VOLUME 1

PARENTAL ADJUSTMENT TO THE BIRTH OF A CHILD WITH CLEFT LIP AND PALATE: THE ROLE OF PRENATAL DIAGNOSIS

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By

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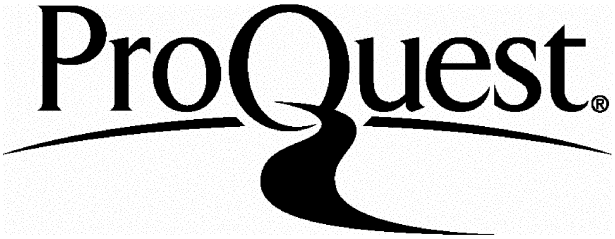

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ABSTRACT

This study aimed to explore parents' experience of the birth of a child with cleft lip and palate, and more specifically, to investigate the impact of prenatal diagnosis of cleft lip and palate on parents' adjustment to the birth of their baby. It was hypothesised that detection of cleft during pregnancy would be associated with successful parental adjustment, following the birth of a child with cleft lip and palate. Adjustment was assessed using three measures: the Reaction to Diagnosis Interview (Pianta & Marvin, 1992), which evaluates parental resolution of grief and trauma associated with the diagnosis of a child's disability; the Indicators of Parental Adjustment to the Birth of a Child with a Congenital Disfigurement (Bradbury & Hewison, 1994); and the General Health Questionnaire (GHQ-28).

Two groups of mothers participated in the study: 20 mothers who had received a prenatal diagnosis of cleft lip and palate, and 24 mothers for whom the diagnosis was given at birth. Comparisons were made on levels of adjustment found in the two groups. Statistical analysis of the interview and questionnaire data obtained, indicated that prenatal detection of cleft lip and palate was associated with more successful adjustment, and more specifically, was related to parental resolution of the trauma and grief following the birth of a child with cleft. Furthermore, results indicated that certain stressors experienced by mothers during the perinatal period were also related to parental adjustment. Possible explanations for the results are discussed, and the theoretical and clinical implications of the findings are explored.

INTRODUCTION

Overview

The birth of a child with cleft lip and palate can have a significant impact on the parent, not only in terms of initial emotional reactions to the birth, but also with respect to potentially stressful caretaking demands that arise during infancy and the preschool years. New developments in antenatal care now mean that for many parents, the diagnosis of cleft lip and palate is given during pregnancy. This study aims to investigate parents' experience of the birth of a child with cleft lip and palate, and in particular, the impact that prenatal diagnosis may have on parental adjustment to the birth. In order to explore this issue, it is important to consider a number of areas of relevant research. Firstly, the psychosocial impact of parenting a child with cleft lip and palate, in addition to the impact of disfigurement on the child, will be explored. Consideration will then be given to possible precursors of the difficulties experienced by parents and children. Issues relating to developments in prenatal screening and diagnosis, and to women's experiences of these procedures, will be examined. The present study will then be described.

Cleft Lip and Palate

Cleft lip and / or palate is one of the most frequent human congenital malformations, with the incidence estimated at approximately 1 in 700 live births (McCarthy, 1990). Both cleft lip and cleft palate are the result of arrested midline facial development during the first trimester of pregnancy. Cleft lip may be unilateral or bilateral and may involve the bones of the upper jaw and / or upper gum ridge as well as the lip, causing mild to severe nasal distortion. Clefts may occur in both the hard or the soft palate. In 75 to 80 per cent of cases the cleft lip is unilateral, and in two thirds of cases it is associated with a cleft palate (Melnick, 1990; Bronshtein, Masiah, Blumenfeld, & Blumenfeld, 1991).

Incidence rates of orofacial clefts vary by ethnic group and by gender, but not by maternal age or socio-economic status (Melnick, 1990). Cleft lip and palate is twice as

common in male fetuses, although in the case of cleft palate alone, males are affected 25 per cent less than females. The prevalence rate of cleft is higher in the asian population, at 1.5 - 2.0 per 1000, compared with approximately 1 per 1000 in the white population, and 0.5 per 1000 in the black population (Melnick, 1990).

Cleft lip and palate may be associated with other fetal abnormalities and can occur as part of a chromosomal or other malformation syndrome, however the majority of cases are isolated. There is a tendency for cleft lip and palate to be accompanied by a range of difficulties including feeding problems associated with the cleft palate and the inability to form adequate oral suction (Clarren, Anderson & Wolf, 1987); chronic ear infections and hearing loss (Hayes, 1994); speech impairments (McWilliams, Morris & Shelton, 1990); and numerous dental and orthodontic problems (Dahllof, Ussisoo-Joandi & Idelberg, 1989).

Despite decades of intensive investigation into the cause of cleft lip and palate, the pathogenesis remains unclear (Melnick, 1990). For the most part, a child with a cleft will be born with no previous family history. However, in some cases there does appear to be a genetic component. Positive family history has been reported in 31 to 51 per cent of cases (Fogh-Anderson, 1971; Bhatia, 1972), and family studies have shown that the presence of a family history of cleft significantly increases the risk of recurrence after the birth of a child with isolated cleft lip and / or palate. Currently the most widely accepted theory pertaining to the aetiology of cleft lip and palate, is a multifactorial model of inheritance (Holder, 1991). This theory contends that the trait is determined by a number of genes at different loci interacting with various, largely unknown, environmental factors.

In most cases the cleft lip is surgically repaired by the end of the third to fourth month of life, with some repairs carried out during the first two weeks. The cleft palate is repaired later, almost always by the end of the first year (Kaufman, 1991). Children subsequently face multiple secondary surgeries and medical interventions that may extend into adolescence, including surgery to reduce facial scarring, bone grafting to support jaw repair, and surgery to reduce nasal airway emissions that affect the quality of speech (The Royal College of Surgeons of England, 1995).

The Impact of Parenting a Child with Cleft Lip and Palate

Initial Responses to the Birth

The birth of any child creates significant disruption within the family, necessitating readjustment and reorganisation within the family system (Hittleman, Emde & Simmons, 1985). For parents whose child is born with a congenital abnormality, the shock may be overwhelming. A number of studies have explored initial parental reactions to the birth of a child with facial cleft, where there has been no prior knowledge of the abnormality (Bradbury & Hewison, 1994; Spriestersbach, 1973; van-Staden & Gerhardt, 1995). Psychological responses documented include shock, denial, anger, and intense distress. The parent may also harbour negative feelings towards the 'imperfect' child (Lax, 1972). Loss of control and a sense of helplessness engendered by the birth has also received attention in the literature; despite all the parent has done to ensure a healthy pregnancy, they have not been able to control the outcome (Bradbury & Hewison, 1994).

On the whole, parental reactions to the birth of a child with a visible anomaly have been conceptualised within a bereavement framework. Klaus & Kennell (1984) proposed that parent's responses progressed through the following stages: shock, denial, sadness and equilibrium. This process has been investigated by several researchers. van-Staden & Gerhardt (1995) explored parental reactions to the birth of a child with facial cleft by means of an open-ended questionnaire. It was found that initial reactions to the child's cleft were characterised by shock and disorganisation, and that parents' reappraisals over time resulted in a phase of reorganisation. A progressive change from loss and threat appraisals to assessing the situation primarily as a challenge was noted. These findings are echoed in a study by Clifford & Crocker (1971), which describes the universal sense of shock experienced by parents following the birth of a baby with cleft lip and palate, but found that this shock dissipated once parents were given information about the possibilities of surgical correction.

The earliest attempts at developing an understanding of parental responses to the birth of a child with a congenital abnormality can be found in psychoanalytic literature. A major contribution to this work comes from the writing of Solnit & Stark (1962), which draws on analytic concepts of narcissism and object loss in order to understand parental responses to the birth of an 'imperfect' baby. Parental responses are conceptualised in terms of mourning for the loss of the healthy, expected baby. Solnit & Stark (1962)

view this mourning process as similar in intensity to the mourning of a dead baby, but suggest that it may take longer to resolve as it becomes complicated by the needs of the living, 'imperfect' child.

This mourning process was considered by Solnit & Stark (1962) as an adaptive response, which facilitated resolution and adjustment. Positive outcome was identified as the parent's ability to separate from the child and integrate the child into the family and to cease devaluing themselves and the child, thereby developing a perception of the child as a separate entity with strengths and weaknesses. However, two extreme reaction patterns that obstruct the normal resolution of grief were identified: the parent who is unable to tolerate her own negative feelings and whose sense of guilt leads her to dedicate herself unremittingly to her child; and the parent whose sense of 'narcissistic injury' proves intolerable, leading her to deny her relationship to the child. Both patterns of responses were thought to obstruct adjustment to the child's abnormality.

This idea has been expanded by other psychoanalytic writers. Lax (1972), described the process of pregnancy as maternal narcissistic self absorption, as the mother merges with the developing baby and experiences the growing child as part of herself. The mother is thought to experience the birth of the 'imperfect 'baby as a 'narcissistic blow' to her self-esteem, as she experiences the child's impairment as her own impairment, thus devaluing both herself and the baby. It was proposed that the degree of distress caused by this was related not to the severity of the impairment, but to factors relating to the woman, and in particular to her relationship with her own mother.

Lax (1972) identified three patterns of responses which enabled the mother to protect herself from this trauma and which reflected the mother's characteristic ways of dealing with conflict. These patterns are expressed as:

* denial of the reality of the child's condition;
* rejection of the child;
* or conversely, extreme over-protection resulting in problems with separation.

The theme of mourning has been continued in the work of Benkendorf (1987). Drawing on her clinical work as a genetic counsellor, she has identified the task of every new parent as reconciling the discrepancy between the fantasised and the real baby. This was seen as part of the process of adjustment experienced by all parents. However, parents of a child with an anomaly are additionally faced with the dual task of mourning and

acceptance. Benkendorf (1987) focused on different themes of mourning including anger and guilt, yet rather than conceptualise the process as a rigid series of stages, mourning was seen as a long process which can involve set backs and reversals, and which follows no predetermined, 'normal' grieving pattern.

Despite its limitations, and in particular the absence of an empirical base, the psychoanalytic approach has been helpful in its emphasis of the psychological processes within the parent as determining and mediating parental responses to the baby, rather than viewing the severity of the anomaly as being the key determinant of those responses. However, the tendency to interpret all responses in terms of mourning, or as defending self-esteem within a narcissistic framework, may be limited. Not all parents follow this observed process of adjustment. Bradbury & Hewison (1994) noted a wide range of responses to the birth of an infant with cleft lip and palate, and found that parents varied considerably in their ability to adapt and adjust to the baby. Some parents experienced great emotional difficulties in coming to terms with their baby, whilst for others, there was unconditional acceptance. Thus, the bereavement model that has predominated the literature, may not necessarily be the only framework in which to conceptualise and understand parental responses to the birth of a child with cleft lip and palate. Recognition of the positive process of adaptation, is also needed.

Stressors During Infancy and Preschool Years

In addition to parents' initial efforts to cope emotionally with having a facially disfigured infant, they may be challenged by a series of potentially stressful demands during infancy and the preschool years. Feeding problems can be severe, resulting from a difficulty in obtaining negative suction, and require specialist feeding methods. Regurgitation of food through the nose is a further problem which parents can find particularly distressing (Speltz, Armsden & Clarren, 1990; Spriestersbach, 1973). The multiple surgeries that infants undergo during the first year of life can also be extremely stressful for families, involving hospitalisations that may last a week to 10 days, cosmetic outcomes that may not meet parental expectations, and extensive post-surgery care (Speltz, Endriga, Fisher & Mason, 1997). In addition to dental and orthodontic problems, hearing difficulties and language delays, families of infants with cleft lip and palate experience negative social reactions to the infant's unusual appearance, and have to contend with the social stigmatisation associated with facial disfigurement (Bull & Rumsey, 1988).

The impact of parenting a child with craniofacial abnormality has been examined by Speltz *et al.* (1990). When compared with mothers of healthy children matched for age and socio-economic status, mothers of preschool-aged children with craniofacial anomalies reported significantly higher levels of stress. Parenting of a young child with craniofacial anomalies was also associated with poorer psychological well-being, and less confidence in parenting skills. This same group of mothers continued to report poorer emotional health several years later (Speltz *et al.*, 1993).

A number of hypotheses have been proposed to explain the lower sense of parental competence observed in mothers of infants with orofacial clefts. It has been suggested that difficulties in adapting to demanding caregiving tasks such as feeding, may leave parents feeling inadequate (Trout, 1983). Alternatively, lower evaluations of self-competence may be the result of a tendency for mothers to attribute the caregiving difficulties of parenting a child with cleft, to self-, rather than child, limitations (Speltz *et al.*, 1990). Although this might be regarded as an unhealthy cognitive process for these mothers, it may have some adaptive significance for both mother and child. Previous research has shown that parental self-blame for problematic child outcomes may facilitate coping. It has been suggested that when parents attribute responsibility on themselves, this allows for a more controllable, and potentially modifiable situation from the child's perspective (Affleck, McGrade, Allen & McQueeney, 1985; Horan, 1982). Thus, mothers of children with clefts who attribute caregiving problems to their 'ineffective' parenting rather than to the child's disability may be more inclined to believe they can facilitate, through future effort, the child's development. Minimising negative attributions regarding the child's characteristics may also serve to promote secure child attachment by preserving positive maternal feelings and behaviour (Speltz *et al.*, 1990).

Research has suggested that parental adaptation to a child with a congenital abnormality may be influenced by the variables such as the quality of the marital relationship, and the adequacy of the social support network (Benson, Gross, Messer, Kellum & Passmore 1991; Friedrich, 1979). It is generally accepted that utilisation of a functioning social support system can be a powerful mediator in the reduction of stress and can positively influence an individual's psychological well-being and physical health (e.g., Mueller, 1980). However, the literature suggests that families of children with craniofacial anomalies do not have adequate support systems.

In an investigation into the social support networks available to families of infants and preschool children with craniofacial anomalies including cleft lip and palate, Benson and his colleagues (1991) identified that parents in the craniofacial group reported significantly less social support and less satisfaction from their social support networks, than did parents in the control group. Furthermore, parents of children who have more severe physical impairments and were rated as less attractive, reported having less available and less satisfying social support.

Several possible explanations for the lack of social support that these parents appear to experience have been suggested (Benson, *et al.*, 1991). Firstly, friends and family members may feel uncomfortable around a child with a craniofacial abnormality, and as such, may have difficulty accepting and responding to a child with a disability. Consequently, they may reduce or avoid social contact. Conversely, it is possible that parents overprotect their child, thereby alienating themselves from an existing social support network (Richman & Harper, 1978). The demands of caretaking may also prevent parents from participating in the lifestyle they had previously enjoyed, thus reducing social contacts and forcing parents of a child with a craniofacial anomaly to become isolated.

Parental embarrassment or an inability to accept the child may also place strain on social outlets. Spriestersbach (1973) explored parents' ability to tolerate the exposure of their child to strangers in a group of mothers of infants with clefts. Results indicated that half of the parents in his group were reluctant to take the baby out prior to surgery, whilst a significant number of mothers reported initial rejection of the child by close relatives, in addition to rejection of the infant by close friends. The absence of a control group is a limitation of this study, however these findings have been substantiated elsewhere. Bradbury & Hewison (1994) reported that many parents of infants with visible clefts were anxious about their infant's exposure to public view and attempted to conceal the infant's impairment. This study also found that parents whose own parents were unsupportive showed the greatest difficulty in adjusting to their infants.

There is also evidence that the marital relationship is negatively affected following the birth of a child with cleft lip and palate. Rates of separation and divorce do not appear to be significantly higher among parents of children with craniofacial abnormalities than parents of non-impaired children (Benson *et al.*, 1991). However, parents of infants with clefts do report significantly greater marital conflict (Benson *et al.*, 1991; Speltz *et al.*,

1990). The birth of a child with cleft lip and palate may place extraordinary stress on the marital dyad in numerous ways. In addition to the emotional turmoil that parents may experience following the birth of the 'imperfect' child, difficulties may arise between parents concerning parental blame for the child's condition, or the responsibility of the child's care (Benson & Gross, 1989). Parents may also need to re-evaluate plans of having additional children in cases where genetic factors must be considered (Carver & Carver, 1972). Furthermore, the feeding difficulties experienced by many children with cleft lip and palate necessitate time-consuming, specialised care. This may mean that parents have less time and energy to devote to the marital relationship (Benson & Gross, 1989). Additionally, parents may experience sexual difficulties following the birth of a child with an anomaly. This has been attributed to exhaustion from caregiving, fear of conception and poor communication (Benson *et al.*, 1991).

It is clear that the birth of a child with cleft lip and palate can have a significant impact on the parent. However, it has been hypothesised that parental adjustment is of particular importance, not just for the parent's well-being, but also for the child's psychological and social development (Bradbury & Hewison, 1994).

The Psychosocial Impact of Disfigurement on the Child

Research has examined whether children with cleft lip and palate have an elevated risk for psychosocial problems. Much of this work has focused on the behavioural adjustment and educational status of affected school-aged children and adolescents (Eder, 1995; Speltz, Armsden & Clarren, 1990). On the whole, studies in this area are fraught with methodological limitations such as small sample size and non-standardised measures, and results have been contradictory, depending to a large extent on how psychopathology and achievement were measured. Nevertheless a number of consistent findings have emerged. Firstly, on measures of behavioural adjustment, children with clefts are rated by themselves, teachers and parents as having more problems than their peers, usually in the direction of more anxious, insecure and socially withdrawn behaviour (Richman, 1978; Richman & Harper, 1979; Simonds & Heimburger, 1978; Spriesterbach, 1973). These findings are supported by more recent evidence (Speltz, Morton, Goodell & Clarren, 1993), which showed that children with craniofacial anomalies (primarily with repaired clefts) were two to three times more likely than peers without clefts to receive parent and teacher reports of behaviour problems above the 98th

percentile at school entry, a factor that is commonly associated with clinically significant disturbance. Furthermore, children and adolescents with clefts have shown poorer self-perception (Broder, Smith & Strauss, 1994; Kapp, 1979; Kapp-Simon, Simon & Kristovich, 1992), and school achievement (Richman, Eliason & Lindgren, 1988) when compared with same-age peers without clefts. The underachievement of children with clefts does not appear to be associated with lower general intelligence (McWilliams & Matthews, 1979; Tobiason, 1984) but rather with more specific language deficits, and also possibly with lower expectations of teachers, parents and children themselves (Richman & Eliason, 1982).

Of particular interest is the relationship between severity of disfigurement and the degree of psychosocial disturbance. One might expect this relationship to be linear, with more severe impairments associated with a higher levels of disturbance. However research has shown that this does not appear to be the case (e.g. Harper, Richman, & Snider, 1980; Lansdown, Lloyd, & Hunter, 1991), and that a mild impairment may be more of a psychological burden than those impairments that are more severe. This observed lack of linearity has been construed in terms of predictability (Lansdown *et al.*, 1991). If children with more severe disfigurements can confidently predict a negative response from most people that they meet, and therefore know that they will be stared at or teased, they can learn to cope with this consistent reality. However, children with milder abnormalities may be less able to predict other people's responses, and this may lead to raised anxiety.

The possible persistence of psychosocial difficulties associated with clefts into adulthood, has received minimal attention in the literature. However, the few existing studies have shown that young adults with repaired clefts tend to marry less often, report less marital and job satisfaction, participate less often in social activities, and report fewer, positive self-attributions than do individuals without clefts (Bjornsson & Agustsdottir, 1987; Heller, Tidmarsh & Pless, 1981; Peter & Chinsky, 1974).

Developmental Precursors of Psychosocial Difficulties in Children with Cleft Lip and Palate

Given that the incidence of cleft lip and / or palate represents a significant number of births each year, it is remarkable how little is known about the origins of psychosocial

problems experienced by children with cleft lip and palate. Even more surprising is the limited understanding that exists within the psychological literature of reactions to the birth and the psychosocial impact of the anomaly on the parents, the child and society. Speltz and his colleagues point to the absence of psychological investigation into the experience of children with cleft under the age of five, despite the intensity of the medical treatment procedures and caregiving difficulties during this period (Speltz, Greenberg, Endriga & Galbreath, 1994). Furthermore, there are no longitudinal studies charting the early course of the parent-child relationship and its effects on the child's later development. Much of what is currently known has been gleaned from retrospective studies which have explored mothers' and children's memories of earlier events or impressions of their relationship as perceived during the child's school years.

The dearth of studies in this area stands in contrast to research on infants and children with other types of chronic medical conditions (e.g., cerebral palsy or Downs syndrome), in which there has been much more emphasis on prospective research that begins in infancy and follows from a theory of social and emotional development (Cicchetti & Schneider-Rosen, 1984; Crnic *et al.*, 1986). Taking a developmental-organisational perspective, one would assume that significant emotional or behavioural disturbances seen in adolescence or in adult life are associated with difficulties that have emerged in infancy or early childhood (Sroufe & Rutter, 1984). It is therefore important to examine the specific pathways and mechanisms by which earlier events may have led to later disturbance, in order to attain a clearer understanding of the development of psychosocial problems related to cleft lip and palate. Two theoretical concepts relevant to this discussion will thus be considered here: a social - psychological theory of disfigurement, and attachment theory.

A Social - Psychological Theory of Disfigurement

> *"What is beautiful is good".*
>
> (Sappho, fragment 101, Dion *et al.*, 1972).

