parental responsibility (Hallberg et al., 2009; Valle, 2017; Woodgate et al., 2015) and fighting for support (Hallberg et al., 2009; Hetherington et al., 2010; Woodgate et al., 2015). In these studies, due to their responsibilities, the parents had developed stress-related symptoms such as chronic pain and insomnia (Hallberg et al., 2009; Woodgate et al., 2015) and reported limited social life and work opportunities (Kantrowitz-Gordon et al., 2016; Woodgate et al., 2015). The task of parenting was described as a labour-intensive undertaking that had put stresses on their partner relationship (Woodgate et al., 2015). Parents described a decline in their personal health due their child’s difficulties as a ‘natural’ cause of parenthood (Kantrowitz-Gordon et al., 2016). Contrary to this, in a study of how parents discussed regret in relation to having children in an anonymous online forum, the

authors found that the most commonly described contextual source of parental regret was the demandingness of raising children described as a ‘sacrifice’ and loss of ‘selfish things’ (Moore & Abetz, 2018). In the present study, the interview setting may have hindered parents from describing disagreement with parental sole responsibility. I will return to this point later when I discuss the limitations of this study.

Gendered roles of parenting were highlighted in the interviews. With a single exception of a two-parent interview in which the father had assumed the primary caregiver’s role, main carers were women. The interviewed women had internalised predominant responsibility for childcare and housekeeping (please consider Ms Turquoise’s and Ms Iolite’s examples on pages 197 and 211, respectively). Yet parents described frustration in relation to the feeling of being a sole person ‘*on top of*’ the overall care and services their children received from various healthcare and educational professionals, and parents expressed hopes of being able to return to work. A finding that women assume the majority of caregiving responsibilities is common (Shirani et al., 2011; Valle, 2017; Wilson & Cook, 2018). Particularly parental responsibility for children with chronic conditions have been framed as a primarily maternal duty (Clarke, 2013; Kanieski, 2010). Additionally, the dominant discourses of attachment and early-parent child relationship have commonly been portrayed as gendered (Macvarish & Lee, 2019). Yet studies among families with children with special needs have also presented contrary findings; parents have described functioning as ‘tag teams’ (i.e., taking turns) or sharing responsibilities in ‘complementary parenting’ (Woodgate et al., 2015). In the interviews, the mothers with children with complex care needs did not discuss ‘tag team’ or ‘complimentary’ caring. The only interviewed couple primarily described a sense of shared responsibilities in the task of parenting, rather than complimenting each other’s roles. As we have learned throughout this thesis, challenges relating to a child’s health may in some situations result in a necessity of intensive parenting practices; the child’s complex care needs may require extensive caregiving efforts. However, no necessity existed for these extensive efforts to be mainly provided by mothers (Webster, 2018).

The idea of an abstract ‘ideal’ interview script, against which the mothers reflected their own experiences (Jordan et al., 1994, p. 23-24) may have in part explained the absence of partners. If the ‘ideal’ parenthood has commonly portrayed the primary caregiver as a woman, then the fathers’ contributions may have been less likely to feature in the accounts. The mothers may have experienced shared responsibilities with their partners, even though the fathers were seemingly absent in the interviews, as they may have fulfilled the socially expected roles of fathers (Shirani et al., 2011), which in these interviews, for example, related to family finances. In their study of how parents negotiated division of domestic responsibilities, Eerola et al.

(2021) found that paternal involvement was commonly framed as a ‘choice’, whereas maternal responsibilities were taken for granted. Fathers’ involvement was described as ‘participating’ and ‘helping’, while mothers had the primary responsibility. Consequently, what the couples described as a shared responsibility was, in reality, culturally identifiable gendered patterns of parenthood (Eerola et al., 2021).

Aligned with previous research (Hallberg et al., 2009; Woodgate et al., 2015), the findings from this study suggested that internalising sole parental responsibility was demanding for parents whose children had complex care needs. In their interviews, both Ms Spinel and Ms Turquoise referred to the impact that their children’s delayed development had had on their roles as parents. Simultaneously planning her daughter’s transition to secondary school, Ms Turquoise expressed feelings of being ‘*stuck*’ in her role as a ‘*new parent*’. Woodgate et al. (2015) reported similar findings among parents of children with complex care needs. Due to the child’s delayed development, the parents in their study reported a lack of feeling of progress, while simultaneously the expectations for the parent role were ever increasing as previous developmental stages were not yet achieved when new stages arrived (i.e., feeling *stuck*). Parents in their study reported anxiety, stress, and chronic pains (Woodgate et al., 2015). In the present study, Ms Turquoise reported clinically significant levels of parenting stress which she related to the ‘*around the clock*’ care needs of her child. In addition, in the present study, parents of children with complex care needs perceived their children growing up commonly as a challenge, as behaviours, such as aggression, became harder to ‘manage’. Similarly, parents of adolescents diagnosed with ADHD reported feeling increasingly anxious about being continuously solely responsible for their children’s well-being (Hallberg et al., 2009).

In the present study, parents of children with limited independence described extensive caregiving efforts to respond to their children’s special needs, but also to provide them with ‘extra’ activities such as adult-assisted cycling and swimming. In the context in which parenting behaviour is regarded as a goal-orientated set of actions to raise an individual with valued adult skills (i.e., ‘successful’ adult) (Maccoby, 1992; Ogbu, 1981), some scholars have suggested that parents with children with special needs are less likely to turn to intensive parenting practices as they may feel that they have already ‘lost’ the opportunity to raise a competent adult (Valle, 2017). The findings from the present study did not fully support this suggestion. In the interviews, the experiences of parents of children who were not anticipated to reach independence showed that for these parents, the extensive caregiving efforts (e.g., 24-hour care) had become a necessity due to their children’s complex care needs (Webster, 2018). Yet, similar to findings reported by Woodgate et al. (2015), the parents constantly strove to

provide their children with “all they needed” and for them to “never go without” (p. 5), even though their parental efforts were not directed to achieve particular competencies (e.g., academic success) (Valle, 2017), but rather to ensure child happiness (Woodgate et al., 2015).

Among typically developing populations, research on intensive parenting practices has commonly focused on parental experiences of providing the children with what is needed to ‘get ahead’ in a race to success such as to maximise cognitive abilities (Wall, 2010). In the parenting culture, the idea of competition has been suggested to originate from neoliberal thinking (Gillies, 2020; Wall, 2010). Contrary to ‘getting ahead’, in the present study, parents of children with special needs discussed wanting their children to be *‘like everyone else’*. The pressures that these parents experienced were related to their child reaching the *‘typical’*, *‘average’* benchmark (please consider Ms Iolite’s example on page 212). Intensive parenting practices among parents of typically developing children may entail practices such extra tutoring, adult-lead and monitored activities (Wall, 2021), but for the interviewed parents of non-typically developing children, comparable extensive parental efforts were needed to reach a seemingly lower benchmark (e.g., *...I just don't want him to always be so far behind...*).

To place the extensive caregiving efforts and prolonged sole parental responsibility into context, it is important to note that the overall survival of infants born EP is increasing (Ancel et al., 2015), while the level of long-term morbidities among the survivors has remained unchanged (Marlow et al., 2021; Moore et al., 2012). Consequently, the absolute number of families with children with disabilities will increase. As in England, this phenomenon is paralleled with limited services for children with disabilities, behavioural or mental health disorders (Ford, John, & Gunnell, 2021), and parental responsibility over societal is prioritised (Macvarish & Lee, 2019), while, for example, financial support for low-income families is reduced (Dermott, 2012), the health and wellbeing of these parents becomes a public health issue (Valle, 2017). Extensive parental efforts have been associated with decreased parental mental health and HRQoL (Hatzmann et al., 2009; Rizzo et al., 2012). Thus, it becomes increasingly important to understand the lived experiences of parents of children with complex care needs long term.

5.5.5 Contextual factors impacting parenting behaviour

As discussed above, findings from the interviews showed that parental behavioural responses were impacted by parental perception of child ability and parental ambitions. Yet, both perceptions were influenced by contextual factors such as sociocultural framings of adult competence and ‘parenting’. Despite that some children appeared to have comparable

characteristics, parents responded to their children's challenges in varying ways. Some parents '*pushed*' harder for their children to learn new skills, whereas others described adjusted ambitions for their children. Ms Quartz, for example, described an adjusted independence for her children as '*never leaving home*'. This ambition/goal may have been influenced by child factors as her children's abilities limited the possibility of a full adult independence. In the future, their developmental challenges, although subtle, may prevent them from securing a job that would sustain independent living (Hallin et al., 2010). In contrast, to Ms Kyanite, university education showcased 'success' which meant, combined with her perception of her child's ability, that 'investing time and working hard' was necessary.

In addition, being culturally situated, some groups of parents may be more exposed to the discourse of parental determinism and therefore, more likely to adhere to intensive parenting practices, such as 'investing time and working hard' or 'pushing for normality'. Gender and social class have been found to influence the level of internalisation of the culture; women more often than men are assumed to take on the primary caregiver's role (Sevón, 2011; Shirani et al., 2011), as was evident among the interviewed parents. The level of education and financial resources may also alter the level of exposure to the intensive parenting culture. Parents with more financial resources are more likely to be surrounded by intensive parenting practices and consequently more likely to reflect their own parenting behaviour against those who 'parent intensively' (Lee et al., 2014, p. 31-33). On the other hand, these parents themselves may have more financial resources to spend on 'parenting' (Hoffman, 2010; Shirani et al., 2011; Wall, 2010). For example, families' financial situations may impact parents' ability to leave paid employment and invest time in 'parenting'. Many of the interviewed parents were working part-time or not at all in paid employment, while their partners were employed in intermediate or high level professions (Office for National Statistics, 2016).

In addition, child factors may be impactful in internalising intensive parenting practices. For example, child school success has been found to promote increased parental involvement (Dermott, 2012; Desforges & Abouchaar, 2003, p. 4). Children from high-resource families (education and income) have generally good school success (Desforges & Abouchaar, 2003, p. 4). Yet, high parental education level may also impact resources (i.e., capacities) that the parent has available to respond to their child's behaviours (Almogbel et al., 2017; Beckwith et al., 1992), which may reduce the stressfulness of intensive parenting practices. Yet, even if parents may have more financial or educational resources available to them to support their parenting task, intensive parenting practices may demand more from the parent, and thus, trigger more stress. The findings from the quantitative strand of this thesis supported this

suggestion as parents with higher educational attainment reported more parental distress than parents with lower levels of education. This finding was further supported by findings from the systematic review, in which Indredavik et al. (2005) reported increased emotional concerns among highly educated parents of preterm born children and Moore et al. (2006) found that high-resource families reported increased family burden over time. Yet, bearing in mind that these studies utilised varying measures of family SES.

5.5.6 Reflections on limitations

This qualitative research strand of a convergent MM study had limitations. The finding that parental ambitions and childrearing goals were a central theme in the parents' interviews might have been influenced by at least three factors. Firstly, due to the semi-structured interview design, unless the parents discussed these topics themselves, they were directly asked about their ambitions for their children and potential goals for being a parent. Secondly, the main study in which this qualitative research strand was situated, investigated long-term child outcomes following EP birth. Thus, the artefact of being in a study of long-term outcomes might have provoked the parents to particularly contemplate on 'the future'. Lastly, the particular time of transition to secondary education might have further focused parents' attention to their child's future. Nevertheless, none of these limitations directed the parents to discuss these particular goals or parenting behaviours. Thus, methodological considerations alone did not explain the findings. The fact that independence and acceptable social behaviour were important, is telling of what adult competencies were valued in this particular context (Ogbu, 1981). Research of childrearing goals among European parents have suggested similar importance on independence (Suizzo, 2016; Tulviste et al., 2007). Similarly, parental behavioural responses could be regarded as descriptions of understandings of 'parenting' in this particular context (Eerola et al., 2021).