This phrase has been used by Dion and her colleagues (Dion, Berscheid & Walster, 1972) to exemplify findings that have consistently emerged from research, which indicate that an individual's physical attractiveness is an important social cue used by adults as a basis for social evaluation. Individuals considered physically attractive are

more likely to be judged intelligent, socially desirable, and successful (Dion *et al.*, 1972). These findings have been replicated in work with children, whereby unattractive children were chosen less often as desired playmates from a range of photographs, and were more frequently described as exhibiting anti-social behaviour (Dion & Berscheid, 1974).

The results from this research appear to have been borne out in parental and peer responses. Facial unattractiveness in non-dysmorphic children has been consistently associated with negative or avoidant peer reactions (Adams, 1980). Furthermore, in a normative sample, mothers of less attractive newborns were less affectionate, playful and attentive to their infants, than were mothers of more facially attractive infants (Langlois, Ritter, Casey & Sawin, 1995). This has led Tobiason (1984) to propose a theory of physical attractiveness as a potential framework for conceptualising the development psychosocial difficulties in children with cleft lip and palate. He hypothesised that in the case of children with cleft, the child's disfigurement is a primary causal variable, which elicits negative expectations from others, and thus leads to rejection or avoidant behaviour from parents or peers. Tobiason (1984) suggests that ultimately this may negatively affect the child's social skills and achievement at school.

In order to gain a clearer understanding of the developmental precursors to psychosocial difficulties experienced by children with cleft lip and palate, it may be helpful to explore the impact of the visible anomaly on the early relationship between the parent and child with cleft more closely. One developmental theory that may be of particular relevance here, pertains to the formation of the attachment relationship between parent and child (Bowlby, 1973; Ainsworth, Blehar, Waters & Wall, 1978).

Disfigurement and the Role of Attachment

Attachment Theory

Bowlby (1969; 1973) drew on analytical theory and ethological observation methods to study maternal behaviour in the development and maintenance of mother-baby attachment. He conceptualised attachment as a behavioural system in which the infant is biologically predisposed to emit 'social signals' such as crying, vocalising, smiling and other facial and motor movements. These behaviours promote increased proximity to

the mother or other attachment figures, resulting in comfort and protection to the infant during dangerous or distressing situations.

It is therefore believed that the formation of attachment begins very early in life (e.g., when the infant first directs signals toward a discriminated parent figure). However, behavioural indicators of attachment are most readily observed when the older infant becomes ambulatory, actively approaching the attachment figure when distressed, resisting separations from her in unfamiliar circumstances, and using her as a 'secure base' from which to explore. Ainsworth *et al.* (1978), developed a standardised assessment of attachment called the 'Strange Situation' in which trained observers code the infant's responses to brief, but stressful separations from the mother, and reunions with both the mother and an unfamiliar female adult. Three types of attachment behaviour have been reliably identified using this means of assessment, each representing an organised strategy for relating to a primary caregiver:

* a secure pattern in which the infant displays moderate separation distress followed by clear approach to the mother during reunion and rapid easing of distress;

* an insecure / avoidant pattern in which the infant shows minimal separation distress (or none at all), followed by ignoring or avoidance of the mother during reunion;

* an insecure / ambivalent pattern characterised by high separation distress followed by contact-seeking and contact-resisting behaviours upon reunion and the absence of return to exploration and play in the mother's presence.

Main & Solomon (1986; 1990) have subsequently identified a fourth category within this group of infants, namely a disorganised / disorientated attachment pattern. This is characterised by contradictory, disorganised behaviour upon reunion (for example, approaching the parent whilst keeping the head averted or seeking refuge under the parent's chair), reflecting the lack of a clear strategy, in addition to evidence of fear or confusion with respect to the caregiver.

Similar patterns of reunion behaviour have been reliably distinguished in toddlers and preschool children, as reflected in both verbal and non-verbal behaviour and expressions of emotion (Cassidy & Marvin, 1988). Further research has identified that the majority of infants and preschool children in non-clinical samples (approximately 70%) are

classified as securely attached, whereas the attachment behaviour of approximately 20 per cent is classified as avoidant, and 10 per cent as ambivalent (van Ijzendoorn & Kroonenberg, 1988).

Bowlby (1973) proposed that infants internalise their attachment patterns such that they form internal working models of their own self worth and of their expectations of care and support from others. This is thought to be of critical importance to later development. The internal working model is conceptualised as a cognitive representation or schema with associated emotional responses. Its primary function is to encode interactions in such a way that will facilitate an ability to predict how significant others will behave and how the self might feel, think and behave in response. It is with the aid of the internal working model that children will predict the likely behaviour of the attachment figure and plan their response. This model thus serves as a 'guide' for the child's subsequent socialisation and a 'filter' for new information about people and relationships (Sroufe & Fleeson, 1986).

Consequently, the child's internal working model reflects the quality and responsiveness of the early caregiving environment and the child's accumulation of experiences in attachment-relevant situations. If the attachment figure has acknowledged the infant's needs for comfort and protection while simultaneously respecting the infant's need for independent exploration of the environment, the child is likely to develop an internal working model of the self as valued and self-reliant. Conversely, if the infant's bids for comfort or independent exploration have frequently been rejected, the child is more likely to develop an internal model of the self as unworthy and incompetent.

Research has demonstrated the tendency for infant attachment patterns to remain stable, with strong continuities found within childhood years (Grossmann & Grossmann, 1991; Main, 1991). However, while resistant to change, internal working models are open to modification over the course of development. The intergenerational transmission of attachment patterns has also been identified by several studies that have provided evidence for associations between parental attachment status and infant attachment (Fonagy, Steele & Steele, 1991; Steele, Steele & Fonagy, 1996).

Results from a number of longitudinal studies that have evaluated the psychosocial status and attachment behaviour of children, suggest that infant attachment status is one of the best single indicators of future psychosocial development, at least for high-risk

infants (Sroufe, Egland & Kreutzer, 1990; Urban, Carlson, Egland & Sroufe, 1991). Attachment insecurity in infancy has been associated with later problems in self-perception and peer relationships (LaFreniere & Sroufe, 1985), and behavioural adjustment (Erickson, Sroufe & Egeland, 1985). Given that certain childhood psychological outcomes associated with ineffective infancy attachments are similar to those problems shown by some children with cleft lip and palate, it is possible that the quality of attachment relations in infants with craniofacial anomalies may to some extent determine the nature of their psychological adjustment in later life. In order to examine this hypothesis, it is necessary to consider the limited empirical evidence available.

Attachment Status of Infants with Cleft Lip and Palate

Almost three decades ago, Clifford (1969) observed that little was known about the early experiences of infants with craniofacial anomalies and their parents, and for the most part, this observation remains true today. Literature on aspects of the parent-infant relationship or family environment that are relevant to attachment theory is sparse. However, three studies have specifically examined the quality of attachment in the craniofacial population. Wasserman, Lennon, Allen & Shilansky (1987) observed 12 month old infants with various congenital anomalies in a variant of the Strange Situation (Ainsworth *et al.*, 1978). Very few infants in this sample had clefts, the majority having orthopaedic impairments. No significant differences were found between this and a control group in terms of attachment. Similarly, Speltz *et al.* (1997) recently compared attachment classifications of 12-month-old infants with orofacial clefts, with those of infants without clefts, using the Strange Situation. Again, no significant differences in attachment status were found.

In a third study, Koomen & Hoeksma (1993) investigated the impact of brief early hospitalisation on the infant's attachment relationship with their mother, in a group of infants who were hospitalised for palatal repair at either 9.5 months of age or 12.5 months of age. A group of children without clefts who were not hospitalised served as controls. Findings showed that hospitalisation has an increasing effect on avoidant behaviours such as lower levels of contact maintenance and resistance, towards the mother after induced stress. Nevertheless, in spite of these heightened levels of avoidant behaviour, hospitalised infants were not found to be at risk of establishing insecure-avoidant attachments to their mothers.

The findings from these studies therefore suggest that, contrary to expectations, children with cleft lip and palate are not necessarily at greater risk for insecure attachment. However, in order to understand the developmental precursors to psychosocial difficulties observed in this group of children, it may be important to identify potential early predictors of attachment.

Early Predictors of Attachment in Infants with Cleft Lip and Palate

There is evidence which suggests the possibility of an elevated risk of attachment problems in children with craniofacial amonalies, including cleft lip and palate, in two particular areas:

* early parent-infant interaction;

* the parent-child relationship during preschool years.

Early Mother-Infant Interactions

Four studies have focused specifically on the interactions between mother and 3- to 6-month old infants with clefts during face to face interactive play. Field & Vega-Lahr (1984) found mothers of infants with clefts to be less responsive and less socially interactive than mothers of 'typical' infants, as indicated by lower ratings of game playing, imitation, contingent responsiveness (i.e., responding when the baby initiated a new behaviour), and facial expressiveness. This study also found that the infants with clefts engaged in less smiling, vocalising and looking at their mothers.

Evidence consistent with these findings comes from second study (Barden, Ford, Jensen, Rogers-Sayler & Sayler, 1989), which found that infants with wide-ranging craniofacial anomalies (i.e. unilateral and bilateral cleft, microfacial microsomia, Crouzon syndrome, and Apert syndrome), were less likely to smile/laugh, vocalise, or show head orientations toward their mothers than non-impaired infants. Despite the small sample size and extreme diagnostic heterogeneity, mothers of infants in the craniofacial group were consistently rated as less active and responsive towards their infants. Similarly,

Wasserman, Allen & Solomon (1985) observed the interactions of mothers of infants with a range of disabilities (including cleft lip and palate), and found them to be significantly less responsive and socially interactive than dyads containing healthy or premature infants. In a fourth study (Endriga, Speltz & Wilson, 1992), mothers of infants with cleft lip and / or palate were observed to smile less often when their baby smiled, when compared with mothers in a control group. This finding suggests that for mothers of infants with clefts, there is less contingency in their response to the infant's display of positive emotion.

A further study examined maternal responsiveness to infants with unrepaired clefts in a feeding situation (Speltz, Goodell, Endriga & Clarren, 1994). Mothers of infants with cleft lip and palate were found to respond less sensitively to their infant during feeding, than mothers of infants with isolated cleft lip or mothers in the control group. This observed difference between mothers of infants with cleft lip and infants with cleft lip and palate, was found to be related not to facial appearance, but to more difficult feeding methods required by infants with cleft of the palate and the lip. Findings also revealed that in comparison to typically developing infants, infants with clefts showed less frequent smiling, and were less clear in their communication during feeding. Lack of clarity was particularly evident in the signalling of their readiness to begin eating and showing the relaxation commonly observed in infants just after the beginning of feeding.

The quality of the early parent-child relationship as measured in teaching interaction also provides information about maternal sensitivity and child responsiveness. Research demonstrates that teaching interaction predicts later child psychosocial outcomes (Barnard, Hammond, Booth, Bee, Mitchell & Spieker, 1989). In an investigation of the cognitive and social development of infants born with craniofacial anomalies, Endriga, Speltz & Mason (1995) observed mother-infant teaching interaction in the second year of life and found differences between cleft and non-cleft groups. Infants with a cleft lip and palate were less clear in their communicative cues than non-impaired infants. Furthermore, mothers of cleft lip and palate infants engaged in less cognitive, growth-fostering activity than did mothers of non-impaired infants.

The results from these investigations suggest that mothers of infants with cleft lip and palate may be less socially responsive to their infants during the first two years of life, than mothers of non-impaired infants, and that infants with clefts may less clear and responsive in their communications.

Preschool Attachment

The relationship between the parent and the preschool child with orofacial cleft is also of relevance to potential problems in the development of attachment. It has been hypothesised that the elevated risk for speech and language delay found in children with cleft lip and palate may lead to a number of difficulties during this postinfancy phase of attachment (Speltz *et al.*, 1994). During this period, attachment relationships are characterised by increasing child autonomy and self-reliance, and gradually become more organised in relation to internal models. This process is facilitated by developmental changes experienced by typical toddlers and preschool children. In particular, the ability to communicate distally with the caregiver through verbal interaction is thought to enable most preschool aged-children to become less dependent upon the caregiver, thereby facilitating the transition from overt to internalised regulation of attachment. Yet for children with cleft lip and palate, this transition may be delayed by speech and hearing problems that complicate parent-child communications (McWilliams *et al.*, 1990). Children with cleft lip and palate have also shown deficits in their use of verbal mediation or 'internal language' during problem solving tasks (Eliason & Richman, 1990). Such deficits may obstruct the child's transition to a more mature form of attachment by limiting internal language as a means of emotional regulation during parental absences.

Furthermore, efforts to become self-reliant during preschool years may be discouraged in the case of the child with cleft lip and palate, by a parenting style that has been observed as over-controlling or over-protective (Tobiasen & Hiebert, 1984). Parental over-protection and intrusiveness is thought to be a result of the emotional upheaval surrounding the birth and early care of the infant with cleft (Speltz *et al.*, 1994), and may create a relationship that impedes the child's efforts to 'individuate' and to acquire age-appropriate skills, by undermining the child's interests in peer relationships. However, as yet, neither of these hypotheses regarding the significance of preschool speech and language delay and family interactional patterns, on attachment relationships, has been investigated in children with clefts.

Although, studies have demonstrated that children with craniofacial anomalies are not more likely to experience attachment difficulties than non-impaired children, parent-child interaction during infancy and preschool years, and in particular the lack of

maternal responsiveness indicated in the investigations reviewed here, certainly suggest an elevated risk of attachment problems for children with orofacial clefts. The issue of maternal responsiveness may possibly contribute to our understanding of the precursers of psychosocial difficulties experienced by these children, and therefore warrants further exploration.

The Antecedents of Maternal Responsiveness

As already discussed, maternal responsiveness is a crucial component in the development of attachment. Mothers rated as more sensitive and contingent in their interactions with the infant during the early months of life are more likely to have securely attached infants, than mothers who are not responsive (Ainsworth *et al.*, 1978). Research into mother-child interactions in the cleft population, has observed that mothers may be less responsive in their interactions with their infants, but has failed to clarify the antecedents to this lack of responsiveness

Speltz *et al.* (1994) suggest that the responsiveness of mothers is likely to be at least partially limited by the clarity of the infant's social behaviours, particularly with respect to the communication of positive emotion and feeding readiness. Another potential explanation might be found in the mother's aversion to the infant's appearance, as originally proposed by Tobiason (1984). However, disfigurement is but one of several adverse factors potentially affecting the infant and parent dyad. The infant with orofacial cleft and her parent are challenged by feeding problems, surgeries and frequent medical clinic visits. Hospitalisations that separate child from parent may also have an adverse impact. Previous longitudinal research has demonstrated that hospitalisation during the first six months of life increases the risk of difficulties in the mother-child relationship by the age of three years (Ludman, Lansdown & Spitz, 1992).

The potential impact of the family environment and social context on maternal responsiveness in the cleft population, can also not be underestimated. Within the developmental literature, maternal responsiveness throughout childhood is thought to be influenced by contextual variables including life stress and family adversity (Spieker & Booth, 1988), the parent's emotional health and stability (Radke-Yarrow, Cummings, Kuczynski & Chapman, 1985), and parental satisfaction with social support (Crnic, Greenberg & Slough, 1986). Evidence from investigations into attachment in normative populations, also reveal strong associations between attachment security in infants, and parents' reports of adequate social support (Crockenberg, 1981), positive marital

relations (Belsky & Isabella, 1988) and family cohesion and adaptability (Bretherton, Ridgeway & Cassidy, 1990).

Speltz and his colleagues (1990) examined relationships between children with craniofacial anomalies and their mothers during late infancy and preschool years. They concluded that parental reaction to the atypical appearance of the child may be more dependent on such variables as maternal psychological status and marital functioning, than upon the nature of the disfigurement itself. Consequently, responsiveness in mothers of infants with clefts, may be adversely affected by psychological and social factors such as high levels of marital conflict (Speltz, *et al.*, 1990), and a lack of available social support (Benson, *et al.*, 1991), which are in turn associated with the child's 'impaired' status.

An important criticism of this body of research is the tendency to include different, heterogeneous conditions with a congenital component related to the face and skull in one research group, thereby falsely assuming homogeneity. Whilst this approach does allow larger sample sizes, it also introduces confounding variables. The physical and psychosocial problems associated with cleft lip and palate may not be shared with other craniofacial anomalies. For example, a child with cleft lip and palate has a constellation of associated features such as hearing difficulties and speech problems, whereas Crouzon's syndrome can be associated with mental retardation. These conditions also differ in their treatment, and in the extent to which they can be corrected. Psychosocial impact may also vary across different craniofacial conditions. Speltz *et al.* (1990; 1993) found that mothers of children with cleft lip and palate reported less satisfaction with social support than did mothers of children with isolated cleft palate or sagittal synostosis (a malformation of the skull bones caused by early suture fusion). Also, as we have already seen, the extent to which self esteem is affected may vary, depending on the severity of the condition (e.g., Lansdowne *et al.*, 1991). Attachment research into the interaction of mothers and babies with facial disfigurement has also attracted criticism for its failure to consider the association between the observed behaviour of the dyad and the meaning of this for the mother (Bradbury, 1995). The effect of parental beliefs on mother-baby interaction has been examined elsewhere (Skinner, 1985; McGillicudy-Delisis, 1982), but not within the cleft population.

Nevertheless, the evidence explored here suggests an ecological, transactional model of parental adaptation to cleft lip and palate, that gives causal significance to both parental

and child risk factors (Belsky, 1984), as opposed to formulations that have given 'main effect' status to craniofacial appearance (e.g., Tobiason, 1984). Of particular relevance to this theory is a meta-analysis carried out by van Ijzendoorn and colleagues (1992), which examined the relative impact of child and maternal problems on attachment in a sample of mothers of children with a range of disabilities (including deafness, developmental delay, Downs syndrome, prematurity and other physical problems). Child disability was found to have little impact on attachment status, whereas the demonstrated effect of maternal problems such as mental illness or child maltreatment by mother, was to increase insecure attachment. Results indicated that when a child had a physical or mental disability, the mother was generally able to compensate for this potential handicap in the dyadic relationship. However, when the mother of a disabled child was suffering from a mental illness or was engaging in inadequate caregiving behaviour, the child was unable to compensate for the lack of maternal responsiveness, and was therefore more vulnerable to developing insecure attachment.

Maternal Adaptation to Cleft Lip and Palate

This notion has been explored by Cox & Lambrenos (1992), who emphasise the need for parents to compensate if their disabled child is less able to communicate needs, register parental communication, or reduce anxiety themselves. These specific disabilities influence the development of the early social and emotional interactions between parents and children, that lead towards the establishment of attachment relationships. Cox & Lambrenos (1992) suggest that maternal adaptation to a child's capacities may compensate for the child's disability at least during the first year of life. Cicchetti & Schneider-Rosen (1988) also draw attention to the wide range of compensatory adjustments that parents of children with disabilities adopt, in order to interact effectively with their child. They suggest that parents of children with disabilities need a higher level of skill to help them read their child's emotional cues, and to communicate effectively.

When applied to children with cleft lip and palate, these ideas suggest that the occurrence of cleft lip and palate alone is probably insufficient to significantly elevate the risk of attachment insecurity. It would appear that the caregiving environment, or more specifically, the mother's ability to adapt to her child's apparent difficulties in

communication, may compensate for limitations imposed by the cleft, such as lack of clarity in the child's communicative cues.

Parents differ in their capacity to adapt and adjust to their child's specific needs (Cicchetti & Schneider-Rosen, 1988). Most parents will compensate effectively, however, there may be a number of factors that impede the ability to compensate, including concurrent adversity, and negative feelings and perceptions that the parent may have, regarding their child. Thus, in some cases, parents may need to be taught compensatory skills, and so the identification of individual differences may be important.

Pianta & Marvin (1993a) suggest that the ability to adapt one's parenting style in order to make the necessary compensations is dependent on the parent's ability to resolve their grief for the loss of the 'healthy' expected child, following the diagnosis of their child's disability. They have developed a theoretical model based on attachment theory, as a framework for understanding the processes involved in parents' resolution of their reaction to receiving a diagnosis of their child's disability. This model assumes that for many parents, receiving a diagnosis that their child has a medical condition, such as cleft lip and palate, is experienced as a crisis which threatens or destabilises the parent's view of their child, themselves as parents, and parenthood. This is consistent with the predominant view in the literature that parents may experience a range of intense and distressing feelings, considered akin to a grieving response, when they first learn of their child's medical problem (e.g., Klaus & Kennell, 1984).

Resolution can be best understood as a process, whereby the parent integrates the information that pairs the child with the disability, and the accompanying emotions, into their representational systems of themselves as parents, of their child, and of their relationship with their child. In essence, the parent needs to integrate their working model of themselves and their child *before* diagnosis, with the corresponding representations, *after* diagnosis.

According to Pianta & Marvin (1993a), the cognitive task of resolution is to integrate this information about the child's disability, without distorting reality (i.e., minimising or denying child's true condition), or focusing too much on this information to the exclusion of other, present-day realities. The emotional work of resolution is to acknowledge the painful feelings associated with the diagnosis, and the ongoing

experience of parenting a child with a disability. Again, this involves not distorting feelings, an acknowledgement of emotional 'moving on' as opposed to emotional disorganisation, whereby there is a focus on the present reality, and a move away from feelings of crisis to coping. It is acknowledged that for parents of a child with a disability, the crisis information and accompanying feelings may periodically resurface as challenges to the equilibrium of the parent's representational model, and so the cognitive and emotional work of resolution is re-engaged. Thus, resolution is viewed as an ongoing process, and the extent of resolution will vary from time to time, depending on the child's and parent's circumstances, and their developmental history.