As a critique of the analytic approach taken in the present study, with a first glimpse, it could be argued that in a thematic analysis, parental ambitions and perceptions of child ability could not be regarded as themes as they were somewhat direct questions in the semi-structured interview guide. Consequently, it could be argued that the interview data were not analysed, but merely described (Braun & Clarke, 2006). Yet, throughout this chapter, my thematic analysis of the interview data has demonstrated how parental perceptions and ambitions were linked to parenting behaviour and thus, they formed important aspects of the construct of parental experiences in this study. Furthermore, my discussion on the key themes, such as parental perception of child ability and its relation to the concept of 'adult competence', and the importance placed on 'the future', has demonstrated that these themes were more than just

mere descriptions of what the parents had answered to a specific interview question, although similarly labelled. The analysis has showcased the social construction of 'parenting' that occurred in these parents' interview accounts.

Another limitation related to the interview setting. In addition to reflecting a cultural perception of competence, the parents' references in their interviews to 'typicality' may have also reflected what the interviewed parents perceived as acceptable to describe to a researcher in an interview about 'parenting'. If being born EP was perceived as a risk factor for future child outcomes, parents may have felt compelled to describe extensive parental efforts and thus, account for how they had 'adequately' responded to the risk factor of EP birth (Faircloth, 2010; Kanieski, 2010). Parents may have been less likely to describe feelings they perceived as negative, such as regret (Moore & Abetz, 2018), especially to a researcher who the parents could have regarded as an 'outsider' (Berger, 2013); not a member of the groups of English parents or parents of children with disabilities. Thus, the finding that parents had predominantly accepted sole parental responsibility and extensive caregiving efforts could partially follow from these limitations. In fact, a few parents with clinically significant (PSI-4-SF score >91st centile) levels of parenting stress were purposefully sampled to take part in the interview. From these parents' accounts it seemed that the stress they reported was related to the described extensive caregiving efforts that they perceived their children needed. Yet regardless that their parenting efforts seemed to impact their personal wellbeing, the parents did not commonly explicitly express personal distress in their interviews. The method of telephone interviewing may have hindered me from achieving depth, as it, for example, prevented me from detecting non-verbal cues (Legard, Keegan, & Ward, 2003, p. 156-157). Yet, for example, in a study utilising feminist research methodology and online-only recruitment methods, the interviewed participants found it easier to discuss sensitive matters in a telephone interview without visual contact (e.g., videoconference) as it was perceived to provide an element of anonymity (Trier-Bieniek, 2012). In fact, along with increasing familiarity with varying communication technologies (e.g., telephone and internet), research participants may perceive non-face-to-face interviewing as a favourable method (Archibald, Ambagtsheer, Casey, & Lawless, 2019).

I have already considered the limitations of this qualitative research strand in relation to the deductive themes of parental ambitions and perceptions of child ability, as well as the limitations of telephone interviewing as a data collection method. Further limitations related to sampling. The aim of the sampling process for this qualitative research strand was to gain a maximum variation sample (Teddlie & Yu, 2007). I had predetermined parental characteristics such as SES to elicit varying parental experiences, although the aim of the sampling process

was not to establish causality or to represent the main EPICure2@11 Study sample directly. When interviewing these women, I realised just how varied the family backgrounds were. For example, a parent of an EP child with a mild disability was additionally a parent of a younger FT born child with severe disabilities. This impacted how she approached the abilities of the child in the main study. Another parent had adopted their EP child and therefore, had not experienced the EP birth themselves, although they had cared for their child during their hospitalisation. These varying family contexts tested the initial attempt to purposefully ‘control’ for varying parental characteristics. The decisions on how parents were sampled were based on a careful consideration of possible dimensions or factors that could influence or signify parental experiences. Researchers use power when they decide which dimensions are important in a specific context, and therefore direct who may be included in the group that contributes data (Mertens, 2007). When creating such pre-existing categories, I “disaggregated” (Mertens, 2007, p. 221) parental experiences on the basis of a number of characteristics. In addition to the ‘selected’ characteristics, the interviewed parents brought many other features and experiences to their interviews. The PSI instrument directed the parents to consider the particular child taking part in the study. Yet, in the interviews, parents discussed more freely their family life, other children, as well as the child in the study. As a limitation in the design of the Parent Questionnaire, questions relating to varying family factors were not included, such as characteristics of siblings.

Some of the aspects mentioned above became apparent during the interview, but also during my role as a clinical assessor in the EPICure2@11 Study. This role provided another point in which the main study’s influence filtered into the qualitative research strand. In my role as a clinical assessor, I had interacted face-to-face with some of the interviewed parents’ children. When some parents during their interviews described their children’s characteristics and parental ambitions for their children, from clinical perspective I was left unsure whether these goals were ‘realistic’. Their perceptions of their children differed to what I had learned about them during the day-long child assessment. However, I had only met some of the children, as also another assessor team worked in the main study. Having interacted with the child previously was a different, more comfortable, starting point for the interview and it potentially allowed me to prompt differently during the interview than with parents whose children I had not met.

Despite efforts, the sample of interviewed parents had mainly high SES, reflecting the sample of families participating in the main study; 14 families’ highest SES was intermediate or high and 15 parents had a university degree or post-secondary education. As discussed earlier, most of the women worked part-time in paid employment or were full-time homemakers. The

sample of the main study influenced the sample of the interviewed parents. Families from high socio-economic backgrounds have more commonly taken part in follow-up studies in comparison with families with low SES, although participation of families from more varied socio-demographic backgrounds may be increased by offering multiple response methods, such as telephone and online options (Johnson et al., 2014). Parental experiences, and parenting behaviour, among working-class parents and among families with low income may differ from that of parents with high SES (Lareau, 2002). Even though that the characteristics of the qualitative sample were relatively similar to the quantitative sample, when considering the qualitative findings, it must be remembered that only a proportion of parents were interviewed. The findings do not represent the whole cohort but rather deepen the understanding of possible parental experiences.

5.5.7 Implications for practice

This analysis has demonstrated that it is relevant for professionals who work with families of children born EP to consider how parents perceive their children's abilities and what ambitions they have for their children as these factors may impact their parenting behaviour and experiences. Echoing previous qualitative studies discussed throughout this chapter, the findings from this study have shown that parents, particularly mothers, may internalise a parental role that is personally demanding. Professionals are in a position to challenge the sociocultural framings of parenthood that prioritise individual maternal responsibility (Widding & Farooqi, 2016). At older child ages, the need for parental support may not be directly linked to a clinically defined level of child disability, as the findings from this analysis have shown that even subtle child developmental challenges (e.g., none/mild child disability level) may provoke intensive parenting practices. I will discuss this matter in detail in the next chapter of this thesis. Support from professionals, such as school personnel, may assist parents in considering their children's abilities and in supporting their children's subtle developmental challenges in a way that does not require extensive personal sacrifices. The analysis has demonstrated that parents of EP children experience widely varied parenting contexts. Parents could be supported in practical aspects of their parenthood, such as in providing respite from their 24-hour care duties and in offering extra support at the time of early adolescence when child behaviours may prove increasingly challenging to manage.

5.5.8 Conclusions and key findings

This qualitative research strand of a convergent MM study was the first in the UK to explore parental experiences among parents of EP YA in the context of the framework of parental determinism. This study has concluded that parental ambitions and perceptions of child ability

impacted parenting behaviour. Parents wanted their child to reach independent adult life. Although some parents described trusting, non-intensive parenting behaviours, parents who anticipated their children to have challenges with future independence described intensive parenting practices to support their child's development. These parental behavioural responses echoed the rationale of parental determinism discussed in detail in Chapter 1. Bearing in mind the labour-intensive caregiving efforts that parents of children with complex care needs described, this study has further directed attention to the health and well-being of parents who care for children with long-term morbidities.

Key findings:

- 1) Parental ambitions and the parents' perceptions of their child's ability to reach adult independence guided how they described their parenting behaviour
- 2) Parents who believed their children would become happy, sociable, and independent adults were generally trusting about their children's future and did not perceive extensive parental involvement in their child's life as desirable
- 3) Parents who anticipated their children to have challenges with future independence described varying parental behavioural responses to support their child's development in accord with parental determinism
- 4) EP birth was not a central feature in parental experiences at the time their children had reached early adolescence
- 5) Parents had internalised sole responsibility for their children which was demanding for parents with children with long-term morbidities

Chapter 6 Discussion

In this thesis, I have reviewed previous research on long-term parent outcomes following EP/VP birth. I have presented findings from a cross-sectional survey of parenting stress and sociodemographic, child and parental health factors collected as part of a national birth cohort study of extreme prematurity in England. I have also explored the parents' experiences at the time their child transitioned to secondary education using qualitative semi-structured telephone interviews. In this final chapter, I will bring together the quantitative and qualitative findings presented earlier in this thesis. As per my MM research question (Creswell & Plano Clark, 2011, p. 212-213), I will explore whether, and if so, how parenting stress and associated sociodemographic, child and parent health factors relate to the parental experiences.

In this convergent multistrand MM study (Teddlie & Tashakkori, 2006), I have explored varying aspects of 'parenting' an EP young adolescent with an overall aim to improve the current understanding of parental experiences among this population. I have examined parent outcomes of parenting stress and HRQoL among EP parents and parents of YA born FT. I have also explored parental perspectives to situate the outcomes in lived experiences. Consequently, in addition to the predefined outcome measure of parenting stress, this study has brought forward other influential factors in parental experiences. Present parental experiences and parenting stress have not previously been examined among English parents of EP YA. Yet, since the two research strands in this MM study did not examine a single concept, such as 'experiences of parenting stress' but instead explored varying aspects of the construct of parenting, instead of attempting to integrate the findings, for example by transforming data (Jang et al., 2008), I will discuss the key strand-specific findings alongside each other to explore how they related.

First, I will present short summaries of the quantitative and qualitative findings. These summaries will be followed by a section in which I will explore how the types of data related to each other and how the findings of this study may help redirect the research field of prematurity to strengthen the investigational focus of parental experiences and well-being among EP parents of YA previously largely lacking from the research agenda. I will then further discuss how the framework of parental determinism contributed to this study. I will discuss how the findings of this study related to the general research on parenting. Lastly, based on my key findings, I will present recommendation for future research and practice. Stemming from the exploratory nature of the present study, this discussion chapter is open-ended (Ribbens McCarthy & Gillies, 2017) inviting further examination and discussion on the concepts explored in the present study.

6.1 Summaries of Quantitative and Qualitative Findings

In the quantitative research strand of this convergent multistrand MM study, I found that parents of EP YA reported higher levels of parenting stress in comparison with controls, that was independent of the presence of child disability, although the average stress scores were within the normal range of parenting stress among both groups of parents (Abidin, 2012). Twelve percent of EP parents experienced high levels of stress in their parenting roles approximately 11 years after giving birth and reported considerably low levels of mental HRQoL. The decreasing trend of parenting stress with increasing child age among EP parents and the opposite trend among the controls was evident. Parents of EP YA who attended SEN schools or units more commonly reported increased levels of total parenting stress in comparison with those parents whose children were educated in mainstream schools. This association diminished when parental mental HRQoL was considered. Parental distress increased among those EP parents who had a university level education, whereas parents of younger EP children and those who received income support benefits reported high child-related parenting stress.

In the qualitative section of this study, I found that parental ambitions and the parents' perceptions of their child's ability to reach adult independence guided how they described their parenting behaviour. Parents who believed their children would become happy, sociable, and independent adults were generally trusting about their children's future and did not perceive intensive parental involvement in their child's life as desirable. Parents who anticipated that their children would have challenges with future independence described varying parental behavioural responses to support their child's development which related to the framework of parental determinism. The EP birth was not a central feature in parental experiences at the time their children had reached early adolescence. The interviewed parents had internalised sole responsibility for their children which was demanding for parents with children with complex care needs.

6.2 Parenting Stress and Experiences: An Integrative Discussion of Key Findings

The quantitative finding that a majority of the EP parents (78%; 147/167) reported total parenting stress scores that were within the normal range of stress could be related to the qualitative theme 1: parental ambitions / childrearing goals identified in the interviews, in which some parents were trusting that their children would reach adult independence and did not perceive intensive parental involvement in their child's life as desirable. The quantitative finding that among the EP parents a high total parenting stress score was associated more strongly with a SEN schooling status than with the level of child disability could be reflected

against the qualitative theme 2: parental perception of child ability, which showcased that among some parents the parental perception of child ability differed from the clinical definition of level of child disability utilised in the EPICure studies. The overlap between parenting stress, child disability, and parental perception of child ability was an overarching topic. The relation between the qualitative sub-theme of ‘pushing for normality’ and high parental distress domain scores is also discussed in the subsequent sections.

6.2.1 ‘All is okay’

In the qualitative chapter of this thesis, within the theme 1: parental ambitions / childrearing goals, I demonstrated how those interviewed parents who anticipated that their children would reach happy, sociable, and independent adult life were generally trusting about their child’s future and did not perceive extensive parental involvement in their child’s life as desirable. In the quantitative section of this study, a majority of EP parents (78%; 147/167) reported parenting stress scores that were within the typical range of responses for the general population (Abidin, 2012). It must be acknowledged, however, that these two findings did not indicate the same measure; in other words, it could not be stated that those parents who were ‘trusting’ of their children’s outcomes did not experience stress in their roles as parents (please consider Ms Onyx’s and Ms Garnet’s examples on pages 194-195). Nevertheless, both findings indicated that in the present study, following EP birth, a proportion of parents were generally coping well in their roles as parents at the time when their children were transitioning to secondary education.

The parenting stress data in this study was novel in that no previous stress outcomes have been reported among a modern cohort (i.e., born in the 2000s) of EP (<27 weeks’ gestation) adolescents’ parents. The findings of this study were aligned with studies of parenting stress among VP born younger populations in that the stress scores that those parents reported were also largely within the normal range of stress (Brummelte et al., 2011; Landsem et al., 2014; Linden et al., 2015; Schappin et al., 2013; Treyvaud et al., 2014). Comparable findings of average stress scores within the normal range have been reported among other clinical study populations, such as among parents of school-aged children with ADHD or ASD (Larkin et al., 2021) and among parents of children in care or adopted from care (McSherry et al., 2019). The findings of a generally low level of parenting stress and somewhat trusting parental perceptions of the possibility of future adult competence among some interviewed parents supported my earlier postulation that the generally negative perception of long-term parent outcomes among prematurity found in my systematic review may not be fully defensible. In the review, I found that the research among prematurity has mainly focused on examining

negative family outcomes. Future research could identify and explore factors that may promote positive family outcomes over time.

Previous qualitative studies among parents of preterm born older children have reported somewhat more critical findings. Yet methodological considerations may in part explain differences in findings. Studies have explored parents' birth stories, reporting that the parents may recall the birth as an emotionally charged experience many years later (Widding & Farooqi, 2016; Widding et al., 2020). Studies among EP parents have also positioned the interviewees to reflect their present parental experiences against the adverse birth experience and the first years of parenthood (Kantrowitz-Gordon et al., 2016; Wilson & Cook, 2018). These studies have noted a methodological limitation. Due to their recruitment methods (e.g., advertisement posters and leaflets) the parents who took part in their investigations may have had particularly strong experiences (Kantrowitz-Gordon et al., 2016; Wilson & Cook, 2018). On the contrary, in the qualitative research strand of the present study, I approached parents who took part in the main study using the strategy discussed in detail earlier. As the case-selection process was researcher-led, it may have prohibited only those parents with particular experiences to come forward for the interview, perhaps resulting in a sample of parents with a wider range of experiences in comparison with previous investigations among comparable populations. In the present study, none of the approached parents declined the interview, suggesting that at the time of early adolescence the parents were generally willing to discuss their parental experiences. Yet, it is important to note that the qualitative sample was a subset of the quantitative sample. Selection bias may have existed as those families who consented to take part in the EPICure2@11 Study may have differed from those who declined participation, as discussed in detail in Chapter 4.

Yet, in both the quantitative and qualitative research strands of the present study, a proportion of EP parents reported high levels of parenting stress, low levels of mental HRQoL, and described labour-intensive parenting efforts (please consider Ms Turquoise's example on pages 191 and 198). Aligned with a previous study among EP families (Minoli et al., 2013), the wide range of parenting stress scores and varying parental experiences among the study population further demonstrated that EP populations represent a heterogeneous group of families of whom many may not consider themselves as 'at-risk'. In fact, studies have suggested that to label individuals (or families) as chronically 'at-risk' based on a single factor such as EP birth, may overlook the dynamics between contextual factors/events and individual characteristics that may change significantly over time (Hoffman, 2010).

The early model of Preterm Parental Distress, discussed in the introductory chapter of this thesis, suggested that parents may display ‘compensatory’ parenting behaviours because of the initial perception of infant ‘fragility’ (Miles & Holditch-Davis, 1997). Sociocultural framings of prematurity, such as the media discourses of ‘miracle babies’ and ‘fighters’ have been suggested to impact parental perceptions at the time of preterm birth (O’Neill, 2019). Yet, following on from the trend of increasing preterm livebirths along with improved survival (Torchin et al., 2020), prematurity as a phenomenon may have become more normative. Present studies have suggested that since preterm birth has become more common, parents may respond to it less intensively (Noy et al., 2015). To some extent the findings from the present study may support this newer hypothesis as ‘prematurity’ was not a prevalent topic in the parents’ accounts when they were not directly asked about the birth. Furthermore, the levels of parenting stress reported by many EP parents were comparable with the control parents. Nevertheless, no direct outcome measure of parental perception of child vulnerability was utilised in the present study. This could have aided to explore the topic of parental perception of ‘fragility’ further. Future research could systematically explore discursive understandings of prematurity in an English sociocultural context and how these framings may impact parents’ and healthcare professionals’ experiences. Such studies could further inform how parents may perceive the phenomenon of ‘prematurity’ (O’Neill, 2019).

6.2.2 Parental perception of child ability and parenting stress

The findings from this MM study have shown that a clinical categorisation of level of child disability may not directly reflect the parental perception of child’s ability to reach adult independence. As was evident in the parents’ quotes presented throughout the Chapter 5 results section, how the level of child disability was categorised in the main study (i.e., none/mild, moderate, or severe), did not directly correlate with parental perception of a possibility of future adult independence. For example, among the interviewed parents, extensive caregiving efforts and perceived ‘indefinite’ parental responsibilities were not necessarily linked to high levels of child morbidity, as was apparent in Ms Spinel’s and Ms Quartz’s examples on pages 197 and 209, respectively. Although, as discussed earlier, this discordance may have related to the presence of child behavioural disorders. I additionally hypothesised that the discrepancy between parental perception of child ability and the clinical categorisation of level of child disability may have related to the particular importance that these parents placed on their child’s social functioning. This interpretation was supported by the association between SEN school attendance and high parenting stress levels. I discuss this argument, as well as other possible interpretations in detail below.

Built upon the findings from the systematic review of long-term parent outcomes following EP/VP birth, I had hypothesised that the potentially increased parenting stress among EP parents would largely be attributed to the presence of child neurodevelopmental disability. Initially, I had presumed child disability to be linked to the extensive caregiving efforts described by some parents in their interviews. Yet, both the quantitative and qualitative findings of this study did not fully support my hypothesis. The differences in the average total parenting stress and child-related parenting stress scores between the groups of parents persisted after excluding parents of children with severe or moderate neurodevelopmental disability. The findings did echo a previous study utilising the PSI-SF instrument (Zerach et al., 2015) in that the differences in parental distress and increased challenges in parent-child interaction diminished when families with children with disabilities were excluded. This finding suggested that the negative parent impact among EP families was partially experienced through its relation to the sequelae of being born prematurely (Drotar et al., 2006).

Yet, when factors associated with high parenting stress solely among EP parents were further assessed using the method of logistic regression, I discovered that rather than the child disability, child attendance at SEN school or unit had a stronger association with high total parenting stress (SEN school: *aOR*: 4.1, 95% *CI*: 1.1, 15.4 and child disability: *aOR*: 1.6, 95% *CI*: 0.5, 5.1, respectively). The type of school was also associated with high parental distress (*aOR*: 4.4, 95% *CI*: 1.1, 17.3) and had a borderline association with child-related stress (*aOR*: 3.0, 95% *CI*: 0.9, 10.2) (please see Table 4-6). When considering these findings in the context of the qualitative research strand, I was unsurprised that rather than the clinical assessment of a level of child disability, a variable that indicated child social functioning (i.e., ‘manage in school’) was more strongly associated with parenting stress. Theme 2: parental perception of child ability, for example for the child to ‘function’ at school, was a central feature in the parents’ accounts (please consider Ms Kyanite’s and Ms and Mr Jade’s examples on pages 200-201). In their interviews, the parents commonly placed high focus on their children’s social behaviour, acceptance by peers, and them being able to ‘manage’ school and ‘function’. Parents described supporting their children’s schoolwork and facilitating friendships as their daily tasks. Some parents felt that their abilities to support their children were diminishing due to the increasingly complex social relations among adolescents. Parents of EP adolescents have previously reported increased concerns about their child’s peer relations (Ritchie et al., 2018; Taylor et al., 2001) which has impacted the parents’ own quality of life even after their preterm born child has reached adulthood (Wolke, Baumann, Busch, & Bartmann, 2017).

Naturally, the type of school correlated with child disability. A majority (20/21) of children attending SEN schools or units had moderate or severe neurodevelopmental disability.

However, of the 63 children with moderate/severe neurodevelopmental disability, 43 were educated in mainstream schools. Consequently, it appeared that in this sample of parents, the joint appearance of child disability and a need for special education was found as most stressful.

Yet, other factors may explain the association between SEN schooling and high total parenting stress scores. Previous studies among other clinical populations, such as among parents of school-aged children with cerebral palsy (Majnemer et al., 2007) and parents of children with ASD or developmental delays (Barroso et al., 2018) have suggested that parenting stress may be associated more strongly with child behavioural and/or psychosocial difficulties rather than with the level of physical child disability (Majnemer et al., 2007) or other chronic conditions (Barroso et al., 2018). This could partially explain the diminishing impact of child disability in the present study, as the disability variable did not include child behavioural factors. The association between SEN schooling and parental distress could reflect child behavioural difficulties. Behavioural difficulties, such as attention deficits, not only affect the child's social interaction outside the home, but also their interaction with their parents (Jaekel et al., 2012). Consequently, the child's ability to behaviourally '*manage*' in a mainstream school and '*function*' may also be indicators of the parent-child relationship and the potential stressors in it. To the best of my knowledge, previous studies assessing long-term parent outcomes following EP birth have not considered the type of school in their analyses. Consequently, it is challenging to examine this hypothesis in the context of previous evidence.