Marvin & Pianta (1993a) have developed a standardised and reliable assessment interview, in order to detect individual differences in resolution of loss, and facilitate an understanding of parents' vulnerability or resilience to the trauma of diagnosis. In a sample of mothers of children with cerebral palsy, the researchers found that status of parental resolution was a more powerful risk factor for insecure attachment, than the child's disability (Pianta & Marvin, 1993c). This is in keeping with the earlier findings of van Ijzendoorn *et al.*, (1992), and the work of Cox and Lambrenos (1992), regarding the relative impact of child and maternal problems on the development of attachment.

The findings from this research have led Pianta & Marvin (1993c) to conclude that failure on the part of parents to grieve or resolve the trauma of receiving the diagnosis, can interfere with sensitive caregiving during infancy and early childhood, leading to an increased risk for attachment difficulties.

A consideration of the resolution process of parents of children with cleft lip and palate, would therefore seem critical to our understanding of parental adaptation to a child with cleft, in addition to the impact of adaptation on the parent-child dyad, and on later outcome for the child. The identification of factors which may facilitate or impede parental resolution to their child's cleft lip and palate may also be important. There may be a number of developmental pathways that lead to resolution or non-resolution in this group of parents. Notably, time was found to be unrelated to resolution (Pianta & Marvin, 1993c), however, many other factors, such as the type and degree of practical and emotional support received by the parent may possibly relate to parental resolution and adjustment.

A further, potentially salient variable in this process that warrants consideration here, relates to the context and timing of the diagnosis. With the development and widespread use of prenatal diagnostic techniques such as ultrasound scan, many mothers are now experiencing the diagnosis of their baby's cleft during pregnancy. Little is known of the impact of this new context, on the process of adjustment and resolution experienced by parents of children with cleft lip and palate. In order to explore this issue, it is first necessary to examine these new developments in antenatal care in greater detail.

Prenatal Screening of Congenital Anomalies

During the last decade, one of the fastest expanding areas in medicine has been the development of techniques for fetal diagnosis for an ever growing list of genetic diseases and congenital malformations. Prenatal screening for fetal abnormalities has become an integral part of antenatal care, and together with neonatal intensive care, testing and screening procedures such as amniocentesis, chorionic villus sampling, and ultrasound scanning have had a more substantial impact on the outcome of a pregnancy than any other obstetric practice (Chard & Macintosh, 1992). While the use of these techniques is still a long way from detecting all serious fetal abnormalities, the birth incidence of a number of conditions, such as neural tube defects, anencephaly and Down's syndrome, has been significantly reduced, in part by a process involving screening, diagnosis and termination (e.g., Limb & Holmes, 1994).

The implicit aim of most fetal testing is thus to improve the outcome of pregnancy. Essentially prenatal screening and diagnostic testing have three objectives:

* to inform and prepare parents for the birth of an affected infant;
* to allow in utero treatment or delivery at a specialist centre;
* to allow termination of an affected fetus.

The advantages of prenatal screening and diagnostic testing outlined in the literature include the reduction of uncertainty as the accuracy of prognosis is improved, the opportunity to avoid having a birth-disabled child through termination, or alternatively to initiate a treatment plan when a problem is found, and otherwise to allay anxiety where no abnormality is evident. Information obtained from a positive test may be of value even when no direct medical action is taken. Farrant (1985) reports that the opportunity to prepare for the birth of a child with an abnormality, was a benefit of prenatal diagnosis frequently cited by mothers of children with disabilities.

Routine ultrasound scanning has become a widely used tool, and is very different from other methods of fetal investigation. As a non-invasive technique, it carries no risk to the fetus, and long waits for results tend to be avoided. In addition to revealing gross structural abnormalities, the scan serves to confirm gestational age, to localise the placenta and to detect multiple pregnancies. Routine anomaly scans are offered by most

antenatal units at 18 to 20 weeks of gestation. However, there are striking differences in the detection rates of different units (Saari-Kemppainen, Karajalainen, Ylostalo, Heinonen, 1990). This has been attributed to differences in experience of staff and in the equipment available.

The relationship of a mother with her child is presumed to begin during pregnancy with both the realities and the mother's fantasies of the developing fetus. This relationship, described as the mother's attachment relationship with her fetus, has been considered both a developmental task of pregnancy and an important prerequisite to successful maternal-infant adaptation (Leifer, 1980; Raphael-Leff, 1991). Clinicians have suggested that ultrasound examinations during pregnancy are related to increased feelings of attachment to the fetus (Heidrich & Cranley, 1989). This observation is supported by descriptive studies in which pregnant women have described the effect of ultrasound as positive and as increasing their feelings of attachment to the fetus (e.g., Fletcher & Evans, 1983; Rothman, 1986). However, despite anecdotal evidence in support of the positive effect of ultrasound on attachment, findings from prospective research which has examined later parent-infant interaction, have not borne out these clinical impressions (Grace, 1984; Sparling, Seeds & Farran, 1988).

The widespread use of sophisticated tests for fetal well-being attests to their popularity, however, this does not necessarily verify their value. It is tempting to believe that early detection of abnormality or disease is worthwhile, and an end in itself. Yet, Mohide & Enkin (1992) warn against identification of 'trivial or untreatable' conditions that may cause anxiety with no useful result. They maintain that it is only worthwhile to screen for disorders that lend themselves to effective intervention.

Prenatal Diagnosis of Cleft Lip and Palate

In recent years, craniofacial abnormalities such as cleft lip or palate can now be detected by ultrasound at approximately twenty weeks gestation (Anderson, Boswell & Duff, 1995). Despite the frequency of cleft lip and palate amongst newborns, relatively few studies have reported prenatal detection. In many cases, non-syndromic or isolated clefts go undetected. This probably reflects both the difficulty in diagnosis, and the fact that sonographic evaluation of the face is not required at routine ultrasonography in low risk obstetric cases (Nyberg, Sickler, Hegge *et al.*, 1995). Furthermore, most isolated cleft

anomalies affect patients with no previous history of fetal malformations, who as a result, are considered low risk (Bronshtein, Blumenfeld & Blumenfeld, 1996). A notable problem of antenatal diagnosis of craniofacial anomaly by ultrasound, is the accuracy of prognosis. Although cleft lip can potentially be detected or ruled out, this is not the case for cleft palate, which generally cannot be picked up at scan. The exact severity of the cleft lip and the size of the fissure are also difficult to assess accurately on scan (Bronshtein *et al.*, 1996).

The few published studies of sonographic detection of craniofacial anomalies have reported wide variations in rates of detection. Anderson *et al.* (1995) reported rates of sonographic detection of fetal abnormality at 16 to 20 weeks gestation in 7880 pregnancies. The prenatal diagnoses obtained at scan were compared with pregnancy outcomes established by pregnancy records and pathologic results. The success rate for sonographic detection of craniofacial abnormality was 25 per cent. This is particularly low when compared with detection rates of 92, and 78 per cent for central nervous and pulmonary anomalies respectively, and is reflected in the findings of other studies (e.g. Luck, 1992; Stoll, Dott, Alembik & Roth, 1995).

However, significantly higher rates of detection have been reported elsewhere. For example, Bronshtein, Blumenfeld, Kohn & Blumenfeld (1994) detected 92 per cent of the clefts in their retrospective study, in which transvaginal sonography was used, rather than transabdominal scan. This effective screening method is, however, less widely used in the U.K. High detection rates using transabdominal scans, have also been reported by a small number of studies. Pilu and colleagues report a false negative rate of one per cent (Pilu, Reece, Romero, Bovicelli & Hobbins, 1986). Additionally, Nyberg *et al.*, (1995) reported a misdiagnosis rate of one per cent, whereby a unilateral cleft was diagnosed as bilateral. There appears to be no discussion of false positive rates in the literature.

The variability observed in prenatal detection rates of cleft lip and palate could be due to possible variations in the quality of sonographic resolution, and in operator skill. Nevertheless, findings indicate that ultrasound could potentially be an accurate and reliable tool for the prenatal diagnosis of craniofacial malformations.

The impact of inaccuracy of diagnosis of cleft lip and palate by ultrasound scan, has received limited attention in the literature. Erdmann & Waterhouse (1992) describe one

case of a false negative result of cleft, which had an adverse effect on the parents. This involved a couple who had a child with bilateral cleft lip and palate, and who intended to terminate the second pregnancy, should this fetus also have cleft. Ultrasound scans were reported as normal, and so the parents elected to continue with the pregnancy. When the cleft was apparent at the birth, the parents were devastated, and were described as rejecting the baby until neonatal surgical repair was carried out. Research into the effect of false positive results of other fetal anomalies on parents, has found that negative results following initially positive results do not necessarily provide complete reassurance that all is well, and that for some women the elevated level anxiety triggered by a false positive result, can extend into the post-partum period (e.g., Marteau, Cook, Kidd, Michie, Johnston, Slack & Shaw, 1992).

More recently, termination following diagnosis of cleft, has been examined by one study. Bronshtein *et al.* (1996) report that during a 10 year period, 15 cases of cleft lip (with / without cleft palate) were diagnosed at 14 to 16 weeks gestation, in 2400 transvaginal scans. Notably, 14 of these cases were terminated. The majority were in low risk patients, with no previous medical history of fetal anomalies. The researchers also report that among a second group of 17 families who already had a child with cleft lip, all of the parents stated that they would choose termination in a subsequent gestation if cleft lip was detected. This is an isolated study, and therefore it is not possible to assess the representativeness of the findings.

The Social Context of Prenatal Screening

A notable gap in the literature on prenatal screening, is the failure to consider women's experiences and attitudes in the wider social context. That context shapes the woman's expectations of the process and her reactions to it. An important element is a woman's own social and family situation, but equally important is her perception of society's attitudes and other people's perceptions of her situation (Green, Statham & Snowden, 1992). Society currently takes a favourable view of screening. Implicit in this view is the notion that the 'perfect baby' is an attainable goal, and its corollary that less-than-perfect babies are not wanted. Green *et al.* (1992) argue that as a means of detecting fetal anomalies, prenatal screening may foster a general attitude that the birth of less-than-perfect babies is preventable.

Women's Experience of Screening and Diagnosis of Fetal Abnormality

Women's experience of prenatal diagnosis of craniofacial anomalies has been completely neglected in the literature. However, research into women's experience of routine prenatal screening and diagnosis of other conditions, has clearly demonstrated that receipt of an abnormal screening result, is associated with high levels of anxiety and distress. This is reflected in mothers' increased worry about the baby's health and in a more negative attitude towards the pregnancy and the baby (e.g., Marteau, Cook, Kidd, Michie, Johnston, Slack & Shaw, 1992).

Richards (1989) has also suggested that the detection, usually by chance, of conditions which are less serious and are generally thought not to lead to severe impairment, may have a negative impact on the parent. He gives the example of a Swedish screening programme for alpha-antitrypsin deficiency, which was discontinued after it was found that the identification of the children led to considerable psychological reactions from some parents and prolonged disturbance of the parent-child relationship (Thelin *et al.*, 1985). Prenatal diagnosis therefore clearly has the potential to increase anxiety and distress, depending on the woman's initial risk status, her understanding of the tests being used, and above all, the quality of communication that accompanies the procedure.

A woman's priorities may be quite different from those of the providers of screening. In one of the earliest studies of prenatal testing, Farrant (1985) pointed to an important distinction between the standpoint of obstetricians who plan and organise prenatal screening, and the mothers who receive it. While mothers tend to seek reassurance that their baby is healthy, obstetricians put the emphasis on the detection of abnormality. However, these obstetric attitudes contradict much of the rhetoric that surrounds prenatal screening, which emphasises the reassurance it can offer women and the choices it provides for parents (Marteau, Plenicar & Kidd, 1993).

In particular, ultrasound scanning may not be viewed by women as a screening or diagnostic test, but rather as a chance to see the baby. Because the detection of fetal abnormality is rarely given as the principle reason for its use, women tend not to feel threatened by it. The reassurance sought by the majority of women from ultrasound scan is the most common outcome. However, for a minority of women, not being aware of

the true purpose of the scan, can leave them completely unprepared for the news that their child has an abnormality.

The manner in which results are communicated is also important for women, and yet it is well documented that problems often exist between professionals and parents of children with disabilities, when the diagnosis is given. Communication problems at this time have been related to difficulties for parents in accepting their child's diagnosis (Edelstein & Strydom, 1981), and in their early treatment of the child (Springer & Steele, 1980). Several themes have emerged from research into parents' experience of the diagnosis of their child's disability (Quine & Pahl, 1986; Quine & Rutter, 1994). Parents appear to favour an early acknowledgement of the problems, even when doctors may be unsure of the exact nature of the child's impairment. Delay in giving the diagnosis has been found to lead to greater parent dissatisfaction. Parents also value a sympathetic and caring approach by doctors and other medical staff. Furthermore, parents want to be given full information about the child's condition.

These needs appear reasonable and unproblematic, and yet it seems they are seldom met. Many studies document high levels of dissatisfaction among parents with the way they were first informed of their child's condition (e.g., Quine & Rutter, 1994). Evidence from these studies indicates that parents often face denial, delay and evasion before the diagnosis is made. Research comparing doctors' views with those of parents, on presenting diagnoses, is consistent with these findings (Nursey, Rohde & Farmer, 1991). It seems that parents are more in favour of being given information about their child's disability at an early stage, than are doctors.

Many of these problems stem from a lack of clear guidelines on what nursing staff should do in the event of the birth of a child with a disability. In the case of ultrasound, Hyde (1986) reports that the reticence of the scan operator, and the poor quality of feedback given, were major negative factors for many women. A further cause of difficulty was the question of authorisation to give the diagnosis. Often, the scan operator was a radiographer, who was working under restrictions of what could be said to mothers. Thus, when an abnormality was detected, the operator had to leave the room and find an obstetrician or radiologist. This was experienced as extremely anxiety-provoking and distressing by the mother. It also appears from the results of another study, that scan operators have difficulty in accurately judging women's anxiety levels or

the amount of information they request during the scan (Barton, Harris, Weinman, Allan & Crawford, 1987).

It has also been argued that the raised anxiety levels associated with prenatal screening, in part reflect the failure of the screening service to provide adequately for the intensive counselling needs created by an abnormal result from a routine screening test (Farrant, 1985). Evidence from screening programmes suggests that reducing the immediate and longer-term distress associated with receipt of positive results on a routine screening test, may be achieved by appropriate preparation for undergoing the test, in addition to the provision of information, counselling, and support post-test (Marteau, 1990).

Clearly, the technology for antenatal diagnosis has grown faster than the ability of the health-care professional to explain the technology, and communicate test results to prospective parents. While this should not be a constraint on the development of antenatal testing, it should nevertheless remain a matter of concern. Of particular importance, is the negative impact of the diagnosis of an abnormality on the mother's psychological health, and on her perception of her pregnancy and child. Given research findings which suggest a link between the feelings and attitudes that mothers have about their pregnancy and the unborn child, and the early postnatal mother-infant relationship (e.g., Condon & Dunn, 1988), a greater understanding is clearly needed, of the long-term implications of prenatal diagnosis of abnormality for women and their children. These issues are only beginning to be addressed.

The Present Study

It is widely accepted that the birth of a child with cleft lip and palate can have a significant impact on the parent. Difficulties in the parent-infant relationship have been found, and psychosocial problems in children with cleft are also frequently observed. However, the developmental precursors to these difficulties are poorly understood. Difficulties experienced by parents in adjusting to their child's disability, and in resolving their reaction to the diagnosis, may possibly have a negative impact on the way in which they respond to their child's needs, and communicate with their child. This in turn may have implications for the child's attachment relationships, and later psychosocial outcome.

Despite prolific technological research into prenatal screening for fetal abnormality, relatively little is known of the impact of prenatal diagnosis of abnormality, on parental adjustment to the child. The detection of cleft lip and palate during pregnancy may provide the parent with an opportunity to prepare for the birth of the child, and thus facilitate adjustment and later coping. Alternatively, it may be argued that an awareness of the child's disability during pregnancy, and inadequate support from antenatal services, may have a negative impact on the parent, reflected in heightened psychological distress and negative perceptions of the pregnancy and the child. These difficulties may lead to poorer adjustment on the part of the parent.

This study proposes to examine the impact of detecting cleft lip or palate by ultrasound during pregnancy on the parent's adjustment to the birth of the child with cleft lip and palate. Previous research has emphasised the importance of social support and marital adjustment in successful adaptation of families of children with disabilities (e.g., Sloper & Turner, 1991). Given the tendency for parents of children with cleft to experience inadequate support systems and greater marital conflict (Benson *et al.*, 1991; Benson & Gross, 1989), this study also aims to examine the role of social support and the quality of the parental relationship on parental adaptation to the birth of a child with cleft lip and palate.

Hypotheses

The experience of mothers who received a prenatal diagnosis of cleft lip and palate, will be compared with that of mothers for whom the diagnosis was given at birth. It is hypothesised that:

1. Prenatal detection of the abnormality will be associated with successful parental adjustment, and resolution of trauma associated with the birth.

2. Prenatal detection of cleft lip and palate will be associated with lower levels of current psychological distress.

3. Prenatal diagnosis of cleft lip and palate will be related to higher levels of social support, and more favourable marital adjustment.

4. Mothers' experience of social support, and the quality of their relationship with their partner will be related to adaptation to the birth of a child with cleft. Higher levels of social support, and more favourable marital adjustment will be associated with successful parental adjustment, and resolution of the trauma experienced at diagnosis, in addition to lower levels of psychological distress.

Parents' experience of the pregnancy and birth, and the impact of perinatal stressors on parental adaptation to the child, will be explored. Similarly, parents' experience of support and information following diagnosis will be examined. Finally, this study will investigate mothers' attitudes and beliefs about prenatal diagnosis of cleft lip and palate.

METHOD

Overview

This study aimed to investigate factors associated with parental adjustment to the birth of a child with cleft lip and palate. In particular, the impact of prenatal diagnosis of craniofacial anomaly on parental adjustment to the birth of a child with cleft lip and palate was examined. Mothers with a prenatal diagnosis of their baby's cleft lip and palate were therefore compared with mothers who had not received a prenatal diagnosis of their child's cleft, on a number of measures that related to adjustment. These included resolution of crisis or trauma associated with the diagnosis, specific indicators relating to emotional, cognitive and behavioural aspects of adaptation to congenital disfigurement, and psychological well-being. The role of social support and quality of the parental relationship in adjustment to the birth of a child with cleft lip and palate was also explored. The methodology used in order to accomplish these aims, is described here.

Participants

A total of 44 mothers with a child who had a cleft lip and palate participated in the study. Twenty of the mothers had received a diagnosis of cleft lip and palate at prenatal screening (Prenatal group), whilst the remaining 24 were mothers for whom the cleft lip and palate was not detected prenatally (No-Prenatal group).

Participants were recruited from four craniofacial units in the North and South Thames regions, where their child had undergone corrective surgery, namely at Chelsea and Westminster Hospital, Mount Vernon Hospital, Great Ormond Street Hospital for Children NHS Trust, and Queen Mary's University Hospital.

Participants in both the Prenatal and No-Prenatal groups met the following inclusion criteria:

1. They were mothers of children who had unilateral or bilateral cleft lip and palate. Parents of children with isolated cleft lip or isolated cleft palate were not asked to

participate, due to the potential differences that these two groups may experience in terms of operations, hospitalisation and feeding difficulties. Cleft lip and palate can also accompany other congenital anomalies or syndromes, and those mothers of children with any known additional medical or physical difficulties were not included in the study.

2. They were the primary caretaker of the identified child.

3. At the time of the study, their child was aged between 11 and 36 months.

4. Surgery to repair the lip and the palate had been completed at least 1 month prior to the study.

5. They were able to be interviewed in English.

Hereafter, the use of the term 'parent' refers to mother.

Ethical Approval

Ethical approval was applied for and obtained from Riverside Research Ethics Committee, in addition to the Research Ethics Committees at Mount Vernon and Watford Hospitals NHS Trust, Great Ormond Street Hospital for Children NHS Trust, and Richmond, Twickenham and Roehamption Healthcare NHS Trust (Appendix I).

Procedure

Recruitment

For each of the craniofacial units, the procedure for recruiting participants was the same. The names, addresses and dates of birth of every child who met the study criteria were accessed from the medical records held at each unit. These details were held either on a computer database or in medical record files, depending on how information was stored by the particular unit. In the case of the latter, the researcher searched through the medical files held in the department in order to collect details of all parents who met the study criteria. Information on the type of cleft and dates of operations, was also accessed at this stage. Additionally, status regarding prenatal diagnosis of the cleft was available

from two of the units. All eligible mothers were then contacted directly by the researcher.

The Initial Contact

Parents of children who met the inclusion criteria were initially approached by a letter, which briefly introduced the study, described the aims of the research, and what was involved in participation (Appendix II). Emphasis was placed on the voluntary nature of participation in the study, and the confidentiality of the information elicited. The letter was sent from the craniofacial unit where the child was being treated. Parents were asked in this letter to return a form to the researcher using a prepaid envelope provided, indicating whether or not they wished to be contacted about participation in the study (Appendix II).

On receipt of this form, the researcher then telephoned parents who indicated that they wished to be contacted, and described the study in greater detail. During this telephone call, the researcher answered any questions posed by the parent and explained their potential involvement in the study. At this point, if the parent was agreeable, the researcher arranged a time for the interview to take place, that was most convenient for the parent during the following fortnight.