It is important to note that in the present study, the SEN school status was a contextually varying factor, and thus, to an extent, an imprecise measure. The type of school was recorded at the time of the clinical child assessment. Some children had '*managed*' in a mainstream primary school but were about to transition to a SEN secondary school. Some children who attended a mainstream school were able to cope because their school provided one-to-one assistance, or the school had only two classes of less than 10 pupils. Some children attended SEN schools as the mainstream schools in their areas provided limited special needs support services. Thus, the findings from this study may be interpreted to suggest that for these EP parents, their child '*managing*' in a mainstream school was an important factor in reducing stress, even if it may have been as a result of contextual rather than child factors.

The reason why the clinically defined level of child disability did not have a priority in parental perceptions of child ability among the interviewed parents may have related to its definition. In the EPICure2@11 Study, '*child disability*' was a categorical status/condition ranging from '*severe*' to '*none/mild*' (Marlow et al., 2021), indicating a level of division from an observable

physical/medical function or structure, highlighting the measurability of the morbidity (Bickenbach, Chatterji, Badley, & Üstün, 1999). ‘Disabilities’ were, for example, limitations in vision, hearing, or cognition. Thus, as in general in the research field of prematurity (Marlow et al., 2020), the disability variable indicated the likelihood of the child to be able to function independently. Even though the parents placed a similarly high focus on future independence, an important part of their perception of adult competence was the child’s ability to function socially (i.e., independent, happy, sociable adult life). For these parents, the experience of their child’s disadvantage in the sociocultural and attitudinal environment seemed pivotal (Bickenbach et al., 1999, p. 1175). As reported by a previous study (Larkin et al., 2021), the way in which parents in this study perceived their child’s ability was meaningful, as it was linked to their ambitions for the child’s future and impacted their parenting behaviour and levels of parenting stress. The findings from my systematic review further supported my conclusion that parental perception of child ability may impact parent outcomes as the reviewed studies reported that parental concern about adolescent health had negatively impacted parents’ emotional health (Indredavik et al., 2005; Saigal, Burrows, et al., 2000), although the parents of preterm born adolescents may view their child’s abilities critically (Hallin et al., 2010; Indredavik et al., 2005; Lund et al., 2011; Saigal et al., 2003),

The EPICure studies have defined child disability following internationally recognised classifications, aligned with other similar longitudinal birth cohort studies. The clinical definitions of impairments and their precise ‘cut-off’ points have focused on medical and longer-term health status interests. Consistent and internationally unified definitions have helped to assess and compare long-term outcomes both within and between the cohorts over time (Marlow et al., 2020). For example, together with survival, the level of child disability has been utilised to indicate/measure effectiveness of previous medical interventions. As discussed in the introductory chapter of this thesis, these outcomes have also been communicated to parents who have faced decisions about their EP infant’s care (O’Neill, 2019). Thus, the outcomes have held the potential to impact care decisions. Aligned with other scholars (Marlow et al., 2020), the finding from the present study that the clinically defined level of child disability did not have a priority in parental perceptions of child ability among the interviewed parents, and that a variable that indicated child social functioning (i.e., type of school) was more strongly associated with high parenting stress scores than the level of child disability, points to the importance in research to utilise outcome measures that are relevant for EP families’ everyday lives. Undoubtedly, it is important to be able to compare outcomes over time and between cohorts to improve care and practices. However, equally important, if not more, is the utilisation of measures/concepts/entities, that relate meaningfully to the

everyday lives of those who live with the consequences of preterm birth and the care decisions that followed the birth.

The findings from this study have shown that, as discussed in the introductory chapter of this thesis, due to the reciprocal nature, parental experiences in the parent-child relationship are co-constructed together with the child (Maccoby, 1992). Parental perception of child ability impacted the interviewed parents' ambitions for their children and how they described their parenting behaviour. In the quantitative section, child factors such as age, level of disability, and SEN schooling were associated with parenting stress scores. The parents' understandings of their roles as parents were aligned with the dominant scientific understanding of 'parenting' to socialise the child (Kuppens & Ceulemans, 2019). Parents' aim was for their child to 'make it' (Ogbu, 1981). Despite the fact that the parental perception of child ability may not be aligned with the clinical assessment of child disability, parental perceptions are important as they may impact parenting behaviour and parent outcomes. In practice, it may be relevant to pay more attention not only on child disability but also to how parents perceive their children's abilities, as this may be an indication of the potential stressors in the family context. On the basis of the present exploratory study, future research could further explore factors in the sociocultural environment that may contribute towards parental experiences and fears for their child's inability to *function*, and how those environmental factors that create disadvantage could be altered (Bickenbach et al., 1999).

6.2.3 'Pushy' parents and parental distress

Terms such as 'acceptance' and 'adjustment' have been utilised to describe the process of change that parents of children with morbidities may begin at the time of diagnosis of child disability (Landsman, 1998). Contrary to 'acceptance', in their interviews, some parents described how, at the time of early adolescence, they still sought to alter/change their children's outcomes. These parents were focused on *'working hard'* and being *'committed as a family'* and *'pushed for normality'*. These parents' accounts demonstrated that in addition to child ability, the *'push'* for change was influenced by their parental aspirations. Contrary to findings from studies among other clinical populations (Hallberg et al., 2009; Roll-Pettersson, 2001), the parental perception of a need for extensive parental efforts was not always linked to severe adolescent morbidity. Interestingly, in the present study, those interviewees who perceived their child's abilities as 'nearly like others' perceived intensive parental involvement as necessary. The findings from this study suggested that the parental attempt to achieve 'typicality' was a labour-intensive undertaking for some parents.

The particularly increased parental distress scores among those EP parents whose children attended special schools was an interesting finding. In the context of the qualitative results, it could be reflected against the theme 3: parental behavioural response of ‘pushing for normality’. Ms Iolite (please see page 203) described how she was reluctant to ‘give up’ on normality and ‘pushed’ for it with an aim of making her child ‘as normal as possible’. Therefore, the particular strain in the Parental Distress domain seemed sensible as the potential tension related more to the parents’ personal ambitions and aspirations, and less to the relationship with the child. Yet, heightened parental distress scores correlated strongly with low mental HRQoL which may have indicated that the personal distress that some parents reported was not solely related to the parenting role. Heightened parental distress scores among those parents whose children attended SEN schools may have carried over from early childhood (Treyvaud et al., 2014) and/or indicated prolonged burden of caring for a child with complex care needs.

The parental behavioural response to ‘push for normality’ had similarities with the ‘fighter’ representation commonly utilised to describe preterm born infants who have been admitted for neonatal intensive care (O’Neill, 2019). Due to their immaturity, EP infants’ survival is uncertain, commonly characterised by an exchange between improvements in condition and setbacks. Thus, framed as a ‘fight for survival’ (O’Neill, 2019). In a parallel manner, the parents in this study expressed a sense of ‘fight’ when they ‘pushed’ for their children to achieve what they perceived as ‘normal’. Instead of fighting for survival, the parents’ focus had shifted to ‘fight’ for their children’s ability to succeed in the surrounding social world.

The infants’ ‘fight’ for survival in the neonatal intensive care unit is temporary; in the best-case scenario it results in the child’s discharge alive from hospital. Similarly in this study, the parents commonly perceived their ‘pushing’ as a temporary necessity; Ms Kyanite was going to ‘push’ until her child was what she was ‘supposed to be’ (please see p. 201) and Ms Fire Agate ‘pushed’ until a ‘turning point’ (please see p. 206). In some ways, the parental sense, that had started in the neonatal unit, of not wanting to ‘give up’ and thus, ‘push’ was still, 11 years later, ongoing. A similar theme of not wanting to ‘give up’ on their child has been described among EP parents in the NICU context (Wraight et al., 2015). A framing as such, however, may be perceived to imply that families who face challenges, do so because of a lack of persistence or a will to ‘fight’ and thus imply ‘giving up’. In fact, in the NICU context, the ‘fighter’ representation has been suggested to be harmful as it may be interpreted to suggest that survival may be impacted by factors such as personal character (O’Neill, 2019).

Parenting behaviour that the interviewed parents described related more to the parent role to promote child outcomes and mitigate potential risks in achieving future competence than to a need for parental protection (Thomasgard & Metz, 1997). References to vulnerability and EP birth had limited appearance in these parents' accounts. When intensive parenting is regarded as a culture, in addition to investigating frequencies of parenting practices (Yerkes et al., 2019) and assessing perceptions of levels of parental involvement (Schiffrin et al., 2013) it may also be important to examine parental perceptions of the purpose of their efforts (e.g., child to 'make it'). In addition to what is done or how often it may be important to also understand why these efforts are made.

Is it justifiable to link the behavioural responses such as those described by the interviewed parents to their parenting stress scores? As per Abidin's (1992) parenting stress model discussed in the introductory chapter of this thesis, an individual's response to the stress trigger is influenced by child and parent factors, contextual factors such as societal expectations of the parenting role as well as by parents' own perceptions of appropriateness of child and parent behaviour, and by a balance between resources and stressors. Therefore, whether parents who 'invest time and work hard' or 'push for normality' experience stress due to their parenting efforts depend on factors such as the availability of family resources to leave paid employment and support received from the parents' social networks (Noy et al., 2015). In one hand, by 'investing time and working hard' some interviewed parents may have been able to sustain a sense of control over their children's outcomes (Yerkes et al., 2019). On the other hand, in previous studies, parents of adolescents with special needs have reported poor parent outcomes such as lack of sleep, pains, and poor physical health due to increased parental efforts linked to their child's complex care needs (Hallberg et al., 2009; Woodgate et al., 2015). A concern in parental responses as described by some parents in the present study is that the potential adverse consequences, such as parenting stress, poor mental health outcomes, loss of job opportunities, or limited social life, may appear acceptable or even be perceived as a 'natural consequence' of parenthood (Kantrowitz-Gordon et al., 2016) which may reduce the likelihood among parents to recognise the negative impact on personal wellbeing.

Despite expressing challenges, generally the interviewed mothers in this study did not question their extensive contributions towards their children, the gendered roles of parenting, or their perceived primary responsibility for their children in relation to receiving support from close relatives, school, or healthcare professionals. The absence of critique, however, cannot be taken to denote its non-existence. Critique might have been absent due to the parents' attempt to adhere to the accountability framework and to show a coherent image (Faircloth, 2010; Jordan et al., 1994, p. 23-24). As I have discussed earlier in Chapter 5 in relation to the

gendered roles of parenting, if the interviewees perceived that parenting ‘should’ be all-consuming labour-intensive ‘hard work’, during their interviews with me, they may have been less likely to raise concerns. To internalise responsibility for one’s child, may be regarded as an attempt to adhere to the dominant discourse which commonly promotes personal responsibility over societal (Taylor et al., 2000).