The Research Intervention

Mothers who agreed to participate in the study were interviewed once, at a time and place that was most suitable for them. Thus for the most part, interviews were carried out in parents' homes, although on two occasions the interview was conducted at the hospital where the child had received surgical treatment.

i. The Explanation of the Study

Before beginning the interview, the researcher explained the study to the parent again, and the opportunity was given to parents to ask questions. Mothers were also given an information sheet to read which reiterated the aims and broader context of the study, what was involved in participating, the confidential status of the information elicited, and the voluntary nature of participation (Appendix III). Parents were required to give written consent in order to be interviewed, and also for the interview to be audiotaped (Appendix III).

ii. The Interview

Once consent was given by the parent, the interview was carried out by the researcher. This lasted one to one and a half hours, and was audiotaped. The interview had a semi-structured format, which incorporated two previously developed measures relating to parental resolution of trauma, and to adjustment to birth of a child with congenital disfigurement. These instruments are described in greater detail in the following section. As noted above, the researcher was aware of whether or not the mother had received a prenatal diagnosis with regards to families from two of the craniofacial units, however for over half of the sample (N=25), the researcher was blind to prenatal status.

iii. The Questionnaires

Following the interview, parents completed three standardised questionnaires, which measured current mood disorder, social support, and quality of the parental relationship. Again, these questionnaires and their psychometric properties are outlined in the following section.

Final Contact

Parents had been given the contact address and telephone number of the researcher, and were asked to contact the researcher with any questions or comments that they might have following the interview. Several weeks after the interview, parents were sent a letter thanking them for their participation (Appendix II). Following the collection and analysis of the data, a brief report summarising the research findings was sent to all parents.

Piloting

The measures were piloted on the first five participants in the study. No changes to the measures or to the research intervention procedure were made, and so all five pilot interviews were included in the final sample group.

Measures

1. The Interview

The interview is semi-structured in format and incorporates two previously developed measures, together with questions specifically designed for this study. In addition to demographic information about the child, parents and family, questions in the interview focus on five main areas:

i. The Cleft Lip and Palate.

The following information was sought about the child's cleft lip and palate:

* Severity of the cleft. This rating was based on the type of cleft. Unilateral cleft lip and palate was rated as less severe than bilateral cleft lip and palate, as it involves less craniofacial structure than the bilateral cleft, and the post-operative results tend to result in less residual disfigurement;
* Prenatal diagnosis of cleft lip and palate;
* Times and numbers of operations;
* Feeding difficulties;
* Family history of cleft lip and palate.

ii. Parental Resolution of Loss (Appendix IV)

Healthy coping or resolution of the loss or trauma associated with the birth of a child with craniofacial abnormality was measured using the Reaction to Diagnosis Interview (RDI) (Pianta & Marvin, 1992; Pianta, Marvin, Britner-Preston & Borowitz, 1996). This interview is intended for research use with populations of parents of children with a disability or chronic illness, and is designed to measure parents' reactions to, and coping strategies for dealing with, the diagnosis of their child's disabling condition.

This is a structured, standardised interview consisting of five questions which explore the following areas:

1. The parent's memories of first realising that their child had a medical problem.
2. Their feelings at this time.

3. The parent's perceptions of how their feelings have changed since the initial realisation.
4. Episodic memory for the moment of receiving the formal diagnosis.
5. Explanations that the parent may have for why their child has a cleft lip and palate.

The interview takes approximately fifteen minutes to administer. The five questions are designed to elicit both content and affect regarding the parent's internal working model of the child's medical condition, as the condition relates to the child, the parent, and to the health care system. The assessment of episodic memory is an important component of this interview, in particular the extent to which parents' recall is clear and unconfounded, or whether defenses prevent memory for detail. Furthermore, search is a normal process following loss or trauma (Bowlby, 1970), however resolution of loss and reorientation to the present is predicated on terminating search behaviour. Consequently, the focus on both functional and existential explanations that parents may have for why their child has cleft lip and palate, allows for an assessment of parent's search behaviour at a mental level.

Probes are limited to re-asking the parent for specific, episodic details when these are not forthcoming, or to clarify a question for the parent. No more than two probes are used to elicit episodic detail for any given part of a question.

The interview is audiotaped and transcribed verbatim with the inclusion of observations of non-verbal communications (e.g. crying), and observations noted by the researcher during the interview concerning silent non-verbal behaviour (e.g. body posture). The transcript of the interview is then coded using a standardised coding procedure (Pianta & Marvin, 1993a; Pianta *et al.*, 1996). Specific verbal and non-verbal events are identified as elements reflecting either resolution of non-resolution. Once these elements are identified, an organisation- or pattern-based judgement is made on whether the mother is resolved or unresolved with regard to the diagnosis.

Major Categories

A classification of '**Resolved**' reflects a strategy in which the mother has moved beyond the crisis associated with the diagnosis: she acknowledges an attenuation in the feelings linked to the initial crisis, a reorientation to present and future, and a realistic acceptance of the reality of the child's condition.

A classification of '**Unresolved**' reflects a strategy whereby the mother has not successfully moved through and past the crisis of the diagnosis: she remains focused in an emotionally overwhelmed or angry stance; or she uses defensive exclusion, denial or deflection to avoid confronting or resolving the crisis.

Sub-classifications

Each parent's response is subsequently classified according to a particular sub-pattern within these two major groups (Pianta *et al.*, 1996). Three sub-classifications of 'Resolved' have been reliably identified:

1. **Feeling Oriented**. The parent makes it clear that expressing feelings was an important part of coping with the diagnosis and related experiences.

2. **Action Oriented.** Whilst acknowledging their feelings, the parent does not emphasise them, but tends towards action and caregiving for the child.

3. **Thinking Oriented.** Refers to a strong intellectual orientation to reporting, with an emphasis on cognitive processes, and on the beliefs that helped.

There are six sub-classifications within the major 'Unresolved' category:

1. **Emotionally Overwhelmed**. The parent is distressed by interview topics or when asked to recall feelings and events, and has great difficulty maintaining composure. The primary feature is that the parent is actually grieving during the interview. She appears to be still actively in the crisis period of grieving the sadness and pain of the diagnosis.

2. **Angrily Preoccupied.** This refers to expression of anger during the interview, which may be directed at a number of targets, and which may be realistic or unrealistic. Anger may also be reflected in the style of response.

3. **Neutralising.** The parent avoids the emotion associated with the interview and the events being recalled. This can be seen as active avoidance where details of

events are not remembered, or a more complicated strategy, where there is recall of detail but no recall of emotion.

4. **Depressed / Passively Resigned**. This may be observed in the parent's style of response or interaction, for example, the parent may appear sad, listless and very passive, requiring many probes and much effort on the part of the interviewer. The content of the interview may also be depressed. The parent may appear stuck in the sadness of the experience, or overwhelmed at the prospect of caring for the child with very little hope for the future.

5. **Distorting.** The primary feature of this sub-category is a lack of balanced perceptions. This may involve unrealistic expectations for the child, or an unbalanced perception regarding the benefits of the experience for the parent. Beliefs have an all or none quality, there are apparent internal contradictions in beliefs that are not acknowledged, and parts of the belief system are separated from others. There may be a disproportionate emphasis on one facet of experience at the expense of other facets.

6. **Confused.** This sub-category is marked by incoherence in the content of responses, and by appearance of confusion and disorganisation in the presentation of the story.

Results from previous studies indicate that the Reaction to Diagnosis Interview is a reliable and valid procedure for assessing a mother's success in resolving the crisis and grief regarding her child's diagnosis (Pianta & Marvin, 1993b, 1993c). Pianta & Marvin (1993b, 1993c) report an inter-rater percentage agreement of 94 and 92%.

Inter-rater Reliability

Thirty of the 44 interview transcripts (68.2%) were coded by two independent raters with previous experience of the standardised coding procedure. The percentage agreement between the two raters on the primary classification of Resolved / Unresolved was 83.3%, and therefore good. Cohen's kappa was calculated in order to control for chance agreement. This gave an agreement statistic of .63, which is marginally acceptable. All disagreements were conferenced. The remaining 14 transcripts were coded jointly by the raters.

The use of two independent raters allowed a comparison of the Reaction to Diagnosis Interview with another measure related to parental adjustment, the Indicators of Parental Adjustment to Congenital Disfigurement (Bradbury & Hewison, 1994), which was rated by the researcher.

iii. Mothers' perceptions of the pregnancy and birth.

Mothers were asked about medical problems experienced during the pregnancy and birth of the child. The impact of a prenatal detection of the cleft lip and palate on mothers' perception of the pregnancy, was also explored where appropriate.

iv. Indicators of Parental Adjustment to Congenital Disfigurement (IPACD)

This is a series of indicators developed by Bradbury and Hewison (1994), which focus on early parental responses to the birth of a baby with congenital anomalies (see Appendix V). This descriptive measure aims to quantify parental responses to the birth of a baby in terms of adjustment, and to identify areas of potential difficulty in a clinical setting. It is retrospective and relies on parental self-report. Developed from attributional interviews with parents of children with congenital anomalies including cleft lip and palate, this is not a standardised measure and there is no information available on its psychometric properties. It was therefore intended that responses elicited by the RDI on resolution of loss and trauma associated with the birth of the child, would provide a source of comparative information, in order to begin to explore whether the RDI and IPACD assess similar constructs, despite the two measures asking very different questions.

There are six items:

* early emotional responses to the baby's condition and duration of overt grieving and distress;
* social exposure of the child;
* the taking of photographs;
* the effect of surgery on the acceptance of the child;
* the effect on decisions about having more children;
* the effect on the mother / father relationship;

Many of the original questions in Bradbury and Hewison's (1994) attributional interview were similar to those used in the RDI. To avoid repetition, a different set of questions was used, which were designed to be directly related to the six indicators of adjustment.

These questions were piloted in the first five interviews conducted in the study. No changes were made.

On the basis of mothers' responses to these questions, each item is rated on a three point scale, indicating level of adjustment. The overall score thus ranges from 6 (most rapidly adjusted) to 18 (slowest to adjust). Nine was designated the threshold score indicating 'well-adjusted' versus "poorly-adjusted' (Bradbury & Hewison, 1994).

iv. Preparation and Support

Mothers were asked about sources of emotional support in addition to sources of information about cleft lip and palate, following the diagnosis.

v. Attitude to Prenatal Diagnosis of Cleft

Mothers were also asked about their attitude to detection of cleft lip and palate during pregnancy.

2. The General Health Questionnaire (GHQ-28)

An assessment of psychological well-being was obtained using the General Health Questionnaire (GHQ-28; Goldberg & Hillier, 1979). This is a self-administered, screening instrument designed to detect current psychiatric illness, rather than make clinical diagnoses. Emphasis is placed on changes in condition, rather than on the absolute level of the problem, and therefore items assess the person's present state in relation to their 'usual' state, with responses ranging from, 'less than usual', to 'much more than usual'. The GHQ-28 is an abbreviated version of the main, sixty-item questionnaire, and contains items selected via factor analyses which have identified four scales measuring somatic symptoms, anxiety and insomnia, social dysfunction and severe depression. These scales are not independent of each other, with correlations ranging from 0.33 to 0.58 (Goldberg & Hillier, 1979).

This version of the GHQ takes 3 - 4 minutes to complete. Items are scored by rating problems as present or absent using a scoring system known as the 'GHQ score', whereby responses are coded 0-0-1-1. The final scores (overall, and for each of the four scales) can be interpreted as indicating the severity of psychological disturbance on a continuum. The threshold score is 4 / 5. This indicates the probability of psychiatric 'caseness' at 0.5 (Goldberg & Hillier, 1979).

The validity of the GHQ-28 has been reviewed by Goldberg & Hillier (1979). They report that correlation of the overall score with the Clinical Interview Schedule was 0.76. Reasonably high correlations of 0.73 and 0.67 were also obtained with a clinical depression rating and an anxiety rating respectively. Using the threshold score of 4 / 5, the sensitivity was 88 per cent, the specificity 84.2 per cent, and the overall misclassification rate was 14.5 per cent.

3. Social Support Questionnaire - Short Form (SSQ6)

Mothers' experience of social support was identified using the short form of the Social Support Questionnaire, (Sarason, Levine, Basham & Sarason, 1983; Sarason, Sarason, Shearin & Pierce, 1987). This Short Form version has six items, and quantifies the perceived availability of, and satisfaction with, current social support. It is designed to reliably evaluate distinct aspects of the concept of social support by assessing social supports at the time of a crisis, support networks that the individual views as unconditional, and support networks available on a daily basis. This version of the instrument takes approximately five minutes to administer.

Respondents are asked to list individuals in their environment who provide help or support. Additionally, respondents are asked to express their degree of satisfaction with that particular support on a six-point Likert scale ranging from very satisfied (1) to very dissatisfied (6). A maximum of nine persons can be listed as supports for each item, their identity being indicated by their initials and relationship to the respondent.

Two sets of summary scores are obtained. A support score for each item is derived from the number of support persons listed (the 'number score'). The mean of these scores across the total number of items gives an overall support score. A satisfaction score is based on the mean of the total number of satisfaction scores. Higher satisfaction scores indicate increased satisfaction with one's social support network.

Good results have been found for internal reliability of the SSQ6, with the alpha coefficients for availability of support and satisfaction reported as 0.90, and 0.93 respectively (Sarason *et al.*, 1987). Test-retest reliability is also acceptable, with correlations of 0.84 for the availability of support items, and 0.85 for satisfaction with support. The SSQ6 correlates highly with the SSQ (Sarason *et al.*, 1987). Significant

negative correlations have also found for the SSQ6 with a variety of personality and social competence variables including measures of anxiety, depression, loneliness, social anxiety and shyness (correlations ranged from -0.17 to -0.60) . There were no significant differences between the correlations of the SSQ6 and the SSQ with these variables. It has therefore been suggested that the SSQ6 can serve as an adequate substitute for the SSQ (Sarason *et al.*, 1987).

4. Dyadic Adjustment Scale (DAS)

The quality of the mother's relationship with her partner was assessed by the Dyadic Adjustment Scale (DAS), (Spanier, 1976). This self-report instrument consists of 32 items that measure satisfaction, cohesion, consensus and affective expression in married and unmarried intimate relationships. Most items are scored on a five- or six-point Likert-type scale. Scores from the 32 items are totalled to obtain a summary score. Total scores on the DAS range from 0 to 151, with higher scores indicative of more favourable adjustment. Scores below 98 are classified as discordant.

Criterion validity of the DAS has been demonstrated using a sample of 218 married and 94 divorced persons in the general population (Spanier, 1976), and in a group of 316 couples with chronically ill children (Gordon Walker, Johnson, Manion, & Cloutier, 1992). The DAS and Locke-Wallace Marital Adjustment Scale were found to correlate .86 for married persons and .88 for divorced persons. Test - retest reliability of the instrument has been demonstrated as favourable, with a coefficient alpha of .96 (Spanier, 1976).

RESULTS

Overview

The results of the descriptive analyses relating to the Prenatal and No-Prenatal groups are presented here. This is followed by the results of the inferential analyses, which aimed to explore differences in the two study groups in terms of adjustment to the birth of the child.

Data Analysis

The analysis of data from the interview and questionnaires followed a number of discrete stages:

1. On determining the acceptance rate for the sample, the demographic characteristics of the two participant groups were summarised in terms of age and gender of child, severity / type of cleft, age and social class of parent, and family composition. The data set was examined in terms of distributions and variances of the two populations.

2. The descriptive analyses investigated differences in the Prenatal and No-Prenatal groups in relation to the following:

   (i). mothers' experience of stressors during pregnancy, the birth and the first year of life;

   (ii). information about cleft lip and palate, and emotional support that women received following the diagnosis;

   (iii). current social support;

   (iv). the quality of the parental relationship.

3. The inferential analyses examined the adjustment of mothers in Prenatal and No-Prenatal groups to the birth of a child with cleft lip and palate, in terms of three psychological constructs:

(i). resolution of trauma associated with the diagnosis of cleft lip and palate;

(ii). parental adjustment to congenital disfigurement;

(iii). current psychological health.

4. Further analyses were carried out in order to examine the association between parental adjustment and key independent variables outlined in the descriptive analyses.

The presentation of results follows this order of analyses. Firstly, the response rate of mothers initially contacted about the study, and the final sample achieved are described. For the purpose of clarity, all percentages have been rounded off to the nearest whole number.

Acceptance Rate

The response rate for mothers who were contacted about the study is presented for each of the four craniofacial units in Table 1. Sixty-three parents were initially contacted, having all met the inclusion criteria for the study. The distribution of this group was approximately equal across the four craniofacial units (20, 24, 27 and 29%). On receipt of the initial contact letter describing the research, only three parents indicated that they did not wish to be contacted further about the study, each from a different unit. None of these parents gave a reason for their refusal. Interestingly, all three of these parents were from minority ethnic groups.

Table 1. The Acceptance Rate

	CRANIOFACIAL UNIT				TOTAL	
	One	Two	Three	Four	N	%
Contacted	15	13	17	18	**63**	(100)
Refused	1	-	1	1	**3**	(5)
No Reply	2	2	6	6	**16**	(25)
Interviewed	**12**	**10**	**10**	**12**	**44**	(70)

A quarter of parents did not reply to the initial letter, and were therefore not contacted again. The 'no response' rate for individual units ranged from 13 per cent for Unit One to 33 per cent for Unit Four. This lack of response may reflect a refusal to participate, or may be due to the letter being sent to an incorrect address.

Every parent who agreed to be contacted about the study and therefore received detailed information on the research, went on to participate. Thus, almost 70 per cent of the initially selected group, took part in the study. This final group was almost equally distributed across the four units. The acceptance rate for individual craniofacial units was generally high, ranging from 80 per cent for Unit One, to 59 per cent for Unit Three. As prenatal status was not known for over half of the parents initially contacted (N=25), it is not possible to calculate the response rates for the Prenatal and No-Prenatal groups. Nevertheless, the distribution of the two participant groups across the four units is outlined in Table 2.

Table 2. Prenatal Status by Unit

	PRENATAL		NO-PRENATAL	
	N	%	N	%
UNIT				
One	6	(30)	6	(25)
Two	6	(30)	4	(17)
Three	4	(20)	6	(25)
Four	4	(20)	8	(33)
TOTAL	**20**	**(100)**	**24**	**(100)**

Descriptive Analyses

The demographic composition of the Prenatal and No-Prenatal groups is described in terms of factors pertaining to the child and then to the mother and the family.

The Child

At the time of interview, the mean age of the child was 19 months (SD = 8.2; range: 9 - 34 months). There was no difference in the age of children in each of the two study groups (Prenatal group: M = 18, SD = 7.5; No-Prenatal group: M = 21, SD =8.7; t = 1.17, N.S.).

The figures in Table 3 show that approximately half of the entire group are female. This even gender distribution is not reflected in the individual study groups. Sixty per cent of the Prenatal group are female, whilst the reverse is true for the No-Prenatal group, of which two thirds are male ($\chi^2 = 2.21$, d.f. = 1, N.S.). The gender distribution of the No-Prenatal group best represents that found in the general cleft lip and palate population (Melnick, 1990).

Table 3. Prenatal Status and Gender of Child

	PRENATAL		NO-PRENATAL		Total	
	N	%	N	%	N	%
Girls	12	(60)	9	(37)	**21**	(48)
Boys	8	(40)	15	(63)	**23**	(52)
TOTAL	**20**		**24**		**44**	**(100)**

There are almost twice as many children in the overall sample with unilateral (less severe) cleft lip and palate as there are children with bilateral (more severe) cleft (Table 4). This difference is also observed in both the Prenatal and No-Prenatal groups, and

reflects the distribution of unilateral and bilateral cleft lip and palates found in the general population (Brohnshtein *et al.*, 1991).

Table 4. Prenatal Status and Cleft Type

	PRENATAL		NO-PRENATAL		Total	
	N	%	N	%	N	%
Unilateral	13	(65)	16	(67)	**29**	(66)
Bilateral	7	(35)	8	(33)	**15**	(34)
TOTAL	**20**		**24**		**44**	**(100.0)**

The Parent and her Family

Mothers in the participant groups were all primary caretakers of the index child. At the time of interview, the mean age of the mothers was 32 years (SD = 4.5; range: 22 to 43 years). There was no difference in the mean ages of mothers in the two study groups (Prenatal: M = 31 years, SD = 4.2; No-Prenatal: M = 32 years, SD = 4.7).

All participants cohabited with the father of the index child. Fathers were aged between 26 and 45 years, with a mean age of 33 years (SD = 4.9). Again, there was no difference in the mean ages of fathers in the two participant groups (Prenatal: 34 years, SD = 5.5; No-Prenatal: 33 years, SD = 4.5). Almost two thirds of the overall participant group (64%) had been married to, or had lived with the father for five or more years. This proportion of relationships of five or more years is reflected in both the Prenatal and No-Prenatal groups (65% and 62% respectively).

The social status of the families in the sample is described in two ways. Firstly, social status was determined by the Registrar General's Classification of Social Class, based on the occupation of the father (Office of Population Censuses and Surveys, 1991), as shown in Table 5. All of the fathers in the sample were in full-time employment. The

overall sample was distributed equally between non-manual (N=22), and manual (N = 22) occupations.

Table 5. Social Class by Occupation of Father

	Prenatal		No-Prenatal		ALL	
	N	(%)	N	(%)	N	(%)
Registrar General's Social Class coding						
I	4	(20)	5	(21)	9	(21)
II	4	(20)	5	(21)	9	(21)
III non-manual	1	(5)	3	(12)	4	(9)
III manual	1	(45)	3	(29)	16	(36)
IV	9	(10)	7	(13)	5	(11)
V	-		1	(4)	1	(2)
	20		**24**		**44**	**(100)**

Similar distributions were found in the Prenatal and No-Prenatal groups. Forty-five per cent of the Prenatal group were classified as non-manual (social classes I, II and III), compared with 54 per cent of the No-Prenatal group. Compared with figures for the 1981 Population Census for England and Wales (Central Statistics Office, 1995) the proportion of families was far higher in Social Class I, and lower in Social Class III non-manual, than would be expected. This disparity is likely to reflect the general bias towards middle and upper class participants, found in psychological research (Graham, 1992).

Additionally, social status was classified according to maternal education, as shown in Table 6.

Table 6. Maternal Educational Status

	Prenatal		No-Prenatal		ALL	
	N	(%)	N	(%)	N	(%)
Educational Status						
1	14	(70)	12	(50)	26	(59)
2	6	(30)	12	(50)	18	(41)
	20		**24**		**44**	**(100)**

On the basis of this classification, Group 1 refers to educational attainment of A level (or equivalent) and above, whilst Group 2 includes O level, CSE and equivalent qualifications, in addition to no qualifications. Overall, 60 per cent of the sample were classified in Group 1. The Prenatal and No-Prenatal groups did not differ significantly in terms of maternal educational status ($\chi^2 = 1.81$, d.f. = 1, N.S.).

The ethnicity of most of the mothers (N=40) was white British. The remaining women (N=4) were white European, namely Irish, Swedish and Hungarian. The absence of mothers from minority ethnic groups in the sample may be understood in a number of ways. Firstly, the incidence of cleft lip and palate in the Black population is lower than in the Caucasian population (Frazer, 1970; Melnick, 1990). This decreases the likelihood that a Black family would be selected in the study. Furthermore, refusals to participate in this study came from parents from minority ethnic groups, which may reflect the tendency for low take-up rates found in Black and Asian populations, resulting in the under-representation of minority ethnic groups in psychological research (Graham, 1992).

In terms of the family composition, the majority of children in this sample (N=20, 45%) were single children. A further 19 children had one sibling, whilst only five children

had three or more siblings. Two children in the group had a twin sibling. There was no difference between the Prenatal and No-Prenatal groups in term of birth order (χ^2 = 0.69, d.f. = 2, N.S.), or family size (χ^2 = 0.64, d.f. = 2, N.S.).

There was a known history of cleft in 10 of the families. In half of these families (N=5) cleft lip and/or palate was traced to a member of the father's family of origin, and in two further cases, the father of the index child had a cleft. Similarly, two mothers in the group had cleft lip and/or palate. In a further case, the sibling of the index child also had cleft lip and palate. In terms of prenatal status, five of the families with a history of cleft had the prenatal diagnosis of cleft (two having been screened specifically for cleft), whilst the remaining five received the diagnosis at birth. Furthermore, the 10 families were equally distributed across the Prenatal and No-Prenatal groups in terms of the family member with cleft (parent vs. other family member).

The experience of mothers during the pregnancy and the birth of the index child, in addition to the first year of the child's life, was examined for each of the two study groups.

The Pregnancy and Birth

The majority of women reported no significant medical problems during their pregnancy (67% in the No-Prenatal group and 75% in the Prenatal group). However, a small group of women (N = 13) did experience physical problems during pregnancy, ranging from prolonged nausia and sickness, to more serious medical complications such as diabetes mellitus and preeclampsia.

All of the mothers in the study had undergone ultrasound scanning as part of routine prenatal screening. A family history of cleft was not necessarily a precursor to specific screening for cleft. Only two of the 10 women with a family history of cleft experienced specific screening. In both cases, the cleft was detected. As one might expect, women did not experienced specific screening when there was no family history of cleft.

Three quarters of women in the Prenatal group (N = 15) reported that the detection of cleft in utero had a negative impact on their pregnancy, clearly borne out in the following statements:

It takes the edge off (the pregnancy). I was definitely more anxious the last six weeks, and was convinced by the end of the pregnancy that he had a club foot as well. I think it's taken the edge off my other pregnancies since.

I was really, really devastated. It was the worse thing that could ever have happened. Before the scan I was over the moon about the baby and starting a family. I felt so proud and well. But after the scan I started to regret the whole thing. It was always on my mind, and for weeks afterwards, every time I thought about it, I burst into tears.

In a number of cases, the diagnosis of cleft lip and palate was made after a repeated scans over several days or even weeks. The detection of cleft also led some women to take further, more invasive tests for chromosomal abnormalities. For some, this was a period of crisis, illustrated by one mother's description:

I felt I'd lost five days out of my life, because it was just horrendous not knowing if anything else was wrong. And also coming to terms with the fact she'd got this defect. I honestly didn't know how we were going to cope with it, and tell our parents, and what it would actually mean when she was born. I don't know how many women actually go through a pregnancy without worrying at all, but it really did ruin my pregnancy. All I could think was I want this to be over. I want her to be born. I want to deal with it.

The remaining 25 per cent of the group reported that the prenatal detection of cleft had made no difference to their experience of being pregnant:

I didn't go around telling people. I didn't feel it was a big deal. I didn't feel I was different and I didn't want to be treated differently.

I was happy with the way the pregnancy was going. I just felt more protective over her after the scan. It never entered my head to terminate just because she wasn't perfect.

On detection of the cleft, four mothers in the Prenatal group (20%) reported that they were offered a termination.

A substantial proportion of mothers reported medical problems during the birth, such as complications arising from toxaemia, or the baby becoming distressed during labour, or having breathing difficulties at birth (No-Prenatal: N = 11, 46%; Prenatal: N = 4; 20%). However, this difference between the two groups fails to reach statistical significance ($\chi^2 = 3.24$, d.f. = 1, $p < .08$).

On the whole, the response of families following the birth of the baby with cleft lip and palate, was reported as positive or neutral by mothers in both the Prenatal and No-Prenatal groups. However, over a third of mothers in both groups reported that their family reacted negatively to the new baby's cleft (Prenatal: 35%; No-Prenatal: 37%):

What hurt me the most was when she (parent's mother) referred to him as ' damaged goods'.

One woman describes the reaction of her mother-in-law:

She wanted me to have an abortion. She said they'd never had 'that sort of thing' in the family before.

The First Year

The immediate difficulty faced by many mothers following the birth, was feeding their baby. Eighty per cent of mothers reported feeding problems arising from the cleft lip and palate, which included the infant being unable to form adequate suction, and the tendency for the infant to swallow too much air, regurgitate feed, or for food to come down the nose. The high proportion of mothers experiencing feeding difficulties was reflected in both the Prenatal and No-Prenatal groups (80 and 83% respectively).

For many mothers, these problems resulted in unsuccessful and often distressing attempts to breastfeed, or in lengthy and frustrating feeding times. It is noteworthy that feeding difficulties were often severely exacerbated by the absence of advice or specialist feeding equipment available during the infant's first few days of life. Problems tended to persist into the first few months of life, and in some cases until the

infant had the palate operation at six to seven months. At the time of interview, significant feeding problems had been resolved for all children.

The operations that the infant underwent during the first year of life in order to repair the cleft lip and palate, were also a significant source of stress reported by mothers. The child's age at the first operation to repair the lip ranged from 2 days to three months. The mean age for children in the Prenatal group was 34.5 days (SD = 1.2), and 26.2 days (SD = 1.3) for children in the No-Prenatal group. The frequency of age at first operation was not normally distributed. A non-parametric test (Mann-Whitney) was therefore computed to test for differences in age at first operation between the two study groups. No difference was found between the age at first operation of children in the Prenatal and No-Prenatal groups (U = 241.5, two-tailed, N.S..).

On average, infants underwent 2 operations, although a small number of infants (N=4) had either four or five operations during the first year. There was no difference in the number of operations in terms of prenatal status. At interview, the time since the child's last operation was also calculated. A wide range was found within the entire sample, from one to 28 months. For children in the Prenatal group, the mean number of months since the last operation was 10.6 (SD = 7.4). The mean number of months in the No-Prenatal group was marginally, but not significantly higher, at 13.7 (SD = 7.7); (t = 1.37, N.S.).

Information and Emotional Support following Diagnosis of Cleft Lip and Palate

Mothers were asked about the information they had received about cleft lip and palate when the diagnosis of cleft was made. Such information pertained to the nature and aetiology of the condition, surgical treatment, in addition to associated problems such as feeding, speech and hearing, and methods of managing these difficulties. There was a strong tendency for mothers in the Prenatal group to emphasise the importance of information at that time, and to describe themselves as actively seeking information about cleft following diagnosis, for example:

I wanted as much information as possible, as quickly as possible, preferably in print.

Whereas mothers in the No-Prenatal group were more likely to see themselves as passive recipients of information, or avoided information altogether:

I didn't go searching for information. I just got it from the surgeon.

I didn't want to know anything. To tell the truth I just wanted to block it all out.

This difference observed between the two groups in terms of information-seeking/avoiding style, was significant ($\chi^2 = 3.26$, d.f. = 1, $p < .05$).

Not surprisingly, the most frequently reported source of information found in the entire sample was the specialist cleft surgical team (93%). Over half of mothers reported that they received information from the Cleft Lip and Palate Association (59%), and a similar proportion of mothers received information from families and friends (54%), and from health professionals who were not specialists in cleft lip and palate (e.g., midwives, paediatricians, health visitors, general practitioners and radiologists) (54%). Other parents of a child with cleft lip and palate were a source of information for 47 per cent of mothers, and books and television programmes informed 34 per cent of mothers about cleft.

The amount of sources of information did not differ significantly between the two groups (t = 0.58, N.S.). Nevertheless, some differences emerged between Prenatal and No-Prenatal groups in terms of where the information about cleft came from. Specialist cleft teams, voluntary organisations such as the Cleft Lip and Palate Association, family, friends and other parents of children with cleft lip and palate were reported as sources of information by mothers in both groups. However, mothers who received the diagnosis of cleft during pregnancy were more likely to seek information from magazine articles, television programmes, medical textbooks or books about childcare ($\chi^2 = 7.13$, d.f. = 1, $p < .01$). In addition, health professionals who were not craniofacial specialists, such as midwives, paediatricians and health visitors, were more than twice as likely to be a source of information about cleft lip and palate when diagnosis was received at birth, than when the cleft was detected in utero (71% vs. 29%; $\chi^2 = 5.64$, d.f. = 1, $p < .05$).

These differences in sources of information reported by Prenatal and No-Prenatal mothers may be related to the differing information-seeking styles observed in the two groups. However, the differing circumstances surrounding receipt of the diagnosis of cleft are more likely to play an explanatory role here. Mothers in the Prenatal group had time during pregnancy to search for information from books and magazine articles themselves, whilst mothers in the No-Prenatal group required information as soon after the birth as possible, and therefore relied on the health professionals that they came into contact with at that time for this information.

It is particularly noteworthy that for those mothers in the No-Prenatal group, the quality of the first information received about cleft may have been less adequate than that given to women who received the diagnosis of cleft during pregnancy, because it was provided by health professionals who may not have specialist knowledge about the condition. This is borne out in the reports of many women in the No-Prenatal group, for example:

I had to find it all out for myself. The doctors and midwives were 10 years behind with the information they could give me. They were ignorant about it and I had to educate them.

It was the lack of explanation, you know. As far as I was concerned, the baby had cleft lip and palate and that was it, and I didn't know you could get them repaired or anything. But there wasn't anybody there to explain to me what happens. So I felt at a loss, and this went on until the surgeon came to see me the next day.

The inadequacy of knowledge available to some mothers at this early stage, had a significant impact on problems such as feeding:

I almost felt we were in limbo. E had a problem and we couldn't feed her, and there was no one there who could help us.

Emotional support following the diagnosis of cleft was considered important by the overwhelming majority of mothers (87% of the No-Prenatal group and 80% of the Prenatal group). Aspects of emotional support valued by mothers included reassurance, allowing the mother to express and explore her feelings, taking a non-judgmental view

of the child, and helping to put the problem into perspective. For some women, emotional support was not available, particularly from health professionals:

I don't think there was any thought for my feelings at all. Nobody asked me how I felt.

Although for others, the need for emotional support was not a priority during the period immediately following diagnosis:

I had a conversation with a CLAPA person who was very supportive and reassuring, but it was too early at that stage to know that everything was going to be alright in the end. I just needed to know what it was and what could be done.

As one might expect, family and friends were the most frequently reported source of emotional support in the sample as a whole (N = 35, 79%). Less than half of the participants (N = 18, 41%) reported that their partner had supported them emotionally at the time of diagnosis. Health professionals such as midwives and paediatricians were viewed by 29 per cent of mothers as supportive. Furthermore, representatives from the Cleft lip and Palate Association were considered a source of emotional support by 14 per cent of the overall participant group, and 11 per cent of mothers reported that members of the cleft surgical team (specialist nurse or surgeon) also fulfilled this role. The number and range of sources of emotional support following the diagnosis of cleft did not differ across the Prenatal and No-Prenatal groups.

Current Social Support

Mothers' experience of social support was examined in terms of the perceived availability of current sources of support, and also satisfaction with support, using the short form of the Social Support Questionnaire (Sarason *et al.*, 1987). The means and standard deviations for numbers of, and satisfaction with available support, are outlined for the two study groups in Table 7. Scores for the number of supports and for satisfaction with support were distributed normally.

Table 7. Social Support and Prenatal Status

	PRENATAL		NO-PRENATAL	
	M	SD	M	SD
Support:				
Number	2.9	1.5	3.7	1.8
Satisfaction	5.2	0.5	5.1	0.5

The perceived number of social supports was not found to differ significantly between mothers with a prenatal diagnosis of cleft, and mothers who did not received a diagnosis until the birth ($t = 1.46$, N.S.). Similarly, satisfaction with this support did not differ significantly between the two participant groups ($t = 0.05$, N.S.).

Differences in the amount of social support available were examined for a number of demographic variables. The median was used as the criterion for dividing ages (of mother, child and father) into two groups. The results presented in Table 8 show that those mothers whose partner was aged 33 years or less, reported a significantly greater number of social supports.

Table 8. Number of Social Supports and Demographic Variables

		SOCIAL SUPPORT: NUMBER		
	Sub-Group	M	SD	(t-value)
Age of child (Months)	≤ 17	3.6	2.1	.92
	>17	3.1	1.2	
Gender	girl	3.6	2.2	.75
	boy	3.2	1.1	
Cleft	unilateral	3.4	1.7	.08
	bilateral	3.3	1.9	
Social Class	I - III(N)	3.9	1.6	1.97
	III(M) - V	2.8	1.7	
Mother's Age	≤ 31	3.4	1.9	.28
	> 31	3.2	1.5	
Father's Age	≤ 33	4.1	2.1	2.24 *
	> 33	2.8	1.2	

* $p < .05$

Table 9 presents the means and standard deviations of satisfaction ratings of social support, for the different demographic variable sub-groups, in addition to the relevant t-values. Mothers from social classifications I, II and III non-manual, as assessed by

father's occupation, reported significantly greater satisfaction with social support. However, due to the narrow range of responses indicating satisfaction, caution must be taken when interpreting the relationship between social class and satisfaction with support.

Table 9. Satisfaction with Social Supports and Demographic Variables

		SOCIAL SUPPORT: SATISFACTION		
	Sub-Group	**M**	**SD**	**(t-value)**
Age of child (Months)	**≤ 17**	5.1	0.6	.92
	>17	5.3	0.5	
Gender	**girl**	5.2	0.6	.75
	boy	5.1	0.5	
Cleft	**unilateral**	5.2	0.5	.04
	bilateral	5.2	0.6	
Social Class	**I - III(N)**	5.4	0.5	2.61*
	III(M) - V	5.0	0.5	
Mother's Age	**≤ 31**	5.1	0.5	.93
	> 31	5.3	0.6	
Father's Age	**≤ 33**	5.2	0.5	.06
	> 33	5.2	0.6	

* $p < .05$

The Parental Relationship

The quality of the mother's relationship with her partner was assessed in the Prenatal and No-Prenatal groups using the Dyadic Adjustment Scale (DAS) (Spanier, 1976). This measure provides a total score indicating the quality of the relationship, which can range from 0 to 151, with higher scores indicative of more favourable adjustment. In the sample as a whole, DAS scores were normally distributed, and ranged from 76 to 133, with a mean relationship score of 109.6 (SD = 13.8).

The overwhelming majority of women in the entire sample (N = 37, 84%) had relationships that scored above the accepted threshold score of 98, and were therefore rated as well-adjusted. As so few relationships (N = 7) were rated as poorly adjusted, the total DAS score has been used to express the quality of the relationship.

Differences in the quality of the parental relationship were examined for mothers in the Prenatal and No-Prenatal groups. The mean DAS score for mothers in the Prenatal group was 113.4 (SD = 13.6). This compares with a mean score of 106.4 (SD = 13.6) for the No-Prenatal group. However, this difference observed in the DAS scores for the study groups failed to reach statistical significance (t = 1.72, N.S.).

Independent t-tests were computed to examine whether there were any differences in DAS scores in terms of a number of demographic variables. Table 10 presents the means and standard deviations of DAS scores for the demographic variable sub-groups, in addition to the relevant t-values.

These figures show a statistically significant difference in DAS scores in terms of gender of the child, whereby mothers of girls have reported better - adjusted relationships with their partners, than have mothers of boys.

Table 10. Parental Relationship and Demographic Variables

		Parental Relationship (DAS)		
	Sub-Group	**M**	**SD**	**(t-value)**
Age of child (Months)	**≤ 17**	110.9	13.7	.73
	>17	107.9	14.0	
Gender	**girl**	116.3	9.3	3.56 **
	boy	103.4	14.5	
Cleft	**unilateral**	108.7	12.1	.54
	bilateral	111.1	16.9	
Social Class	**I - III(N)**	111.0	14.1	.68
	III(M) - VI	108.1	13.7	
Mother's Age	**≤ 31**	110.9	14.3	.67
	> 31	108.1	13.3	
Father's Age	**≤ 33**	113.8	12.2	1.82
	> 33	106.4	14.2	

** $p < .01$

Inferential Analyses

Parental adjustment to the birth of a child with cleft lip and palate was explored in the Prenatal and No-Prenatal groups, with respect to three different constructs:

* Resolution of trauma associated with the diagnosis of cleft lip and palate;
* Parental adjustment to congenital disfigurement;
* Current psychological health.

The results for each of these measures of adjustment are presented for the two study groups.

1. Resolution of Trauma associated with the Diagnosis of Cleft Lip and Palate

Firstly, parental resolution of trauma and grief associated with the diagnosis of cleft was explored using the Reaction to Diagnosis Interview (Pianta *et al.*, 1996). The impact of prenatal status on maternal resolution was examined. The results are shown in Table 11.

Table 11. Resolution of Loss in Prenatal and No-Prenatal Groups

	PRENATAL		NO-PRENATAL		Total	
	N	%	N	%	N	%
Resolved	9	(45)	4	(17)	**13**	(30)
Unresolved	11	(55)	20	(83)	**31**	(70)
TOTAL	**20**		**24**		**44**	**(100)**

Thirty per cent of mothers in the overall sample were found to be Resolved. In terms of prenatal status, a significant difference between the two groups was found, with mothers who had received the diagnosis of cleft during pregnancy more than twice as likely to be rated as Resolved, as mothers who discovered the cleft at birth (45% vs. 17%; χ^2 = 4.20, d.f. = 1, $p < .05$).

Sub-patterns of resolution and lack of resolution were explored in greater detail for the entire sample rather than for the Prenatal study groups. A meaningful comparison of resolution patterns across the two study groups was not possible, given the large number of sub-patterns relative to the sample size. Consequently, sub-classification ratings for the Resolved and Unresolved groups are presented for the whole sample in Table 12.

Table 12. Distribution of Sub-classifications for Resolved and Unresolved Groups

	UNRESOLVED			RESOLVED	
	N	%		N	%
Emot. Overwhelm	11	(36)	**Thinking**	7	(54)
Neutralising	6	(19)	**Action**	4	(31)
Distorting	5	(16)	**Feeling**	2	(15)
Confused	5	(16)			
Angrily Preoccup.	2	(6)			
Depressed/Passive	2	(6)			
TOTAL	**31**	**(100)**	**TOTAL**	**13**	**(100)**

The relative sub-group frequencies were examined within the major categories. Among the mothers in the Resolved group, over half (N = 7) were rated as Thinking-Oriented.

These parents tended to focus on their thoughts and beliefs about their experiences, particularly when asked about how they felt.

A substantial proportion of mothers within the Unresolved group were rated as Emotionally overwhelmed (N = 11). For these parents, the interview topics tended to be emotionally charged. There was also a tendency for the parent to appear to be actively in the crisis period of grieving the sadness and pain of the diagnosis. A very small number of parents were rated as either Passive / Depressed or Angrily Preoccupied. Whilst the remaining parents were almost evenly distributed across the Neutralising, Distorting and Confused sub-patterns.

In order to gain a clearer understanding of the association between prenatal detection of cleft lip and palate and successful resolution of trauma, the analyses aimed to answer the following questions:

i. Is resolution associated with background variables relating to the mother, child or family?