6.3 The Research Field of Prematurity and Parental Determinism

As I have demonstrated throughout this thesis, studies in the research field of prematurity have implicitly based their theoretical grounds on the psychoanalytic framework and attachment theory when examining the relationship between parenting behaviour and child outcomes. The findings from my systematic review showed that the rationale of parental determinism appeared in research frameworks. In some of the reviewed studies, parenting behaviour was understood as a means to promote child outcomes (Jaekel et al., 2012) or as a risk factor that could explain differing outcomes among adolescents/adults born prematurely (Pyhala et al., 2011). “Parenting issues” (p.251) were hypothesised to be linked to poor adolescent mental health outcomes among VLBW survivors (Pyhala et al., 2011). Yet, the reviewed studies also demonstrated that the evidence-base of long-term parent outcomes following EP/VP birth was uncertain. Methodological limitations, such as reliance on a retrospective self-report of parenting behaviour (Pyhala et al., 2011) and the potential for reverse causality between parenting behaviour and child outcomes (Treyvaud et al., 2020) were apparent. Nevertheless, as in other areas of parenting research (Macvarish & Lee, 2019), psychoanalytic and attachment theories, as well as parental determinism remained influential in the conceptualisation of ‘parenting’, regardless of limited evidence-base. Findings from the present study showed that at the time of early adolescence, parental determinism appeared in the interview accounts. Some of the interviewed parents were ‘*working hard*’ for their children in the belief that they could improve their children’s outcomes, consequently feeling responsible for those outcomes. Yet, the parents referred less to their early experiences of EP birth in comparison with previous reports among comparable populations (Widding et al., 2020). The findings from this study highlight the impact that the selected conceptual frameworks have on research outcomes. It has been suggested that conceptual frameworks such as above may miss the complexity and contextual variability of ‘parenting’ (Gillies, 2008).

The labour-intensive caregiving efforts described by EP parents in this study, and in other reports (Kantrowitz-Gordon et al., 2016; Wilson & Cook, 2018), form an integral part of the phenomenon of extreme prematurity in England. Despite that preterm survivors have

discursively been stated to “owe their lives to modern neonatal intensive care” (BBC, 2004a), following discharge from hospital the responsibility for long-term child outcomes has largely remained with the parents (Shaw et al., 2016). While, with their intensive parenting efforts, some parents in this study may have focused on ‘making the “miracles” happen’, on occasions, their own personal health and wellbeing had suffered, which related to their labour-intensive parenting efforts. A proportion of parents in this study reported high parenting stress scores and low levels of mental HRQoL. These outcomes were present among some parents of YA born EP who felt necessary to *‘push’* for their children to meet the social standards of adult competence; become a happy, independent, and sociable adult, and thus, to ‘make it’ in the society. Yet, it is important to note that the qualitative sample of this study was limited and not designed to draw conclusions of potential causality between factors or to generalise findings. Nevertheless, as was demonstrated by my systematic review, the research on long-term parent outcomes following EP/VP birth has been uncommon. Consequently, a limited view of the phenomenon of extreme prematurity in England has been portrayed. To establish a wider picture, together with infant survival and child disability, future practice could consider parent outcomes as part of the primary outcome measures of extreme prematurity. As the evidence-base of long-term parent outcomes following EP birth is further strengthened, it may be communicated with more certainty to parents who are at the risk of preterm labour or whose child is born preterm.

My criticism of parental determinism and the key argument of this study may seem conflicting; how can I conclude that 1) the parents, with their excessive caregiving efforts are the ones making the “miracles” happen and 2) the research field of prematurity should abandon the thinking that parents have direct causal impact on their children’s outcomes. The latter invalidates the first. If we look back, for example, at Ms and Mr Jade’s experiences (please see page 200) of being *‘committed as a family’* to tirelessly *‘almost train’* their child to behave in a *‘socially acceptable’* manner, it would be challenging to disregard their pivotal roles in contributing towards their child’s future. However, the criticism of parental determinism does not suggest that parents do not influence or impact, promote or hinder, their children’s outcomes. By arguing that parental determinism is socially constructed, I am not stating that research results, such as those demonstrating sensitive parent behaviour to promote child task persistence (Jaekel et al., 2012) are not ‘true’ or ‘valid’. Rather, I am directing attention to the critical question why such a high importance has been placed on parent behaviour, (by both researchers and parents themselves) when the evidence-base that proves a link with child outcomes is much less factual (Dermott, 2012). After all, as found in the systematic review, previous research on long-term parent outcomes, including parent behaviour, has been scarce and subject to methodological disparities. The social focus on parenting appears extensive in

comparison to its scientific evidence-base. In the next section I will explore this question further.

6.4 ‘Good enough’ Parenting: A General Comment on the Research on Parenting Behaviour

Throughout this thesis, I have sought to demonstrate how sociocultural assumptions, or even political agenda, have been present in parenting research (Ribbens McCarthy & Gillies, 2017), and also in research among families with children born preterm. The discussed studies have demonstrated that a cultural normativity may moderate parenting behaviour (Eerola et al., 2021; Widding & Farooqi, 2016). In the methods section of this thesis, in the view that ‘parenting’ may be regarded as a socially constructed entity, I postulated that its framing may also entail political agenda (Rosen & Suissa, 2020). The examples of political framings of parenting have mainly referred to the idea that when parents are placed in a position of primary responsibility over their children’s outcomes, societal responsibility for child outcomes can be minimised (Dermott, 2012; Macvarish & Lee, 2019; Ribbens McCarthy & Gillies, 2017) (please consider the opening quote of this thesis from DfES). For example, when parents with low SES are positioned to bear responsibility for their children’s poor school success, the government may claim to have actioned with limited input when implementing interventions that target specifically ‘parenting’ (e.g., improve ‘parenting competence’), instead of strengthening costly societal structures such as the income support and housing and education systems (Dermott, 2012). In the present study, high child-related parenting stress was associated with receiving income support benefits. Low SES has been associated with higher levels of parenting stress among parents of VP born children (Singer et al., 2010; Zerach et al., 2015). Among EP families, availability of support services may impact the level of personal responsibility required from the parents, such as in relation to parental financial sacrifices following EP birth (Kantrowitz-Gordon et al., 2016) versus a sense of security gained from the availability of a national low-cost healthcare system (Jaekel et al., 2012). Findings from the present study, as well as from studies among other populations (Barnes & Power, 2012; Wall, 2010), have indicated that parents may internalise responsibility for their child’s outcomes even in a context in which they have limited possibilities to influence these outcomes, such as in extreme poverty (Barnes & Power, 2012) or in relation to their child’s ‘brain potential’ (Wall, 2010). As was concluded by a parent in the present study: *“I’m probably trying to do something impossible”*.

The findings from the present study do not necessarily tap into a this kind of political discussion, but they do relate closely to the discussion of parenting behaviour; when and by

what definition is parenting behaviour ineffective (Maccoby, 1992), inappropriate (Gillies, 2008; Ribbens McCarthy & Gillies, 2017), overprotective (Day, Dobson, et al., 2018; Day, Schmidt, et al., 2018), hostile (Pyhala et al., 2011), positive (Treyvaud et al., 2020), cognitively stimulating (Wolke, Jaekel, et al., 2013) or ‘good enough’ (Taylor et al., 2000). At the start of the 21st century, Taylor et al. (2000) called for a practical, culturally and contextually sensitive, definition of parenting that could be used to identify appropriate levels of caring from in appropriate (Kotchick & Forehand, 2002; Taylor et al., 2000, p. 114). Today, scholars are still reporting parental anxieties of over- and under-parenting of their adolescent children based on social understandings and expectations. This is especially at the time of adolescence, when independence should be nurtured while the surrounding social risks may increase (Wall, 2021). Furthermore, as discussed earlier, the appropriateness of parenting behaviour continues to be contextually varying. It may be perceived as appropriate for parents to monitor their children’s sleep or diet (Símonardóttir, 2016) but not their play (Dodd & Lester, 2021). The various studies of parenting behaviour discussed throughout this thesis had used standardised validated instruments to assess parenting behaviour. Yet these studies had generally relied on a retrospective parent or child self-report of parenting behaviour, introducing obvious methodological limitations of a potential of reverse causality and recall and/or information biases. Other investigators had observed and assessed parent-child interactions utilising standardised scoring instruments. Yet the observed parent-child interaction sessions had been short (e.g., 10 minutes) and low levels of inter-rater reliability were reported (Treyvaud et al., 2020). How these parents interacted with their children over the years outside of the study settings remained unknown in these investigations.

The difficulty to define parenting behaviour that is ‘good enough’ is a general challenge in parenting research, while the extremes of poor as well as good parenting may be identifiable (Taylor et al., 2000). Certain parenting styles, such as authoritative parenting has been shown to correlate positively with adolescent outcomes, such as social competence and academic achievements, among general population samples (Kuppens & Ceulemans, 2019). Highly involved parenting behaviour has been associated with poor adolescent mental health (Schiffrin et al., 2013). Yet, as discussed earlier, even if parenting behaviour has been associated with various child outcomes, it does not follow that parenting could be regarded as an “independent mechanism through which negative outcomes for children can be avoided” (Dermott, 2012, p. 2). In other words, negative outcomes cannot be necessarily avoided, even if parents intensively promote their child’s positive development.

Even more so, aligned with earlier scholarly discussion (Dermott, 2012), the findings from my interviews showed that parenting behaviour is not a unitary concept, but in reality it refers to

a variety of activities of caring and perceptions of responsibilities. Similarly, when examined from the child perspective, a universal definition of ‘optimal’ parenting behaviour would assume that a single set of actions (i.e., parenting) is universally applicable to all children which would diminish the importance of the long-standing parent-child relationship built on unique personalities and temperaments (Gillies, 2008; Wall, 2021). In addition, the discussion in this thesis has demonstrated that it may be challenging to separate the impacts of varying factors such as parent and child health factors and families’ SES from the impact of parenting behaviour (Gillies, 2008), as Hallin et al. (2012) noted in relation to adult attachment experiences following EP birth. Parenting behaviour may be regarded as a proximal factor of more distal impact factors such as SES (Taylor et al., 2000). As I contemplated at the beginning of this thesis, the varying impact of child, parent, family, and contextual factors on parenting behaviour and child and parent outcomes do not only interact with each other, but also the movement in the process may be bidirectional. Therefore, simple linear models of causality may prove too simplistic to capture a concept as varying as parenting behaviour (Gillies, 2008).

As discussed at the beginning of this thesis, in her early review of parenting research, Maccoby (1992) concluded that if to ‘succeed’ parents are to “accepted almost unlimited, long-term commitment to promoting the child's welfare” (Maccoby, 1992, p. 1015). Over 30 years later, a similar acceptance of responsibility appeared in some interviewed parents’ accounts in the present study (i.e., *“if she’s got something on, whatever I’ve got on doesn’t happen”*). Nevertheless, resistant voices questioning the discursively represented ‘total’ parenting (Smith, 2010) have emerged (Joiko, 2020). The coronavirus disease (COVID-19) pandemic has further highlighted the limited services available for children with special needs and their families in England (Ford et al., 2021). Simultaneously reports have emerged that question whether it is warranted for parents to sacrifice their personal life to care for children in a taxing situation of a national lockdown (Jayanetti, 2021; Joiko, 2020). For example, The Guardian interviewed three women who had given up paid employment to care for their children with mental health disorders in the context of unavailable services for such families in England. Despite the report partially framed the issue as the mothers being unable to do another ‘selfless’ job as a frontline NHS healthcare professional during the pandemic due to their child’s ill-health, the reporter concluded that when caring for a child with complex care needs “the effect on parents is often overlooked – forced to abandon their careers so they can look after their children full-time and fight endless battles with cash-strapped bureaucracies for the most basic support” (Jayanetti, 2021). Extraordinary circumstances, such as a prolonged national lockdown, may help to call further attention to the lived experiences of parental sole responsibility in the context of child morbidity. The present study has in part showcased such

parental challenges and brought them forward for discussion. This point relates to my earlier discussion that EP parents' wellbeing is an emerging public health issue as the absolute number of families with preterm born children with long-term morbidities is increasing. Scholars, as well as healthcare professionals, are well positioned to contest discursive framings of 'optimal' parenting behaviour (Widding & Farooqi, 2016). In the present study, a common feature amongst the interviewed parents' perceptions of their roles, was that their child's potential future success would constitute them as merely '*lucky*', and not be evidence of their labour-intensive parenting efforts.