The descriptive analyses demonstrated that the Prenatal and No-Prenatal groups did not differ significantly in terms of demographic variables. However, it is also important to investigate the potentially confounding role of demographic variables relating to the mother, child and family. Analyses showed that resolution was not related to the child's age ($t = 1.07$, N.S.), or gender ($\chi^2 = 0.63$, d.f. = 1, N.S.), or to the severity of the child's cleft ($\chi^2 = 0.15$, d.f. = 1, N.S.). Nor was resolution associated with the unit where the child received treatment ($\chi^2 = 2.76$, d.f. = 3, N.S.).

Furthermore, there was no difference in resolution status in terms of mother's age ($t = 0.7$, N.S.), father's age ($t = 0.60$, N.S.), social class by paternal occupation ($\chi^2 = 0.98$, d.f. = 1, N.S.), or family history of cleft ($\chi^2 = 0.57$, d.f. = 1, N.S.).

These results suggest that the relationship between resolution and prenatal status is not confounded by demographic variables.

ii. Is the trauma or crisis associated with diagnosis more likely to resolve over time?

This question was initially addressed by investigating the relationship between age of child and resolution of trauma. An independent samples t-test showed that mothers of older children were not more likely to be resolved, than those mothers of younger children ($t = 1.07$, N.S.).

The question was subsequently explored in more detail, by examining the relationship between resolution and months since diagnosis either at prenatal scan, or at the birth. The mean number of months since diagnosis was 23.1 (SD: 9.1) for mothers who were rated Resolved, and 20.9 (SD = 7.5) for mothers rated Unresolved. An independent samples t-test showed that the length of time since diagnosis of cleft was not significantly greater for mothers who were resolved, compared with mothers who were rated Unresolved ($t = 0.84$, N.S.). As the distribution of months since diagnosis was positively skewed, a non-parametric test was also computed (Mann-Whitney). The results confirm the findings from the parametric test ($U = 169.5$, N.S.).

iii. Is lack of resolution related to the experience of stressors during the pregnancy, birth and infancy?

Again, the descriptive analyses demonstrated that the Prenatal and No-Prenatal groups did not differ significantly in terms of stressors experienced in pregnancy, at the birth or during the first year. Further analyses showed that resolution status was not related to difficulties experienced either during pregnancy, or at the birth.
These include:

* the presence of medical problems during pregnancy ($\chi^2 = 1.77$, d.f. = 1, N.S.);
* the negative impact of diagnosis on the pregnancy ($\chi^2 = 0.07$, d.f. = 1, N.S.);
* the presence of medical problems at the birth ($\chi^2 = 0.99$, d.f. = 1, N.S.).

In addition , resolution was not found to be associated with family response to the baby following the birth ($\chi^2 = 0.04$, d.f. = 1, N.S.).

There was also no association between parental resolution of trauma and stressors that occur during the first year of life such as feeding difficulties ($\chi^2 = 0.14$, d.f. = 2, N.S.), or multiple operations ($F = 0.18$, N.S.).

iv. Does the amount and nature of support and information received after diagnosis of cleft, impact on the mother's resolution of trauma?

A recognition of the role of initial emotional support in coping with the diagnosis of cleft, was associated, albeit not significantly, with resolution of trauma and crisis. All of the mothers rated as Resolved reported that emotional support from either family and friends, health professionals or voluntary organisations was crucial when they first received the diagnosis. This compares with only 77.4% of mothers rated as Unresolved However, this tendency failed to reach statistical significance (χ^2 = 3.49, d.f. = 1, Fisher's Exact Test: one tail, p <.07).

The number of sources of emotional support and of information that mothers received following diagnosis of cleft was not related to resolution. Furthermore, the actual source of information or support, whether it be the specialist team, or friends and family, was not associated with whether or not the mother was resolved. Findings therefore suggest that the actual existence or availability and perhaps also the quality, rather than the quantity or source of support and information, in addition to the mother's ability to acknowledge her emotional needs, may be more important in the resolution of grief and trauma associated with the birth of a child with cleft lip and palate.

v. Are mothers more likely to have resolved the grief and trauma that they have experienced, if they have adequate social support?

Social support appears to have little impact on resolution of trauma. There was no difference in the perceived number of supports currently available to mothers who were classified as Resolved, compared with those who were Unresolved (t = 1.09, N.S.). Similarly, mothers' reported satisfaction with social support did not differ in terms of resolution status (t = 0.20, N.S.).

Further analyses were conducted to examine the potentially moderating effect of social support on the relationship between prenatal status and resolution of trauma. The results from a logistic regression show no evidence of an interaction effect for the number of current social support on the significant association between prenatal status and resolution of trauma (B = -0.01, Wald = 0.00, d.f. = 1, N.S.).

Similarly, maternal satisfaction with support did not have a moderating effect on the relationship between prenatal status and resolution of trauma (B = -0.17, Wald = 0.14, d.f. = 1, N.S.).

vi. Does the quality of the mother's relationship with her partner influence her ability to resolve her grief?

The quality of the mother's relationship with her partner, as measured by the DAS, is not related to resolution of loss. No difference in resolution was found between those mothers whose relationship was rated as poorly adjusted, compared with those with well adjusted relationships (χ^2 = 0.93, d.f. = 1, N.S.). Similarly, when viewed as a continuum, the adjustment of the parents relationship was not significantly greater for those parents who were rated as Resolved (t = 0.44, N.S.).

A logistic regression was computed to investigate the possibility that the quality of the parental relationship may have a moderating effect on the relationship between prenatal status and resolution of trauma. However, results confirmed the absence of an interaction effect (B = -.08, Wald = 1.8, d.f. = 1, N.S.).

2. Parental Adjustment to Congenital Disfigurement

Mothers' adjustment to congenital disfigurement of their child was explored by investigating parents' emotional, cognitive and behavioural responses following the birth of the child, and during early infancy, as measured by the Indicators of Parental Adjustment to Congenital Disfigurement (IPACD) (Bradbury & Hewison, 1994).

In the sample as a whole, the mean total score indicating overall adjustment to the birth of a child with a congenital disfigurement, was 10.8 (SD = 2.4). Scores were normally distributed, and ranged from 7 to 17, with higher scores indicating more difficult adjustment. The mean IPACD scores for the two study groups were not significantly different (Prenatal: M = 10.3, SD = 2.1; No-Prenatal: M = 11.33, SD = 2.5; t = 1.42, N.S.). Consequently, prenatal diagnosis of cleft lip and palate is not related to parental

adjustment to the birth of a child with congenital disfigurement, as measured by the IPACD.

Further analyses were carried out within the entire sample, in order to gain a more comprehensive understanding of parents' adjustment to congenital disfigurement. These analyses focused on IPACD scores in relation to background or demographic variables, pre- and postnatal stressors, information and support received following diagnosis of cleft, current social support, and the quality of the parental relationship.

i. The Child, the Mother and the Family

The relationship between parental adjustment to congenital disfigurement, expressed by the total IPACD score, and background variables relating to the child and the parents, was investigated by computing a number of independent t-tests. The results are presented in Table 14.

A number of differences emerged at the five per cent level of significance. Mothers in social classes IV (i.e. III manual), V and VI scored higher in terms of adjustment to congenital disfigurement, than did mothers in social classes I to III. A higher level of adjustment was also found in mothers over the age of 31 years (median age). Furthermore, adjustment to congenital disfigurement was not related to craniofacial unit ($F = .92$, N.S.).

Table 13. Adjustment to Congenital Disfigurement and Demographic Variables

		IPACD		
	Sub-Group	**M**	**SD**	**(t-value)**
Age of child (Months)	**<=17**	11.55	2.5	1.75
	>17	10.3	2.2	
Gender	**girl**	10.8	2.0	.14
	boy	10.9	2.7	
Cleft	**unilateral**	10.8	2.3	.14
	bilateral	10.9	2.7	
Social Class	**I - III(N)**	10.4	2.0	2.35 *
	III(M) - VI	11.7	2.5	
Mother's Age	**≤ 31**	10.1	1.8	2.29 *
	> 31	11.7	2.7	
Father's Age	**≤ 33**	10.8	2.3	.17
	> 33	10.9	2.5	
Family History	**no**	10.8	2.7	.28
	yes	11.0	1.4	

* $p < .05$

ii. Pre- and Postnatal Stressors

* Pregnancy

The emotional impact of prenatal diagnosis of cleft on the pregnancy was not related to adjustment, as measured by the IPACD ($t = .84$, N.S.). However, the presence of medical problems during pregnancy was related to parental adjustment to congenital disfigurement. The adjustment of mothers who did not experience problems was rated as significantly higher, than the adjustment of mothers who had prenatal medical difficulties ($t = 2.74$, $p < .01$).

* Birth

The presence of medical difficulties at the birth was not related to parental adjustment to congenital disfigurement ($t = 1.90$, N.S.). Furthermore, parental adjustment to congenital disfigurement was related to mother's report of her family's initial response to the baby. Mothers whose family had reacted negatively, were rated as significantly less adjusted than those mothers whose family's response was positive or neutral ($t = 2.18$, $p < .05$).

* Infancy

There was no relationship between parental adjustment to congenital disfigurement and feeding difficulties ($t = .79$, N.S.), or the number of operations that the child has undergone during the first year of life ($F = .79$, N.S.).

iii. Information and Emotional Support at Diagnosis

The number of sources of information about cleft lip and palate, and of emotional support received by mothers following diagnosis of cleft, were not related to parental adjustment to congenital disfigurement ($t = 1.08$, N.S.; $t = .73$, N.S.).

iv. Current Social Support

Correlational analyses were computed in order to examine the association between social support and adjustment to congenital disfigurement. Neither the number of supports available ($r = -0.08$. N.S.), or maternal satisfaction with support ($r = -0.23$,

N.S.), were significantly associated with parental adjustment to congenital disfigurement of their child.

v. Quality of Parental Relationship

The quality of mothers' relationships with their partners was found to negatively correlate with levels of adjustment to congenital disfigurement ($r = -0.34$, $p < .05$). This indicates that mothers whose relationships were more adjusted, had fewer difficulties in adjusting to their child's cleft lip and palate.

The six individual indicators of adjustment to congenital disfigurement were explored for mothers in the Prenatal and No-Prenatal groups.

i Early Emotional Responses to the Baby's Condition: Duration of Overt Distress.

The most common emotion experienced by parents when given the diagnosis cleft lip and palate was one of shock. This was reported both by women who received the diagnosis at scan, and those who first learned of the cleft at the birth. For women in the latter group, the shock may have been all the more intense, given the unexpected nature of the cleft, and the likelihood that few people will have seen an unrepaired cleft lip and palate previously.

When I saw her I was devastated. I hadn't seen it before, I'd only seen repairs before.

It was just sort of, absolute shock, 'cos you always expect to produce a healthy, perfect child. You just don't expect anything will go wrong.

My reaction was one of total disbelief, because she looked so horrendous. I remember saying over and over again, 'It can't be true. This can't be happening'.

The sense of sadness and disappointment was strong for many women.

I was overwhelmingly upset. The tears came and it's hard to describe, like losing a member of the family, I was grief stricken.

I was just really, really upset because he wasn't perfect.

Whilst all parents experienced some element of shock, some parents also described feelings of denial.

It was very strange. I didn't focus on the mouth. He had a slight problem with his eye, nothing major, and that was the first thing I noticed. I don't know whether I was avoiding looking at his mouth.

A number of mothers reported experiencing a sense of helplessness:

There's this baby inside you with something wrong, and there's nothing you can do.

There was just this empty despair, 'Oh my God, what's going to happen?'.

Feelings of shame, guilt and anger also characterised many women's initial responses to their child:

I remember sitting there crying and saying, 'What have I done wrong?'.

All I could think was, 'What are people going to think?' and, 'Why me? What have I done to deserve this?'.

In a few cases, guilt was related not to a sense of responsibility for the cleft, but to feelings of revulsion on seeing the child:

I still feel guilty as my first reaction on seeing my baby, which should have been ' how lovely', was just sort of recoiling and 'how horrible'.

The scoring for this item focuses on the reported time it takes for parents to work through their distress and reach a sense of acceptance. For a considerable proportion of the sample, (20.5%, N = 9) this happened soon after the birth. The proportion of mothers with a prenatal diagnosis whose emotional responses followed this pattern, was four times that of those mothers who received the diagnosis of cleft at birth, although this tendency failed to reach statistical significance (35% vs. 8%; $\chi^2 = 5.10$, d.f. = 2, N.S.).

For the majority however, the experience of overt distress was a prolonged process. Approximately half of mothers in each participant group experienced distress and grieving for some weeks or months (Prenatal: N = 9, 45%; No-Prenatal: N = 13, 54%). A further 29 per cent of the overall sample (N = 13) experienced considerable distress related to the birth, at the time of interview (Prenatal: N = 4, 20%; No-Prenatal: N = 9, 37%).

ii. Social Exposure of the Child

Parents had to deal not only with their own responses to the disfigurement, but also with those of other people, both in the hospital, and when the baby was taken home. The importance of the baby's physical appearance was acknowledged by many women:

The face is probably the most important feature about you. It's all you ever see of a baby, isn't it, and people can be so harsh in their judgements. Everyone was incredibly kind, but there was just that feeling that you weren't showing off something, you know, perfect. It was a bit damaged.

Difficulties in coping with the responses of others were commonly reported:

People found it quite difficult knowing what to say, particularly when she was first born. And it was much easier for us when people acknowledged it, than when people said, 'hasn't she got beautiful hands', or, 'hasn't she got lovely hair', and you'd say, ' yes, but look at her face!'.

Preparing others for how their baby looked was a strategy used by many mothers, in order to avoid negative reactions:

I had to warn people before they came to see him. Um, and the reason I had to warn them was because I couldn't, I couldn't bear to see the look on their faces {mother starts to cry}. When they sort of, saw him without, without knowing, they'd sort of have a look of horror.

Parental apprehension about exposing their child to the gaze of others was not related to prenatal status ($\chi^2 = 0.63$, d.f. = 2, N.S.). In the entire sample, parent's responses were

distributed almost evenly across the three anxiety ratings. Seventeen parents (38%) reported that they were able to take their baby out and let others see their baby without much anxiety:

Yes, I did get some nasty looks. That's the upsetting bit. People are not very discrete, but I wouldn't hide her. At first you wonder what they're thinking. But then I s'pose you get past caring what they think..

Whilst 12 mothers (27%) were anxious about taking the baby out, and sometimes covered the cleft. A further 15 mothers (34%) could only take the baby out if the cleft was covered, or would not take the baby out at all. One mother, who covered her baby's lip with a plaster when they went out, described her concerns:

It's a dilemma. Do you let every one see her and not care and be proud of her, or do you not show her to every one? No matter what, people always do remember, and they do make judgements. You can't get away from it. It doesn't look nice and people don't react well to it.

iii. Taking Photographs

Almost half of the entire sample (N = 21) freely took and displayed photographs of their new baby prior to the repair of the cleft. For some, photographs acted as a 'medical record', documenting the changes that came with surgical repair. For others, photographs served to affirm the parent's acceptance of her child:

I thought, if I didn't have any pictures of her, she'll think I'm ashamed of her.

A similar proportion of mothers (N = 18, 41%) took photographs, but covered the cleft in the photo:

We've got a couple of interesting ones where the hand is covering it over, and on some we tried to take pictures of her without it showing, but they weren't terribly successful. It's such a big thing to hide as it's right in the middle of her face.

Alternatively, the photographs were kept out of sight. A small group of mothers (N = 5, 11.4%) did not take any photographs prior to surgery:

No, I didn't take any pictures. To me he was like an alien.

There were no differences in the taking of photographs, between the Prenatal and No-Prenatal groups ($\chi^2 = 0.77$, d.f. = 2, N.S.).

iv. The Effect of Surgery on Acceptance of the Child

Surgical repair of the child's cleft lip provoked a range of responses from mothers. A substantial number of parents (N = 9, 20%) found it hard to accept their child until the lip was repaired.

I couldn't bond with him until after the operation, when he started to look like a normal baby.

I didn't feel close to her until after the operation. I suppose she was so ugly before, I didn't cuddle her like you do with normal babies, until afterwards.

After surgery, it was like having a new baby. Having her returned from the recovery room was like delivering a newborn baby without a problem, to me.

However, for the majority (N = 33, 79%), surgery had little or no impact on how they felt about their child:

She did look different, but I felt the same. This was just how she was supposed to look , - she was finished off.

When she came back again she was all gunge and stitches, and I thought, "Why did they have to touch her, she looked fine the way she was', 'cos I'd got used to her little funny lip.

Many women also reported that the operation was fraught with anxiety, and proved a traumatic time:

That (surgery)was the most worrying part, 'cos he was so tiny. I was even trying to convince myself that he didn't even need an operation.

Again, time of diagnosis (prenatal vs. birth) was not associated with parental acceptance of the child before surgery ($\chi^2 = 0.44$, d.f. = 2, N.S.).

v. The Effect on Decisions about Having More Children

Parents were asked about the influence of their baby's condition on their views on having more children. Most parents expressed some degree of concern about the possibility of having another baby with a cleft. Almost half of the entire group (N = 21) reported anxiety about this risk, and indicated that they would like reassurance from prenatal screening, that future children did not have the condition.

I'd want to know whether another child might have the same. Not that it would stop me having another one, but I'd like to be prepared this time.

For over a third of parents (N = 15), the impact of having a child with cleft lip and palate was such that they had chosen not to take the risk of having a future child with cleft lip and palate.

There's a risk that another child might have it, and we wouldn't want to put another child through what we've had to put T through.

Whilst the remaining eight mothers reported that their child's cleft lip and palate did not effect their decision to have children in the future. Prenatal status was not associated with decisions regarding future children ($\chi^2 = 0.13$, d.f. = 2, N.S.).

vi. The Effect on the Parental Relationship

The birth of a child with cleft lip and palate imposed some degree of stress on the parental relationship, for a considerable proportion of the sample (N = 15, 34%). Strain not only arose as a result of difficulties relating to the diagnosis, to caretaking tasks, or

surgery, but may have also been associated with the lack of synchronicity found in process of adjustment experienced by the two parents. However, most parents in the sample reported that the experience of having a child with cleft lip and palate, had drawn the couple together in mutual support (N = 29, 66%). Prenatal status was not related to the impact of the birth of a child with cleft on the parental relationship (χ^2 = 1.82, d.f. = 2, N.S.).

Consequently, prenatal diagnosis of cleft lip and palate was not associated with individual emotional, cognitive and behavioural indicators of parental adjustment to the birth of a child with congenital disfigurement.

3. Current Psychological Health

Mother' psychological well-being was assessed using the GHQ-28 (Goldberg & Hillier, 1979). Within the entire sample, seventeen women obtained a score of 0 on this instrument, indicating that they experienced no psychiatric symptomatology (Prenatal group: N = 10, 50%; No-Prenatal group: N = 7, 29%). The mean total GHQ-28 scores were 2.9 (SD = 3.9) for the Prenatal group, and 5.1 (SD = 4.9) for women in the No-Prenatal group.

A third of the entire sample (N = 15) achieved a score that indicated psychiatric 'caseness', using the accepted threshold of 4/5. The figures in Table 15 show than the proportion of mothers in the No-Prenatal group who were experiencing psychiatric disturbance, was considerably greater than that of mothers with a prenatal diagnosis. However, this difference is not statistically significant ($\chi^2 = 1.34$, d.f. = 1, N.S.).

Table 14. Psychiatric Illness and Prenatal Status

	PRENATAL		NO-PRENATAL	
	N	%	N	%
Psychiatric Disturbance:				
Present	5	(25)	10	(42)
Absent	15	(75)	14	(58)
TOTAL	**20**		**24**	

The presence of psychiatric 'caseness' was not associated with demographic variables, or with the experience of pre- and postnatal stressors, with the exception of the family's reaction to the baby. There was a tendency for mothers whose families had responded in an unsupportive or rejecting manner, to experience a clinical level of psychiatric disturbance, with half of the mothers in this group reporting symptoms indicative of psychiatric disturbance (N = 8), compared with only a quarter of those mothers whose

families responded well to the baby (N = 7). However this trend failed to reach statistical significance (χ^2 = 2.8, d.f. = 1, p < .09). The presence of psychiatric 'caseness' was also not associated with receipt of information and emotional support following diagnosis of cleft.

Additionally, current psychiatric disturbance in mothers was not associated with social support in terms of the amount of support (t = 1.59, N.S.), or satisfaction (t = .61, N.S.). There was also no relationship between psychiatric disturbance and quality of the parental relationship (t = .24, N.S.).

The presence of four symptom groups was investigated in the Prenatal and No-Prenatal groups. The majority of mothers in both study groups did not report any psychiatric symptoms. Consequently, the distributions for each of the symptom groups are positively skewed. Non-parametric tests (Mann-Whitney) have therefore been computed in order to examine the relationship between symptom group and prenatal status. The means and standard deviations of the symptom scores found in the Prenatal and No-Prenatal groups are outlined in Table 16, in addition to the numbers of mothers who reported one or more of the symptoms in each symptom grouping. Mann Whitney U values are also presented.