6.5 Methodological Limitations and Strengths of the Study

The primary strength of this study was its mixed methods design as it allowed me to relate data from different ontological viewpoints. I was able to examine parental experiences from a wider perspective and, consequently, improve both the "depth and breadth" (Bazeley, 2018, p. 12) of the current understanding of parenting an EP YA in England. This study provided an insight into parents' lived experiences as well as evidence of parenting stress levels and parental HRQoL previously lacking from the evidence-base of long-term parent outcomes following EP birth. Nevertheless, the convergent multistrand mixed methods design of this study had limitations. I have discussed the limitations of each research strand earlier in this thesis. In this section, I will focus on the limitations of my mixed methods study design.

Firstly, I want to return to the point I presented in the methods chapter of this thesis, that the design of this convergent multistrand MM study was opportunistic rather than strictly following a defined MM design (Jang et al., 2008; Teddlie & Tashakkori, 2006). Aligned with experiences of other doctoral students (Clarke & Visser, 2018), I had not fully appreciated the challenges that my methodological approach would pose. In a convergent multistrand MM design, the research strands should remain separate until the point of interpretation and inferences (Jang et al., 2008; Teddlie & Tashakkori, 2006). Yet, as discussed earlier, in this study the two research strands interlinked at multiple points, mainly in sampling and data analyses. My earlier discussion in Chapter 3 pointed to the flexibility of MM designs. Jang et al. (2008) stated that while "mixed methods design alternatives are useful for clarifying the inquiry purpose and linking it to methodologies, in practice such design options are neither exclusive nor singular because actual mixed methods studies are often much more complex than any single-design alternative can adequately represent" (page 224). This was evident in the present study.

The findings from my systematic review demonstrated the need for a qualitative inquiry on parental experiences following EP birth. I could have chosen a purely qualitative approach, yet I was reluctant to miss the opportunity to report quantitative long-term parent outcomes in this population as these were additionally largely missing from the evidence-base among EP populations. Since my doctoral studies were situated within an ongoing longitudinal birth cohort study, it provided an outstanding opportunity for a mixed methods inquiry. Yet, both the time constrain (i.e., the need for my project to run parallel with the main study) and the resource constrains (i.e., I was the only qualitative researcher) meant that only a convergent multistrand design was feasible. For example, a sequential design of a qualitative study first, could have informed the selection of outcome instruments to be utilised in the quantitative study. Alternatively, the preliminary quantitative findings could have informed the design of the qualitative interview guide. In the present study, parenting stress and parental experiences signified distinct aspects of the construct of 'parenting', resulting in a study in which the two research strands ran parallel and interlinked but were merged with limitations. Other MM scholars have noted similar constrains (Jang et al., 2008).

Nevertheless, the context of an existing longitudinal birth cohort study provided advantages. The main study, EPICure2@11, provided me with access to a cohort of research participants and access to vast contextual data on background characteristics which would have been challenging to source otherwise. Yet, the context of a larger cohort study with its own imperatives posed limitations. Due to the data needs of the main study, the Parent Questionnaire was extensive. Consequently, I was limited with what instruments I could include the questionnaire to assess long-term parent outcomes. Ideally, in addition to parenting stress, I would have included instruments that measured levels of social support and parent mental health outcomes more specifically. Future research could explore EP parents' outcomes more widely, such as mental health outcomes, physical health, reproductive behaviour, employment outcomes, and other social outcomes by utilising validated standardised instruments. A recent Europewide effort to harmonise the VP birth cohorts' outcome data (Zeitlin et al., 2020) would provide an ideal context for a cross-sectional geographical comparison of parent outcomes among the cohorts. These findings could then direct more detailed investigations among the EP parents.

As discussed earlier in this thesis, the analyses of data were somewhat mixed. As I was working alone on the data analyses, it was impossible for me to keep them fully separate, as I could not 'unknow' what I had discovered from the quantitative and qualitative analyses, respectively. The conversation between the types of data meant that when I turned to the other analysis, I had already formed a certain idea of the data (e.g., impact of maternal caregiving

efforts, level of child disability, parent SES). This may have led me to then focus more on those aspects in the data and I potentially overlooked other features. Yet, my hypotheses that emerged from the data analyses were aligned with previous research, as has been discussed throughout this thesis. In addition, due to my role in the main study, as opposed to merely analysing the quantitative data, I had formatted the questionnaire, designed the study database, entered a part of the data, and cleaned and prepared the data sets for analyses. As was the case for the qualitative interviews. Consequently, even before embarking on the analyses, I had formed a comprehensive sense of both types of data, further strengthening the validity of the findings of this study (Braun & Clarke, 2006; Nowell et al., 2017).

The present study has strengthened the research field of prematurity with its constructivist viewpoint. As discussed earlier in this thesis, and what was demonstrated by the findings from my systematic review, in research among prematurity on long-term parent outcomes, considerations of the sociocultural aspects of constructs such as parenting have largely been lacking. The findings from the present study have demonstrated that such social understandings may not appear only in research agenda but also in parents' understandings of their roles as parents. In addition, the constructivist viewpoint may also showcase the possibility to construct the 'parent' with the presentation of data (Marvasti, 2019). For example, by reporting long-term parent outcomes that are within the normal range of outcomes among the general population in a negative tone may blur the boundaries between 'normal' and 'abnormal' and distract focus from those groups who could benefit from research attention (Macvarish & Lee, 2019).

In this thesis, I have criticised the concept of 'parenting' lacking cultural contextuality. I have discussed the Western understanding of normative parenting behaviour. Yet, simultaneously, this 'Western understanding of parenting' may vary significantly (Símonardóttir, 2016), for example among parents from different social classes (Dermott, 2012; Gillies, 2008). Scholars have suggested that parents are positioned to reflect their actions against the culture of intensive parenting. Yet some elements of the framework of parental determinism may be less dominant in a particular parenting context. In the present study, regardless that parental determinism and intensive parenting practices were present in some parents' accounts, some elements of the framework of parental determinisms, such as reliance on 'expert knowledge', were less evident. In my earlier discussion of parental determinism in the introductory chapter of this thesis, I postulated that EP parents may be particularly willing to rely on the 'expert knowledge' of parenting due to child morbidity. Yet this postulation was not fully supported in this study. Rather, many interviewed parents felt trusting about their children's outcomes which may have been supported by older child age, as discussed earlier in Chapter 5.

A telling feature of the samples of parents taking part in this study was the participants' predominant female gender. Missing perspectives of fathers among research in prematurity have been noted previously (Schuetz Haemmerli et al., 2020). In both the quantitative and qualitative samples, most respondents were women. Even though the present study does not tell how the partners of these women would have described their parenting experiences (with one exception), it is important to note that the fathers/partners did have an equal opportunity to be invited to participate; when appropriate, the posted Parent Questionnaires were addressed to both caregivers in the household. Within the family, I invited the adult who self-identified as the primary carer of the child in the Parent Questionnaire to take part in the interview.

6.6 Study Contributions and Recommendations for Future Research and Practice

I began this thesis by introducing the context of this study; I described the EPICure cohorts and challenges around defining long-term parent outcomes following preterm birth. I discussed parenting culture and discursive representations of prematurity. I then presented a systematic review of long-term parent outcomes following VP birth, to provide the reader with a complete view of previous research in the topic area. Following my research methodology, I presented findings from a cross-sectional survey about parenting stress, sociodemographic, child and parental health factors collected as a part of a national birth cohort study of extreme prematurity in England. I also explored parental experiences at the time their child transitioned to secondary education using qualitative semi-structured telephone interviews. Finally, in the preceding sections, I converged these two research approaches and further examined them in the context of previous research in the research field of prematurity. The data integration resulted in recommendations for future research and practice, with an aim to take forward research on longer term impact of extreme prematurity and improve long-term parent outcomes. In this final section, I will collate those recommendations together with key contributions of this study.

6.6.1 Key study findings relating to the research field of prematurity and suggestions for future research

This investigation has demonstrated that previous studies assessing long-term parent outcomes following VP birth have predominantly emerged from a positivistic research practice. In previous studies, parenting behaviour has been framed as a means to promote child outcomes. This study has demonstrated that dominant discursive concepts such as parental determinism appear in research practices among the research field of prematurity. Based on these findings, I recommend future research to better acknowledge and expect the socially constructed nature of phenomena and concepts under study, such as parenting, when preparing research proposals

and interpreting results on long-term parent outcomes. Special attention could be paid to explicitly state and critically examine the conceptual basis from which research proposals emerge such as the psychoanalytic and attachment theories.

Recommendation 1: Critically examine the theoretical point of departure for the research agenda and clearly define the operative meanings of concepts under study

This study has demonstrated that research on long-term parent outcomes following VP birth has been uncommon. The findings have also showed that parents may direct excessive caregiving efforts towards their children, which on occasion may affect their own health and wellbeing. Parental contributions and poor long-term parent outcomes have less often been acknowledged in the research among prematurity. Thus, a limited image of the phenomenon of extreme prematurity has been portrayed. Based on these findings, I suggest that more focus is directed towards examining long-term parent outcomes following preterm birth. In future research, along with infant survival and child disability, parent outcomes could be regarded as an integral part of the primary outcome measures following EP birth when the evidence-base is further strengthened.

Recommendation 2: Include measures of and a focus on parental items alongside the child-focused measures in longitudinal studies

6.6.2 Key study findings relating to and recommendations for practice

The findings of this study have demonstrated that a small proportion of EP parents experienced high levels of parenting stress and low levels of mental HRQoL, approximately 11 years after giving birth. Together with these findings, sociocultural factors of internalised sole parental responsibility for the child, gendered roles of parenting, and the excessive caregiving efforts cited in this study should be regarded in the context that while overall survival of infants born EP has increased, the level of morbidities among the survivors has remained unchanged. Consequently, the absolute number of families with children with morbidities will increase. Bearing in mind the labour-intensive caregiving efforts that parents of children with complex care needs described, this study has further directed attention to the health and well-being of parents who care for children with long-term morbidities. Based on these findings, I recommend that the health and wellbeing of parents who have children with long-term morbidities is regarded as an important and valid public health matter.

Recommendation 3: Regard the health and wellbeing of parents with children with long-term morbidities as an important public health matter

The findings from this study have demonstrated that the acceptance of excessive parental caregiving efforts may contribute towards an acceptance of ‘personal sacrifice’ in the parent role. From the viewpoint of a healthcare professional, I challenge the appropriateness of this finding. Aligned with other scholars, I recommend that nurses, clinicians, educators, and other professionals who liaise with families with EP children are sensitive to question and challenge the discursive framing of parenting that potentially negatively impacts parents’ personal health and wellbeing. Screening tools, such as the PSI-4-SF and SF-12v1 are readily available instruments. These tools, together with immediate scoring and interpretation, could help to initiate discussions about parent outcomes in follow-up care of preterm born children. Those parents who may benefit from further support, could be identified, and referred to follow-up services. I am conscious, however, that such practices are not likely to systematically take place until parent outcomes are widely internalised as an integral part of the long-term outcomes of EP birth.