Table 15. Psychiatric Symptom Group and Prenatal Status

	PRENATAL			NO-PRENATAL			
	N	M	SD	N	M	SD	U
Symptom Group:							
Somatic	8	1.2	1.7	14	1.7	1.8	193.0
Anxiety	8	0.8	1.3	12	1.5	2.1	205.0
Social Dysfunction	7	0.8	1.6	10	1.3	2.1	216.5
Severe Depression	0	-	-	6	0.37	0.8	180.0 *

* p < .05

No relationship was found between prenatal status and current experience of symptoms indicative of somatic symptoms, anxiety, or social dysfunction. However, prenatal status was related to severe depression. All six mothers who reported symptoms indicative of severe depression were in the No-Prenatal group. Thus mothers who had not received a prenatal diagnosis of cleft lip and palate, experienced significantly more symptoms of depression, compared with mothers who had a prenatal diagnosis.

4. The Relationships between Measures of Adjustment

The relationships between mothers scores on each of the three measures relating to adjustment, were explored. Firstly, this analysis showed a tendency for ratings of resolution on the RDI to be associated with scores on the GHQ-28. Thus, mothers who were unresolved with respect to their reaction to the diagnosis, were more likely to experience symptoms indicative of psychiatric disturbance (Unresolved: N = 13, 42%; Resolved: N = 2, 15%). However, this trend did not reach statistical significance (χ^2 = 2.8, d.f. = 1, $p < .09$). Resolution was not significantly related to either of the four psychiatric symptom groups also measured by the GHQ-28, although it is notable that of the six women who reported symptoms of severe depression, four were unresolved with respect to grief or trauma associated with the diagnosis.

Neither resolution of grief and trauma (RDI), or psychiatric disturbance (GHQ-28), were related to adjustment to congenital disfigurement, as measured by the IPACD (t = 1.7, N.S.; t = 0.6, N.S.; respectively). Similarly, a series of correlational analyses showed that adjustment as assessed by the IPACD, was not related to either of the four psychiatric symptom groups (somatic symptoms: r = -0.11, N.S.; anxiety: r = 0.05, N.S.; social dysfunction: r = -0.04, N.S.; severe depression: r = 0.09, N.S.)

Furthermore, no there were significant relationships found between resolution of trauma, or psychiatric disturbance, and the six individual IPACD indicators of adjustment.

5. Predictors of Parental Adjustment

Regression analyses were computed in order to investigate possible predictors of adjustment as measured by the RDI, IPACD and GHQ-28. On the basis of results from the inferential analyses described earlier, the following variables were identified as associated with the three measures of adjustment:

* prenatal diagnosis of cleft;
* medical problems during pregnancy;
* family response to the birth of the child;

The predictive value of each of these variables was explored for the three adjustment variables, using logistic and linear regression methods.

i. Resolution of Grief and Trauma (RDI)

Overall, prenatal diagnosis, medical problems in pregnancy and family response were not predictive of resolution of grief and trauma (χ^2 = 6.0, d.f. = 3, N.S.). However, the effect of prenatal diagnosis of cleft was significant (B = 1.4 , Wald = 3.6 . d.f. = 1, p =.05).

ii. Psychiatric Disturbance (GHQ-28)

Overall, prenatal diagnosis, problems in pregnancy and family response were not predictive of psychiatric disturbance (R^2 = 0.16; F = 2.6, N.S.). However, the effect of family response was significant (B= -2.84, t = 2.08, p <.05).

iii. Adjustment to Congenital Disfigurement (IPACD)

The predictive power of family response, problems during pregnancy and prenatal diagnosis together was significant (R^2 = 0.26; F = 4.58, p <.01.). Furthermore, family response and problems in pregnancy were both independently predictive of parental adjustment to congenital disfigurement (B = -1.34, t = 1.96, p <.05; B = 1.78, t = 2.45, p <.05; respectively).

6. Mothers' Attitudes Towards Prenatal Diagnosis

Finally, mothers were asked about their attitude to prenatal detection of cleft lip and palate, and whether they thought it was helpful to know about the cleft before the birth. The overwhelming majority of women responded in favour of prenatal diagnosis of cleft (N = 36, 81%). This group included women who felt their own experience of prenatal diagnosis had been positive:

It was so much better for us to know then, and to be able to tell our friends, than for everybody to have to find out about it, and get used to the idea at the same time as we were. So that when he was born, um, it wasn't such a shock for us and they weren't stuck for something to say at the time, because they all knew about it. It just made it so much easier when he was born.

Before she was born, all the visits to the doctors and the literature I'd read, it was all a bit unreal. But the minute she was born, when it became reality, suddenly it was normal and part of life. I think all the preparation for me beforehand made it so much easier to deal with.

And also those women who had wished that their baby's cleft had been diagnosed at scan:

I had quite a late scan and they said there was no problem. But I wonder, if only I'd known. If only I'd had some preparation, it would have been easier, I think.

Now, when I look back, it's the shock of first seeing her and that first week that haunts me. So, I think it might have been better to know in advance.

Ten of the women in favour of prenatal diagnosis, also voiced some ambivalence about the procedure:

I don't know which, I think it's got to be six of one and half a dozen of another. I think in terms of making the birth a positive experience, knowing beforehand, when you actually get the child, it has got to be better. But that time between finding out and then finding

out the real implications of it all and actually having the child, is dreadful. But all in all, I think it's best to know beforehand.

Only eight women (18%) had voiced misgivings about prenatal diagnosis:

I'm glad we didn't know, because I'd have imagined the worst, and found that last twenty weeks of pregnancy really bad. And I'd have worried that other things were wrong with her. Getting through labour would've been really terrifying. I'd have been terrified to look at her.

Women's views on prenatal diagnosis appeared to be influenced by personal experience. All of the women who had previously had a prenatal diagnosis, were in support of diagnosis during pregnancy, whilst, the small group of women who were not in favour of prenatal diagnosis, had not actually experienced it themselves. This observed relationship between past experience and current views of prenatal diagnosis was significant ($\chi^2 = 8.32$ d.f. = 2, $p < .05$).

DISCUSSION

Overview

The primary aim of this study was to investigate the impact of prenatal diagnosis of cleft lip and palate, on parental adjustment to the birth of a child with cleft. This discussion examines the findings in the light of the study's aims, and the methods used. The limitations and strengths of the study are then examined. This is followed by a consideration of the findings within the wider theoretical context, and possible directions for future research are proposed. Finally, the implications of the results for professional practice are explored.

The Main Findings

The study aimed to explore the relationship between prenatal diagnosis and parental adjustment to the birth of a child with cleft lip and palate. Parental adjustment was assessed in terms of three psychological constructs:

(i). resolution of trauma associated with the diagnosis of cleft lip and palate (RDI);

(ii). parental adjustment to congenital disfigurement (IPACD);

(iii). current psychological health (GHQ-28).

The main findings concerning the impact of prenatal diagnosis and other key independent variables central to the study's hypotheses, on parental adjustment as measured by each of these three constructs, are considered here.

Parental Adjustment to the Birth of a Child Cleft Lip and Palate

The results of this study suggest that the majority of parents of children with cleft lip and palate experience adjustment difficulties. Furthermore, when compared with rates of adjustment difficulties observed in other populations, the rates found in this study were higher than anticipated. Yet, it is also the case that a substantial number of mothers of children with cleft appear to be well-adjusted. This suggests that, contrary to the prevailing view, negative outcome is not inevitable for parents of children with cleft lip and palate.

In order to understand parents' experience of adaptation to their child, it seems important to establish why higher rates of problematic adjustment may have occurred for mothers in this study.

Firstly, the rates of resolution classifications found in this sample, as assessed by the RDI, were compared with those in previous studies. Pianta & Marvin (1993b, 1993c, 1996) reported rates of resolution ranging from 42 to 54 per cent, for several populations of mothers of children with cerebral palsy. In the present study, the proportion of mothers with prenatal diagnosis who were classified as resolved, is comparable with these rates. Yet, a considerably lower rate of resolution (17%) was found in the group of women who did not receive prenatal diagnosis. The disparity observed here could possibly be understood in terms of differences between the two disabling conditions.

For example, there are certain difficulties specific to cleft lip and palate such as the highly visible and often shocking nature of the disfigurement, and early feeding problems, which may impede resolution. Also relevant, may be the transitional nature of the first few years of life of the child with cleft, due to the many operations that occur. During this period, parents are thus generally looking towards the 'next operation', which promises improvement for their child. This level of instability, which may last until the main surgical repairs have been carried out, may also hinder the process of resolution. Pianta & Marvin (1992) illustrate this notion with the hypothesis that, although potentially devastating, some conditions such as cerebral palsy, are 'static', and may therefore be more likely to lead to resolution, than volatile and unstable conditions such as epilepsy. Consequently, there appear to be a number of difficulties specific to the condition of cleft lip and palate, which may impede parental resolution, and thus lead to the lower rate found in the cleft sample.

Of particular interest is the similarity found between rates of resolution in parents who have prenatal knowledge of the diagnosis, and rates found in previous population. Potential explanations for this finding are discussed later.

The rate of poor adjustment to congenital disfigurement (IPACD) observed for the majority of mothers, irrespective of prenatal status, was also higher than rates found in previous research, where adjustment for 50 per cent of the sample was rated as poor (Bradbury & Hewison, 1994). This compares with poor adjustment found for 66 per cent of mothers in the present study, when the threshold score was used (Bradbury & Hewison, 1994). This discrepancy may possibly be related to differences observed between the two samples. The previous sample studied by Bradbury & Hewison (1994) was smaller, and included children up to 11 years of age. Items on the IPACD rely on parents' recall of events and the behaviour of parents and significant others, following diagnosis of cleft. One could hypothesise that in the Bradbury & Hewison (1994) study, the accounts of parents of older children may have been subject to recall bias, whereby fewer difficulties following diagnosis were remembered.

The final indicator of parental adjustment was current psychiatric disturbance (GHQ-28). The level of psychiatric difficulties experienced mothers of children with cleft, was no greater than expected, as the proportion of women who experienced psychiatric disturbance using the GHQ-28 matched rates found previously in samples of general practice attenders (Goldberg & Hillier, 1979).

However, the higher rates of adaptation difficulties, indicated by lack of resolution and poor adjustment to congenital disfigurement, do suggest that parental adjustment to children with cleft lip and palate may be more problematic than adjustment in other areas of disability. Further research on this issue is required before any firm conclusions can be drawn.

Internal versus External Adjustment

Adjustment to congenital disfigurement (IPACD), was not related to resolution of trauma (RDI), or to psychological well-being (GHQ-28). However, a non-significant relationship was found between lack of resolution of trauma and grief, and the presence of psychiatric disturbance. This suggests that the three measures of parental adjustment to the birth of a child with cleft lip and palate, may assess different aspects of

adjustment. This finding gives rise to the tentative hypothesis that two distinct components of adjustment may be measured in this study:

* 'internal' adjustment, reflecting psychological factors including resolution of grief and depression, assessed by the RDI and GHQ-28;

* 'external' adjustment, which relates to parents' social and behavioural coping responses. Particular emphasis is placed on the way in which the mother responds to the cleft and her child in relation to the world around her. This is measured by the IPACD.

On the basis of this dichotomy, a mother may have adjusted well in terms of overt, external coping, but may not necessarily have resolved the grief that she has experienced. In theory, the lack of resolution observed in this example would suggest the possibility of poor outcome for the mother and child (Pianta & Marvin, 1992). Yet, it may be also possible that external adjustment is in itself sufficient for positive outcome.

The possibility nevertheless remains that this distinction may have emerged as a result of the measures used, rather than from any conceptual basis. For example, the collection of items in the IPACD have not yet been demonstrated as valid or reliable, and it is possible that they do not reflect a single construct relating to adjustment. Therefore the conclusions drawn from this finding must be treated with caution.

Prenatal Diagnosis and Adjustment

The potential impact of prenatal diagnosis on parental adjustment to the birth of a child with cleft, is an issue central to this investigation. As hypothesised, the results indicate that prenatal diagnosis does have a positive impact on parental adjustment. However, prenatal detection of cleft is not the only variable predictive of successful adjustment. Other factors related to the perinatal period also impact on adjustment, namely, medical or physical problems during pregnancy, and initial family response to the child.

Prenatal detection was associated with the resolution of trauma and grief related to the diagnosis, with those mothers who had prenatal knowledge of their baby's cleft, more likely to have resolved their grief. The absence of prenatal detection was also associated

with psychiatric disturbance (i.e., severe depression), however, numbers were too small for inferences to be drawn with any confidence from this finding.

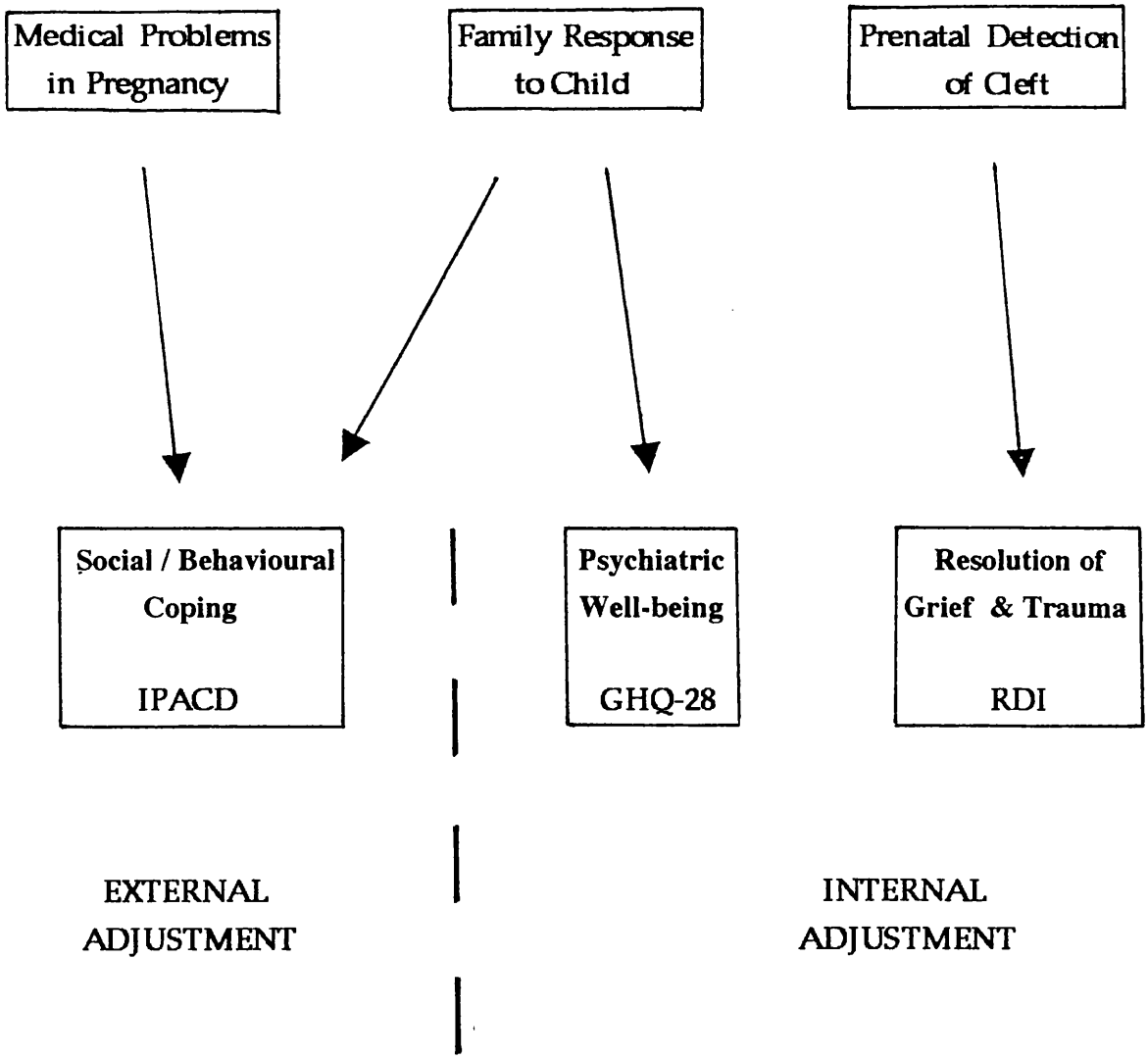
Mothers' reported experience of medical problems during pregnancy, was found to be predictive of problematic adjustment to congenital disfigurement, as measured by the IPACD. Furthermore, initial negative family response to the birth of the child was also found to be predictive of poor parental adjustment to congenital disfigurement. A trend was also found between initial family response and psychiatric disturbance. Mothers whose families responded in an unsupportive or rejecting manner were more likely to experience symptoms indicative of psychiatric disturbance.

Notably, these variables appear to relate to the different aspects of adjustment that have been hypothesised here. Potential pathways that have emerged in the results between perinatal factors and the different components of adjustment are presented in Figure 1.

As we have seen, the results demonstrate that prenatal detection may have a positive impact on parental resolution, which may be related to an internal component of adjustment. Meanwhile, mothers' reported experience of medical problems during pregnancy has been found to lead to difficulties in overt behavioural / social coping strategies, and therefore to external adjustment. Of particular significance is the role of family response to the birth of a child with cleft, in both external and internal adjustment processes. Findings suggest that rejecting and unsupportive family reactions may lead to poor coping, in addition to psychiatric disturbance.

Figure 1.

Predictors of Parental Adjustment to the Birth of a Child with Cleft Lip and Palate



Within the narrow constraints of this study, one can only hypothesise about the causal factors or processes underlying these relationships. Firstly, in the case of prenatal diagnosis and resolution, it is possible that prenatal diagnosis may allow the expectant mother time to prepare for the birth of a child with cleft lip and palate, by accessing specialist feeding equipment, alerting family and friends, finding information about the appearance, nature and treatment of cleft, and establishing links with the surgical team. The opportunity for cognitive and emotional preparation may also be a crucial feature of prenatal diagnosis. Preparation techniques such as cognitive rehearsal, realistic fear appraisal and planning a variety of coping strategies (Sherr, 1995) may be employed during the pregnancy. Prenatal knowledge about the baby's cleft may also provide the expectant mother with the opportunity to begin to reconcile the image or fantasy she has developed during pregnancy of her 'ideal' or 'perfect' baby, with the real, 'imperfect' baby, before the actual birth. This cognitive and emotional process is thought to be crucial in successful transition to parenthood (Beckendorf, 1984). Pianta & Marvin (1993c) also describe this process in terms of integration of the information that pairs the child with the disability, and the accompanying emotions, into representational systems that the parent has of themselves as parents, of their child, and of their relationship with their child. It is this process which is considered central to resolution.

For any parent, the birth and early postnatal period can be an emotionally overwhelming time. However, when a baby with cleft is born, the intensity of the parent's emotional response to cleft, combined with the enormity of the task of parenting a new baby, the demands specific to caring for a baby with cleft, and the extensive amount of new information that rapidly needs to be absorbed, may serve to impede the parent's attempts at this process of integration. One mother described the overwhelming shock she experienced at the birth, when she discovered her baby had cleft. For this mother, adjusting to having a child with cleft has been problematic.

M (partner) said he had something wrong with his lip and a hole in his mouth. I just couldn't come to terms with it. I was so shocked, I think with having the caesarian early, it was just one big shock.... I was so upset and you know, I just couldn't bond with him at all. I just thought, what have I been given, 'cause he was so awful to look at. I was really upset, I didn't want any pictures taken of him. I thought why did it happen to me, you know, 'what have I done to deserve this ?'.....It's been very hard. I'm coming to

terms with it, but I don't think I'll ever come to terms with it completely. I get really bad days when I do get upset. And going for operations, well, that's another nightmare.

Consequently, by providing an opportunity for practical, cognitive and emotional preparation, prenatal diagnosis may facilitate successful resolution of grief and trauma associated with the diagnosis.

The link between negative family reaction to the child and psychiatric disturbance may be indicative of chronic difficulties that existed prior to the birth. However, the possibility that lack of support and criticism from families, at a time when parents are in need of a supportive environment, might lead mothers to experience symptoms indicative of psychiatric distress, is also very real. Similarly, the absence of a supportive family may also exacerbate difficulty in social and behavioural aspects of adjustment. Finally, the relationship between medical problems in pregnancy and later external adjustment, may potentially be understood in terms of the impact of a culmination of difficulties. Not only does the mother experience medical or physical problems whilst she is pregnant, but she also gives birth to a child with cleft lip and palate. One could thus hypothesise that the stress associated with this heightened level of problems, makes it more difficult for the mother to cope with the day to day realities of parenting a child with a cleft lip and palate.

It must be stressed that the hypotheses and model proposed here are extremely tentative. However, it would seem that the preliminary patterns that have emerged, may possibly contribute to a clearer understanding parental adjustment to the birth of a child with cleft lip and palate.

A number of other interesting findings relating to the experience of parents of children with cleft lip and palate, have emerged in this study.

Social support and Quality of the Parental Relationship

Contrary to expectations, current social support was not associated with resolution of trauma, parental adjustment to congenital disfigurement, or psychiatric disturbance. Furthermore, social support did not have a moderating effect on the relationship between

prenatal status and resolution. In fact, parents in this study did not experience specific difficulties with social support. Ratings of social support were higher than those found in previous investigations into the social support of parents of children with craniofacial abnormalities using the same measure, but were comparable with ratings found in a sample of parents of children with no significant physical or behavioural problems (Benson *et al.*, 1991). One explanation for higher levels of social support than expected, may be the bias towards upper and middle social class found in the sample. Benson *et al.* (1991) suggests that higher socio-economic status may be a protective factor against lack of social support.