Recommendation 4: Integrate systematic assessment of personal health and wellbeing of parents into the long-term follow-up of EP children

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Appendices

Appendix A Search terms and the search configuration for the systematic review

(exp Infant, Premature/ OR exp Premature Birth/ OR exp Infant, Extremely Premature/ OR exp Obstetric Labor, Premature/ OR exp Infant, Extremely Low Birth Weight/ OR exp Infant, Very Low Birth Weight/ OR exp Infant, Low Birth Weight)

AND

(exp Parenting/ OR exp Single-Parent Family/ OR exp Parent-Child Relations/ OR exp Parents/ OR exp Single Parent/ OR exp Education, Parenting/ OR exp Parenting Education/ OR exp Family Therapy/ OR exp Family Health/ OR exp Family Relations/ OR exp Family/ OR exp Aid to Families with Dependent Children/ OR exp Family Characteristics/ OR exp Nuclear Family/ OR exp Domestic Violence/ OR exp Family Conflict/ OR exp Mothers/ OR exp Mother-Child Relations/ OR exp Maternal Age/ OR exp Maternal Death/ OR exp Maternal Exposure/ OR exp Maternal-Child Nursing/ OR exp Maternal Welfare/ OR exp Maternal Health/ OR exp Maternal Deprivation/ OR exp Maternal Behavior/ OR exp Educational Status/ OR exp Fathers/ OR exp Father-Child Relations/ OR exp Paternity/ OR exp Paternal Behavior/ OR exp Paternal Deprivation/ OR exp Paternal Age/ OR exp Paternal Death/ OR exp Paternity Benefits/ OR exp Sibling Relations/ OR exp Siblings)

AND

(exp Adolescent Behavior/ OR exp Psychology, Adolescent/ OR exp Adolescent, Institutionalized/ OR exp Adolescent/ OR exp Adolescent Psychiatry/ OR exp Adolescent Development/ OR exp Adolescent, Hospitalized/ OR exp Adolescent Health/ OR exp Adolescent Health Services/ OR exp Adolescent Medicine/ OR exp Adolescent Welfare/ OR exp Adolescent Well Being/ OR exp Adolescent Well-Being/ OR exp Underage Drinking/ OR exp Young Adult/ OR exp Personality Development/ OR exp Puberty/ OR exp Puberty, Delayed/ OR exp men/ OR exp women)

Appendix B Tool to assess risk of bias in cohort studies utilised in the systematic review of long-term parent outcomes following very preterm birth

Tool to Assess Risk of Bias in Cohort Studies

1. Was selection of exposed and non-exposed cohorts drawn from the same population?

Definitely yes (low risk of bias)	Probably yes	Probably no	Definitely no (high risk of bias)
--------------------------------------	--------------	-------------	--------------------------------------

Examples of low risk of bias: Exposed and unexposed drawn from same administrative data base of patients presenting at same points of care over the same time frame

Examples of high risk of bias: exposed and unexposed presenting to different points of care or over a different time frame

2. Can we be confident in the assessment of exposure?

Definitely yes (low risk of bias)	Probably yes	Probably no	Definitely no (high risk of bias)
--------------------------------------	--------------	-------------	--------------------------------------

Examples of low risk of bias: Secure record [e.g. surgical records, pharmacy records]; Repeated interview or other ascertainment asking about current use/exposure

Examples of higher risk of bias: Structured interview at a single point in time; Written self report; Individuals who are asked to retrospectively confirm their exposure status may be subject to recall bias – less likely to recall an exposure if they have not developed an adverse outcome, and more likely to recall an exposure (whether an exposure occurred or not) if they have developed an adverse outcome.

Examples of high risk of bias: uncertain how exposure information obtained

3. Can we be confident that the outcome of interest was not present at start of study

Definitely yes (low risk of bias)	Probably yes	Probably no	Definitely no (high risk of bias)
--------------------------------------	--------------	-------------	--------------------------------------

4. Did the study match exposed and unexposed for all variables that are associated with the outcome of interest or did the statistical analysis adjust for these prognostic variables?

Definitely yes (low risk of bias)	Mostly yes	Mostly no	Definitely no (high risk of bias)
--------------------------------------	------------	-----------	--------------------------------------

Examples of low risk of bias: comprehensive matching or adjustment for all plausible prognostic variables

Examples of higher risk of bias: matching or adjustment for most plausible prognostic variables

Examples of high risk of bias: matching or adjustment for a minority of plausible prognostic variables, or no matching or adjustment at all. Statements of no differences between groups or that differences were not statistically significant are not sufficient for establishing comparability.

5. Can we be confident in the assessment of the presence or absence of prognostic factors?

Definitely yes (low risk of bias)	Probably yes	Probably no	Definitely no (high risk of bias)
--------------------------------------	--------------	-------------	--------------------------------------

Examples of low risk of bias: Interview of all participants; self-completed survey from all participants; review of charts with reproducibility demonstrated; from data base with documentation of accuracy of abstraction of prognostic data

Examples of higher risk of bias: Chart review without demonstration of reproducibility; data base with uncertain quality of abstraction of prognostic information

Examples of high risk of bias: Prognostic information from data base with no available documentation of quality of abstraction of prognostic variables

6. Can we be confident in the assessment of outcome?

Definitely yes (low risk of bias)	Probably yes	Probably no	Definitely no (high risk of bias)
--------------------------------------	--------------	-------------	--------------------------------------

Examples of low risk of bias: Independent blind assessment; Record linkage; For some outcomes (e.g. fractured hip), reference to the medical record is sufficient to satisfy the requirement for confirmation of the fracture.

Examples of higher risk of bias: Independent assessment unblinded; self-report; For some outcomes (e.g. vertebral fracture where reference to x-rays would be required) reference to the medical record would not be adequate outcomes.

Examples of high risk of bias: uncertain (no description)

7. Was the follow up of cohorts adequate?

Definitely yes (low risk of bias)	Probably yes	Probably no	Definitely no (high risk of bias)
--------------------------------------	--------------	-------------	--------------------------------------

Examples of low risk of bias: No missing outcome data; Reasons for missing outcome data unlikely to be related to true outcome (for survival data, censoring is unlikely to introduce bias); Missing outcome data balanced in numbers across intervention groups, with similar reasons for missing data across groups; For dichotomous outcome data, the proportion of missing outcomes compared with observed event risk is not enough to have a important impact on the intervention effect estimate; For continuous outcome data, plausible effect size (difference in means or standardized difference in means) among missing outcomes is not large enough to have an important impact on the observed effect size; Missing data have been imputed using appropriate methods.

Examples of high risk of bias: Reason for missing outcome data likely to be related to true outcome, with either imbalance in numbers or reasons for missing data across intervention groups; For dichotomous outcome data, the proportion of missing outcomes compared with observed event risk is enough to induce important bias in intervention effect estimate; For continuous outcome data, plausible effect size (difference in means or standardized difference in means) among missing outcomes is large enough to induce clinically relevant bias in the observed effect size.

8. Were co-Interventions similar between groups?

Definitely yes
(low risk of bias)

Probably yes

Probably no

Definitely no
(high risk of bias)

Examples of low risk of bias: Most or all relevant co-interventions that might influence the outcome of interest are documented to be similar in the exposed and unexposed.

Examples of high risk of bias: Few or no relevant co-interventions that might influence the outcome of interest are documented to be similar in the exposed and unexposed.

Appendix C Supplementary Table 1 Reasons why children of consented families did not undergo clinical child assessment (n=20)

Reason for child not being assessed	Frequency (n)	Percentage (%)
Unable to get in contact with family or school following consent	14	70.0
Unable to find a suitable assessment time	2	10.0
Family emergency	2	10.0
Living abroad	1	5.0
Withdrew	1	5.0
Total	20	100.0

Appendix D Supplementary Table 2 A comparison of sociodemographic characteristics between parents with complete data (n=279) and parents with data missing or incomplete (n=21) among participants who returned their Parent Questionnaires (Total Stress score, child assessment and/or IMD decile)

	Data available n=279/300 (93.0%)	Data missing or incomplete n=21/300 (7.0%)	<i>p</i> value¹
Parent Characteristics			
Respondent			
Mother	248 (90.8)	17 (89.5)	
Father	21 (7.7)	2 (10.5)	0.79
Carer/Foster parent/Grandparent/Friend	4 (1.5)	0 (0)	
Age, median (IQR)	46.0 (41.9-49.3)	44.4 (42.9-50.6)	0.51
Ethnicity			
White	199 (71.3)	15 (71.4)	
Mixed/Multiple ethnic groups	9 (3.2)	0 (0)	0.72
Asian/Asian British	39 (14.0)	1 (4.8)	
Black/African/Caribbean/Black British	26 (9.3)	3 (14.3)	
Other ethnic groups	6 (2.2)	2 (9.5)	
Employment status			
Employed/Self-employed/Full-time student	220 (79.1)	15 (79.0)	
Retired/Semi-retired/Long-term illness	6 (2.2)	0 (0)	0.93
Homemaker/Carer	38 (13.7)	3 (15.8)	
Unemployed	14 (5.0)	1 (5.3)	

ST 2 (continued)

	Data available n=279/300 (93.0%)	Data missing or incomplete n=21/300 (7.0%)	p value ¹
Marital status			
Married/Living with a partner	222 (79.9)	14 (66.7)	
Separated/Divorced/Widowed	31 (11.2)	1 (4.8)	0.02
Single	25 (9.0)	6 (28.6)	
Living with the father or mother of the study child	211 (75.9)	12 (57.1)	0.06
Highest academic qualification			
University degree or above	114 (43.9)	11 (64.7)	
Some post-secondary education	74 (28.5)	1 (5.9)	0.10
Secondary education or less	72 (27.7)	5 (29.4)	
Receiving benefits			
Family benefits, income support, tax credits	151 (54.1)	15 (71.4)	0.12
Incapability and disability benefits	61 (21.9)	9 (42.9)	0.03
Receiving more than 1 type of benefit	45 (15.4)	9 (42.9)	0.001
Child characteristics²			
Male sex	134 (48.0)	11 (52.4)	0.70
Age at the assessment, mean (SD)	11.8 (0.57)	11.6 (0.68)	0.37
Number of siblings, mean (SD)	1.2 (1.0)	1.5 (0.8)	0.20
School type			
Mainstream	257 (92.5)	17 (85.0)	
SEN school or unit	18 (6.5)	3 (15.0)	0.32
Home educated	3 (1.1)	0 (0)	

ST 2 (continued)

	Data available n=279/300 (93.0%)	Data missing or incomplete n=21/300 (7.0%)	p value ¹
Overall neurodevelopmental disability			
None or mild	219 (78.5)	11 (61.1)	
Moderate	35 (12.5)	2 (11.1)	0.04
Severe	25 (9.0)	5 (27.8)	

Data are presented in n (%) unless otherwise stated

Note: Missing data accounts for differing totals

¹Chi Square Test for categorical variables and Welch's t-Test for continuous variables were used

²Those assessed and Parent Questionnaire returned (EP n=172 and FT n=125)

Appendix E Supplementary Table 3 A comparison of characteristics at birth between those biological parents who returned their Parent Questionnaires at 11 years (n=171) and mothers in the remaining EPICure 2 cohort (n=866)

Characteristics	Parent Questionnaire returned n=171/1037 (16.5%)	No Parent Questionnaire n=866/1037 (83.5%)	p value¹
Infant male sex	87 (50.9)	415 (47.9)	0.48
IMD at delivery, mean (SD)	4.7 (2.6)	4.2 (2.9)	0.03
Multiple birth	43 (25.2)	208 (24.0)	0.75
Mother's age at birth, mean (SD)	31.6 (5.8)	29.2 (6.6)	<0.001
Maternal ethnicity			
White	110 (64.3)	558 (65.3)	
Mixed/Multiple ethnic groups	5 (2.9)	16 (1.9)	
Asian/Asian British	28 (16.4)	100 (11.7)	0.05
Black/African/Caribbean/Black British	23 (13.5)	170 (19.9)	
Other ethnic groups	5 (2.9)	10 (1.2)	

Data are presented in n (%) unless otherwise stated

Note: Missing data accounts for differing totals

¹Chi Square Test for categorical variables and Welch's t-Test for continuous variables were used

Appendix F Participant information sheet for the semi-structured telephone interviews



EPICure2@11 - Parent Study

Information about the telephone interview

We are inviting you to take part in a telephone discussion to help us understand your experiences as a parent. We are particularly interested in your aspirations and ambitions as a parent, and potential hurdles and issues at the time your child is transitioning to secondary school.

Please take the time to read the following information and let us know if anything is unclear or if you have any concerns.

What is the purpose of the study? In addition to the excitement and happiness parents may feel close to the time of their child's birth and during the early childhood, they may also find this period stressful. This occurs particularly if parents have concerns about their child's health. Studies have also shown that feelings of stress may continue or re-occur when a child approaches adolescence.

We would like better to understand parents' feelings and experiences about parenting at the time their child transitions to adolescence. We hope the findings from this research will influence the development of services available to parents and allow us to improve support for families at all stages of their child's development.

Who is funding and organising this research? The EPICure2@11 study is funded by the Medical Research Council (MRC). The Parent sub-study is being undertaken by a researcher Ms Emmi Suonpera based at University College London (UCL). Professor Neil Marlow is responsible for the project and all the data.

Who has reviewed the study? To protect your interests, all research based at UCL is looked at by an independent group of individuals called a Research Ethics Committee. This study has received a favourable opinion by the UCL Research Ethics Committee (Ref:10175/001).

Why have I been asked to take part? We are inviting all parents in the EPICure2@11 study to consider taking part in the telephone discussion. *We are interested in everyone's opinions, even if you have not experienced stress or other challenges in your parenting.*

Do I have to take part? No. It is entirely your decision whether you would like to take part in the interview or not. If you choose not to take part, or decide to withdraw from the interview at any time, it will not affect your child's participation in the EPICure2@11 study in any way. If you do not wish to take part or you change your mind about taking part at any time, you do not need to provide a reason.

What will I have to do if I take part? If you are willing to consider taking part, the EPICure Study Co-ordinator will pass your phone number or email address to the researcher, who will contact you to setup a convenient date and time for you for a brief telephone interview.

Specifically, on the day of the interview the researcher will call you on the telephone and confirm that you still wish to participate in an approximately half an hour conversation. The discussion will cover the context of your family life, your parenting experiences, your perceptions about your child's characteristics, thoughts about her/him growing up and parental support.

You don't need to answer any questions you don't want to and you don't need to disclose any personal information if you don't want to. If you want to stop at any point, you just need to let the researcher leading the discussion know.

Will my information be kept confidential? At the start of the interview we will ask your permission to record the interview. The recording will be held on a password-protected recorder and deleted as soon as it is transferred to a secure university computer. The only people who will listen to the recorded discussion are the researcher and the team typing up the record. All answers that you provide us are anonymous and will be kept securely at UCL. We do not ask for names so you cannot be identified from your answers. We will not

share the information you give us, unless we are concerned that you are at risk of harming yourself or others, and then only with your permission. Any notes will be kept in a locked cabinet in the researcher's office and all information gathered will be kept confidentially and securely for a time specified in the UCL data protection policies. It will then be destroyed.

What will happen to the results of the study? Results will be written up for publication in academic journals. When the results are presented, no sound recordings or names will be used and any quotes will be anonymous. If you are interested, we can send you a copy of the results and we will post summaries of the results on the EPICure website (www.epicure.ac.uk).

What are the possible benefits or advantages of taking part? We cannot promise that the study will help you, but the information from this study will help us to learn more about your experiences as a parent. You may enjoy knowing that you are contributing to the improvement of services in the future and you may find it helpful sharing your experiences with someone else.

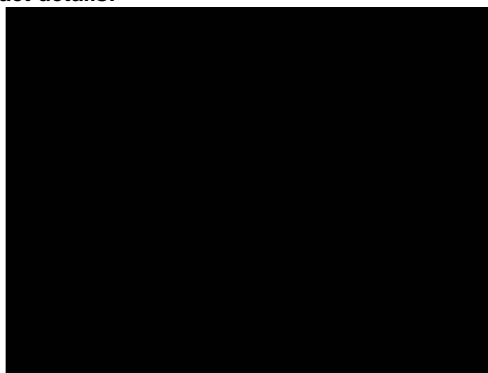
What are the possible disadvantages of taking part? We believe that it is unlikely that you will come to any harm through taking part in the interview. However, it is possible that asking you to talk about potentially difficult experiences could be upsetting for you. The conversation will be very open so you can choose what experiences you feel comfortable about sharing; we can stop the conversation at any time.

What if I feel upset after the interview or wish to discuss something further? We understand that some people may find some of the questions upsetting. If you are upset by any of the issues we talk about please let the researcher know. We can find you further support.

What if there is a problem? If you have any complaints or concerns about the research you can complain via UCL. Please contact the Chief Investigator Professor Neil Marlow using the email address: epicure@ucl.ac.uk.

Thank you very much for taking the time to consider participating in this study

Contact details:



Study website: www.epicure.ac.uk

Support and advice relating to parenting is available from:

Bliss helpline:

Tel: 0808 801 0322

Email: hello@bliss.org.uk

Information can also be found on BLISS website:

<https://www.bliss.org.uk/Pages/Category/support>

Or you can find local support services on NHS CHOICES website:

<http://www.nhs.uk/pages/home.aspx>

Appendix G Topic guide for the semi-structured telephone interviews

PARENTING EXPERIENCES – TOPIC GUIDE FOR THE TELEPHONE INTERVIEWS

Topic areas:

- A) FAMILY COMPOSITION AND CONTEXT
- B) CHILD CHARACTERISTICS
- C) TRANSITION TO ADOLESCENCE
- D) PARENTING EXPERIENCES
 - D1) INFLUENCES
 - D2) IDENTITY / LIFE PERCEPTION
 - D3) PARENTAL SUPPORT

Examples of questions and possible points of discussion:

A) FAMILY COMPOSITION AND CONTEXT

- Could you tell me about your family?
- Could tell me about the 'make up' of your family
- Could you tell me about your family's daily life?

"I have seen your parent questionnaire so I know little bit about your family but could you just..."

- Members
- Other children in the family
- Extended family
- Significant others - daily/intermittent
- Daily life
- Living area
- Work
- School
- Hobbies
- Evenings

B) CHILD CHARACTERISTICS

- If you were to think about X how would you describe her/him as a child?
- If you were to think about X as a child, is there something that has impacted your parenting?

"I briefly met [] during the assessment so I know little bit about him/her but how would you describe [] as a child..."

You mentioned x/prematurity/illness **could you give me an example from your daily life?**

How your parenting has been affected by/how have you managed /coped with this?

Consideration on other siblings and how they may have been affected

Do you think there are factors that influence your child other than your parenting?

Sort of things like characteristics or personality

Emotional/behavioural characteristics, Developmental/learning difficulties

C) TRANSITION TO ADOLESCENCE

“we briefly touched the topic of x starting secondary school now in September,

could you tell me little bit more about that? How has it been?

“... has now started secondary school, right, how has that been/how do you feel about it?”

How growing up has influenced your experiences as a parent?

[] has started secondary school/will start soon...is that something you think about?

What do you think about the change of schools?

Have other siblings started secondary school, - does it feel different?

What do you think is expected from you as a parent during this time?

Do you think that differs to what has been before?

How do you feel about having these expectations, tasks, or roles?

How do you feel about the change as a parent, are you still needed as much as before?

What do you do in your daily life to meet these expectations, tasks, or roles?

Do you have any particular ambitions for your child?

Are there particular challenges in reaching these ambitions?

How do you support x in reaching these goals?

If you were to think about your role as a parent in future, how do you see it?

Thinking about future, how do you feel about it? Is that something you think about?

D) PARENTING EXPERIENCE

“Now I would like to move on a little bit and ask more about your experiences as a parent overall, so if you were to think about”

-> role as a parent -> influences -> priorities -> goal for parenting -> challenges achieving these goals -> support in parenting

D1) INFLUENCES (and how it is seen!)

What factors influence your parenting?

For example, is there something that influences “how you do things” as a parent?

Are there things that particularly influence your parenting?

Could you explain me little bit more about that, how do you see it?

Could you give an example of that?

Do you have priorities as a parent? –something that has always been important for you in your parenting?

Is there something that you find challenging in your parenting?

What would you need to achieve these goals?

Notes:

You mentioned xxx earlier is that something that influences your parenting?

Are these something that you prioritise in your parenting?

You mentioned ... is that something that you see as a goal in your parenting?

D2) IDENTITY / LIFE PERCEPTIONS

If you were to think about yourself as a person, is that something that impacts your parenting?

Do you think that your personality influences your parenting?

Emotional positivity

Social confidence

Feelings of competence

Worry

Can do attitude

A balanced view

Resilience

Humour

Notes:

Sort of things like... personality, outlook on life?

Wear a different hat, as a mother how do you feel about that?

As a person what do you think of that?

You mentioned x earlier, could you tell me more about it?

Could you give me an example from your daily life? Perhaps a recent one.

Some other people have mentioned this ... , what do you think of it?

Why do you think that might be?

How do you feel about it?

Are there any things that you would change in your parenting?

If you had unlimited resources, what would you do differently?

Notes:

“ouh that sounds like your days are full, from morning till evening, do you feel like you get time for yourself as well? Do you find time to relax?”

Up to now, we've been talking about XXX. Now I'd like to ask you about...'

You just mentioned X that reminded me to ask you about...

D3) PARENTAL SUPPORT

Now that we have discussed a lot about the transition to secondary school and possible changes ahead, what do you feel you need as a parent to assist you through the transition years?

Do you receive support in your role as a parent?

Who do you talk to about the topics discussed above?

Do you share responsibilities with your partner?

Do you see that you and your partner have similar roles in parenting?

The role that you have as [] parent is very important and valuable; do you feel like you receive support in your role as a parent?

Can do attitude

Lack of support

Not being able

Barriers limiting possibilities

Availability of services for growing children

Support during early years

Long lasting connections with support services

Notes:

Cooling down: Is there something else you would like to discuss about that has not been mentioned yet?

What you have shared with me is really important information and will be very useful for the study. Thank you so much for taking time to talk with me and for sharing your experiences