There was evidence to suggest a relationship between the quality of the of the mothers relationship with her partner, and external indicators of adjustment. Mothers who reported discordant relationships were more likely to experience difficulties in social / behavioural adjustment to their child's cleft. Generally, low rates of marital discord were found in the sample. This is inconsistent with findings from other studies which report high levels of discord in parents of children with clefts (e.g., Speltz *et al.*, 1990). The quality of the parent's relationship in this study, was found to match ratings for other populations of parents of children without congenital abnormalities or other problems (Heyman, Sayers & Bellack, 1994). This unexpected finding requires further investigation. The potential interactive effect of the quality of the parental relationship on the association between perinatal factors and 'external' adjustment, also merits further research.

Parental Adjustment and Support Following Diagnosis of Cleft

Mothers who received the diagnosis of their child's cleft at the birth, were more likely to receive initial information about cleft from a health professional such as a midwife or paediatrician, who may have possessed limited knowledge about the condition. This first information may therefore have been less adequate than that given to women who received the diagnosis of cleft during pregnancy, and who were more likely to have had contact with a health professional with specialist knowledge of cleft soon after the diagnosis. Yet, this difference did not have a statistically significant impact on mothers' later adjustment.

Parental Adjustment and Severity of Disfigurement

Resolution, adjustment to congenital disfigurement as measured by the IPACD, and mother's psychological health, were not associated with the severity of the child's cleft lip and palate. This is consistent with research findings of Bradbury & Hewison (1994), and MacGregor (1992), and also with Pianta and Marvin's (1993c) finding that caregiver's resolution status was independent of the severity of the child's cerebral palsy.

Prenatal Status and Experience of Pregnancy and Birth

On the whole, the diagnosis of cleft lip and palate at prenatal screening was considered by mothers to have a negative impact on their experience of pregnancy. However, this appears to have had few repercussions on the pregnancy in terms of medical problems. Mothers with a prenatal diagnosis had comparable levels of medical problems during pregnancy as those mothers with the diagnosis at birth.

Termination was offered to mothers on detection of the cleft in a fifth of the cases, and in a further case, a mother's request for a termination was refused. Information is not systematically collected on numbers of women offered a termination when cleft is detected prenatally. Therefore, it is not possible to assess the representativeness of the number of terminations offered to women in this study.

Family history of cleft lip and palate was not related to requests for specific screening of the cleft by parents at scan. This was in part due to mother's ignorance about the genetic component of the condition, and to mothers not being fully aware of their relative having a cleft at the time. Remarkably, in two cases, parents with clefts failed to acknowledge the strong possibility that cleft may be passed on to their child, during the pregnancy, and therefore did not ask for screening. It is difficult to know whether this response involved some element of denial on the part of the parents.

The negative impact of diagnosis on pregnancy did not appear to influence the actual birth. In fact, the reverse proved to be the case, as there was a non-significant tendency for women who did not have a prenatal diagnosis, to perceive the birth as more difficult, and also to report more medical problems. This may possibly reflect greater shock and disappointment experienced by women who were not aware of their child's cleft before the birth and who were therefore not prepared for their child's disfigurement. The

intensity of these feelings may then have negatively influenced the mother's view of the birth.

Maternal Attitudes to Prenatal Diagnosis of Cleft Lip and Palate

Finally, mothers' views on the value of prenatal diagnosis of cleft were explored. In general, mothers supported the use of prenatal screening for cleft lip and palate, although for some women, there were mixed feelings. Attitudes towards the value of prenatal diagnosis tended to be related to past experience. All of the mothers who voiced strong misgivings about prenatal diagnosis, had in fact not had a prenatal diagnosis themselves. Consistent with the belief, 'what is must be best', observed by Porter & Macintyre (1984) in their investigation of women's views on antenatal care, the trend observed here reflects the principle of cognitive consistency: the need to redefine the world in ways that make sense (Festinger, 1957). Thus, women's beliefs are consistent with their own experience. Those women with the diagnosis are more likely to consider it worthwhile, whereas the reverse may be true for women who have not experienced the diagnosis.

At this point, some consideration should be given to the strength and significance of the findings. One advantage of this study was the homogeneity of the two participant groups. The descriptive analyses demonstrated an extremely high level of concordance between the Prenatal and No-Prenatal participant groups, in terms of demographic variables, and independent variables including, number of operations, feeding difficulties, social support and quality of the parental relationship. Potentially confounding variables were thus controlled for, thereby reducing potential error in the analyses, and increasing statistical power.

However, a large number of statistical tests were computed, and with significance set at the conventional criterion of .05, caution must be taken when interpreting the results. One could argue nevertheless, that the large number of statistical tests reflected a conservative approach to the analysis of the data, as the majority of these tests served as a check for the presence of confounding variables, and were not part of the inferential analyses that related to the study's hypotheses.

Limitations of the Present Study

Having assessed the strength and significance of the findings, it is important to examine the extent to which they can be generalised beyond the immediate context of the study. Consideration is therefore given to methodological limitations relating to the design and measures used.

Although the retrospective design of this study has provided an efficient means of collecting data, it has also led to a number of significant methodological difficulties. The retrospective study of a psychological construct such as adjustment is particularly problematic. The process of adjustment is dynamic rather than linear, whereby a family's functioning may be considered to fluctuate over time, with highs and lows that presumably relate to stressors impinging on the family system. As a result, measures of adjustment related to current functioning should ideally be considered relative to pre-diagnosis status, rather than in absolute terms. As Kazak (1989) argues, marital functioning prior to the birth of a child with a disability may well be the best predictor of long-term marital adjustment. Similarly, it may be that those parents of children with clefts who report psychiatric disturbance, may be those who experienced psychological problems before the birth. Exceptions to this criticism are resolution of loss or trauma, and indicators of parental adjustment to congenital disfigurement, both of which are directly related to the diagnosis or birth of a child with a disability, and thus cannot be measured prior to diagnosis.

There are a number of other limitations associated with the retrospective nature of the study. Firstly, reliance on mothers' memory for much of the data may have led to the introduction of error as a result of inaccurate recall. The use of retrospective accounts also negated access to reliable, in-depth information on, for example, the impact of prenatal diagnosis on mothers' psychological health during pregnancy. Nor was it possible to establish whether the levels of anxiety experienced by pregnant mothers following prenatal diagnosis, were in fact any different from that experienced during pregnancy by mothers without the prenatal diagnosis. There was also no reliable information available on the quality or timing of support or information given to mothers following diagnosis. Also, the impact of the way in which women were informed of the diagnosis on later adjustment, could not be investigated. Ideally, a longitudinal approach to the study of parental adjustment to the birth of a child with cleft

lip and palate would have overcome these limitations. Unfortunately, given the relatively narrow remit of this study, a prospective design was not possible.

Yet, these limitations should not undermine the validity of the perceptions of the mothers in this study. Subjective reality for these mothers may be particularly relevant to adjustment and to the way in which they cope with adversity. Consequently, although the retrospective nature of this research necessitates some caution when interpreting the results, it would be inappropriate to suggest that this invalidates the findings presented here.

A further criticism of the study is that the experience of fathers has been neglected, as is often the case in psychological research, and particularly in the area of cleft lip and palate. However, as the major burden of care of the child tends to fall on the mother (as borne out by this study), and given the constraints of time and resources, maternal adjustment to the birth of a child with cleft lip and palate was given greater priority.

The researcher was not blind to the prenatal status of mothers from two of the four units. However, as there were no differences in adjustment ratings found between the four units, it seems that this knowledge did not bias results. A further important limitation of the study is the relatively small sample size. Although the acceptance rate was high, there were a number of criteria that limited the target population. The relative rarity of cleft lip *and* palate, and low prenatal detection rates of cleft, in addition to the narrow age range specified by the study, all contributed to the small sample size, and also resulted in a reliance on 'convenience sampling', whereby whoever met the inclusion criteria was asked to participate.

In an attempt to increase sample size, without broadening the inclusion criteria to incorporate other forms of cleft, participants were recruited from four craniofacial units. The inclusion of participants from different units could be viewed as a strength of the study, in allowing a wider range of services to be examined, thus increasing the generalisability of the results. However, recruitment from four units also served to increase the variability of treatment received by participants and their children. Specific differences did exist between the treatment and care provided by the four units, primarily with regard to the timing of operations. However, this variability did not impact on maternal adjustment.

While these limitations may restrict the validity of the results to some extent, the lack of variability between the Prenatal and No-Prenatal groups with respect to many potentially confounding demographic and background variables, should mean that the differences observed in resolution of trauma between the two groups, are reasonably robust.

Finally, there are a number of limitations related specifically to the measures used in the study. As a measure of parental adjustment to the birth of a child with cleft lip and palate, the IPACD (Bradbury & Hewison, 1994) requires further work on establishing psychometric properties. It is an extremely useful descriptive measure, however there is little evidence to support the validity and reliability of this scale. The scoring categories are crude, and the designated threshold is arbitrary. The IPACD is also reliant on retrospective self-report, and therefore the reliability of information elicited may be questionable. As the IPACD did not correlate with the Reaction to Diagnosis Interview (Pianta & Marvin, 1992), or the GHQ-28 (Goldberg & Hillier, 1979), and would seem to assess 'external' rather than 'internal', psychological elements of adjustment, it is not possible to comment on the construct validity of this measure. However, in spite of the psychometric limitations outlined here, the IPACD provides a wealth of descriptive information that relates specifically to the difficulties faced by parents of a child with a disfigurement, and may also be extremely valuable in a clinical context. Further evaluation of the content and construct validity of this measure is required, however, it is hoped that the results of this study have contributed further to an understanding of this measure.

Whilst the GHQ-28 (Goldberg & Hillier, 1979) is considered a valid and reliable method of screening for general psychological and psychiatric disorder, the design of this instrument has also met with criticism. The scale measures acute rather than chronic conditions. The response categories ask whether each symptom is worse than usual, and therefore, if a person has suffered a symptom for a long time and has come to consider it 'usual', the scale will not identify it as a problem. Thus changes in a person's condition rather than the absolute level of the problem are measured in this study. This introduces some doubt onto the reliability of mothers' responses on this instrument, however Goldberg & Williams (1988) argue that the loss of cases is minimal because many people hold the concept of their 'usual self' as being without symptoms.

A final limitation relates to the inter-rater reliability for the RDI. Previous research using this measure reported inter-rater agreement for Resolved / Unresolved

classifications as 92% and 94% (Pianta & Marvin, 1993b, 1993c). This compares well with the percentage agreement found in this study (83.3%). However, the kappa statistic for inter-rater reliability for the RDI ratings was adequate, if only moderately acceptable. Pianta & Marvin (1993b, 1993c) have not published a kappa statistic for inter-rater reliability, and so a comparison can not be made. However, one reason for the low reliability statistic observed for inter-rater reliability, may be related to the use of transcripts rather than video recordings of interviews, as was used in previous research.

Whilst these limitations must be considered when drawing conclusions from the results of this study, the methodological strengths of this study should also be borne in mind. The use of established, valid and reliable measures such as GHQ-28 and RDI, has served to enhance the strength of the results. Moreover, where possible, a combination of quantitative and to some extent qualitative self-report methods have been used. Rather than a total reliance on questionnaires, much of the information comes from face to face interviews carried out in parents homes. It is hoped that this approach would increase the quality and reliability of the information elicited, in order to get a more accurate and in-depth understanding of mothers' experience following the birth of a child with cleft lip and palate.

Theoretical Implications of the Research

This is a new area of research. The adjustment of mothers with a prenatal diagnosis of cleft lip and palate has not previously been studied, nor has the concept of parental resolution of grief and trauma associated with the diagnosis of disability, been applied to this population. A number of interesting issues, salient to the areas of both cleft lip and palate, and prenatal screening, have emerged from the findings.

The results from the present study suggest that factors relating to mothers' perinatal experience, may be associated with parental adaptation to the birth of a child with cleft lip and palate. In particular, prenatal diagnosis of cleft appears to be related to parental resolution of the grief and trauma associated with the diagnosis. This is consistent with the view that prenatal diagnosis of an abnormality can allow the mother time to prepare

for the birth, as suggested primarily by anecdotal evidence from mothers (Farrant, 1985). It therefore seems that this opportunity to prepare, may facilitate parents' ability to integrate the information about their child's diagnosis into their mental models of the child and of themselves as parents. It is this process that enables the parent to resolve their own reaction to the diagnosis more successfully (Pianta & Marvin, 1992).

However, it is important to consider these findings within the broader context of the literature on prenatal diagnosis of congenital abnormality. Potential drawbacks of prenatal diagnosis of cleft lip and palate have been well-documented. Concerns in the literature have focused on the possibility that a prenatal diagnosis of less serious congenital abnormalities such as cleft lip and palate, may provoke anxiety (e.g., Farrant, 1985), and have more fundamental effects on the experience of pregnancy itself (e.g., Rothman, 1986; Richards, 1989). This concern was shared by some of the mothers in this study:

I wouldn't want to have a scan, because it would change your feelings about the pregnancy and giving birth. I'd worry like mad.

Personally, I wouldn't want a scan. I think it would set a precedence before the baby's born that this is a problem baby, and the worry and anxiety would be too much to cope with.

Although it was not possible to directly assess psychological disturbance of these women during pregnancy, the majority of mothers for whom the cleft was detected prenatally, reported that the diagnosis had negatively affected their experience of pregnancy, as a result of the distress and anxiety provoked by the news that their baby had cleft lip and palate. However, few of these women went on to experience difficulties during the birth, nor were they more likely than women without the prenatal diagnosis, to experience psychological adjustment difficulties, such as lack of resolution or depression. Consequently, although prenatal diagnosis may lead to heightened anxiety and distress during the final trimester of pregnancy, this does not appear to have a significant impact on later adjustment.

A second issue that has emerged in the literature with regard to prenatal detection of cleft lip and palate, is the increased possibility that women will choose to terminate the pregnancy as a result of the cleft. This is currently the subject of much debate, as some

health professionals have questioned the justification for the termination of a baby with a cleft, on the grounds that cleft is a relatively minor abnormality which can be surgically repaired (e.g., Bronshtein, Blumenfeld & Blumenfeld, 1996; Butler & Sommerlad, 1997). The present study investigates the experience of women who had a prenatal diagnosis of cleft lip and palate, and who continued with the pregnancy. Significantly, the majority of women were not offered a termination on detection of the cleft. The experience of mothers in this study therefore suggests that on the whole, termination is not presented as an option when cleft lip and palate is detected at prenatal screening.

Yet, little is known of those women who had a prenatal diagnosis of cleft lip and palate, and went on to terminate the pregnancy. Rates of terminations for cleft lip with / or without cleft palate in the North West Thames region, are documented as 18 per cent in 1994, and 16 per cent in 1995 (North Thames (West) Malformation Register, 1996). However, these figures are confounded by the inclusion of clefts associated with other malformations such as chromosomal syndromes. It is therefore impossible to determine the exact rate of terminations for isolated cleft lip and palate, and whether this rate has seen an increase in correspondence with growing detection rates.

In addition to the role of prenatal diagnosis in adjustment to the birth of a child with cleft, there are number of noteworthy findings which require further consideration in relation to the relevant literature. Firstly, maternal adjustment was not associated with the severity of the child's cleft lip and palate. This result supports findings elsewhere in the literature (Bradbury & Hewison, 1994; MacGregor, 1992), and in doing so challenges the view that the degree of distress experienced by parents does has a linear relationship with the degree of disfigurement.

Surprisingly, social support appeared to have had little impact on parental adjustment. This is inconsistent with the literature on the functioning of families with a disabled child (e.g., Sloper, Knussen, Turner & Cunningham, 1991), which highlights the role of social support in how parents adapt to their child. The adequate levels of social support reported in this study are also inconsistent with the accepted view that parents of children with cleft lip and palate are socially isolated and unsupported (e.g., Benson, *et al.*, 1991). However, a supportive parental relationship, in addition to perceived family reaction following the birth appears to be related to parental adjustment in the present study. This is consistent with previous work on the importance of the marital dyad in

family functioning (Benson & Gross, 1989), and also with research by Bradbury & Hewison (1994) which highlights the role of transgenerational issues in parental adjustment. Emphasis has also been placed on the importance of support offered by grandparents to parents, following the birth of a disabled child (Beresford, 1994).

Issues relating to the assessment of resolution of loss or trauma also merit some consideration here. The Reaction to Diagnosis Interview was designed for research use with parents of children with some form of disability or chronic illness. Previously used with parents of children with cerebral palsy, epilepsy and learning disabilities (Pianta & Marvin, 1993b, 1993c, 1996), the RDI has never previously been used with mothers of children with cleft lip and palate. Findings from the present study demonstrate that the concepts of loss and trauma associated with the birth of a child with a disability, and the need for mothers to resolve this loss, are valid and meaningful in relation to the experience of parents with children with cleft lip and palate.

Resolution of trauma was also found to be unrelated to time since diagnosis. Consistent with previous research (Pianta & Marvin, 1993b), this finding supports the argument for a process rather than a stage theory of mourning and resolution. Data from interviews contradicted the idea proposed by some 'stage' theories (e.g., Blacher, 1984), that there is a state of complete resolution where the experience of loss has been integrated to the extent that it is no longer of much concern to the parent. Parents in the present study reported an ongoing effort to remain focused on the needs of the child, whilst coping with feelings of loss or sadness:

I think (my feelings) still go up and down really. I've had all the information I could possibly ask for and I think I've just sort of got on with it really, you know. The operations have been the hardest bit. And I mean there are still tears about it, and there's still worry about it, but now his feeding's sorted out, and basically, between operations it's been fine.

I don't feel angry anymore. And I don't feel sorry for myself anymore. But I do feel different from other mothers, and I feel sad for him. And I just feel exhausted, because of all the problems, with his hearing and speech.

The need for parents to compensate for their child's disability has been emphasised as important for the development of secure attachment relationships (Cox & Lambrenos,

1992). Previous research has also suggested that failure to resolve the grief and trauma associated with the diagnosis of disability may negatively influence the mother's capacity to adapt and respond sensitively to her child's cues, and may therefore lead to an elevated risk of attachment difficulties (Pianta & Marvin, 1993c, 1996). This gives rise to the hypothesis that, for those mothers of children with cleft who are unresolved with respect to the trauma of the diagnosis, there is an increased risk of an insecure attachment relationship with their child.

The manner in which a diagnosis of congenital abnormality is given, may also have a significant impact on parents, and is therefore relevant to this discussion. There is evidence to suggest that parents generally consider the news that their child has cleft lip and palate to be traumatic (Bradbury & Hewison, 1994). The way in which parents are told of their child's disability may affect the way they adjust to the situation, and how they react to, and interact with their child (Pianta & Marvin, 1993c; Springer & Steele, 1980). There is also considerable evidence from the disability literature, of parental dissatisfaction with disclosure of the diagnosis (e.g., Sloper & Turner, 1993b).

Significantly, many mothers in this study reported that the diagnosis given at scan or at the birth was delivered in a manner indicative of poor practice. One mother described how she first learned of her son's cleft lip on palate when she had an ultrasound scan, and read the diagnosis of cleft, as it was being typed up on a screen. Another mother was simply handed the baby in silence at the birth, and therefore saw her baby's cleft, without being given a diagnosis, or any form of preparation. Often there was a dearth of information available about the condition and treatment at diagnosis, and in some cases, parents received misinformation from health professionals. For many women, the anger they still felt about how they were informed of their child's cleft, was evident in their accounts. One mother describes her experience:

There was one midwife who delivered me, and I think it (the cleft) shocked her. It was their faces, it was the look on their faces. I was so angry with the way I was told, the fact they treated him like he was a freak. That was the way I felt, and I still feel angry at the way they were.

Another mother reflected on first being given the diagnosis at a scan:

I think what really threw us was the fact that one of the consultants said there are only two decisions we need to make at this stage. Um, one was did we want to an amniocentesis to find out more, and indeed, whether we wanted to continue with the pregnancy. So that made us very worried because we thought if they were happy to terminate the pregnancy for something like this, it must be very, very serious. And I was horrified, completely at sea..... It makes me so cross now, because I now know it is something that can be corrected and dealt with. It gave an inaccurate reflection of the severity of the condition, and when you're in that vulnerable position any negative comments are unhelpful.

Accounts of these women suggest to some extent, that the way a diagnosis of cleft lip and palate is conveyed, either on scan, or at the birth, may affect parental adjustment to the birth of a child with cleft lip and palate.

Consistent with the broader disability literature on the functioning of parents who have a child with a disability, studies on the adaptation of parents with children who have craniofacial anomalies, provide reasonably firm evidence that these parents are more likely than parents of non-impaired children to experience higher levels of stress, anxiety and depression (Speltz *et al.*, 1990, 1993). And yet, it is imperative to stress that these findings are not definitive. As findings from the present study attest, for some parents, the impact of the birth of a baby with cleft may be less significant. However, the prevailing body of research has focused solely on negative outcome. Sloper and Turner (1993a) have argued that the effect of this approach has been to stereotype families, and to assume inevitable distress. This may be reflected in professionals' views, as research evidence shows that health professionals tend to predict more negative effects on families, that parents' own responses indicate (Sloper & Turner, 1993a).

Recent work in the field of disability has emphasised the strengths and successful adaptation of many families, and the need to identify both child and family factors relating to successful adaptation, in addition to risk factors for poor adaptation (Byrne & Cunningham 1985; Sloper & Turner, 1993a). Models of stress and coping (Lazarus & Folkman, 1984; McCubbin & Patterson, 1983) have provided a useful framework for the identification of important risk, resource and coping variables (e.g., Sloper, Knussen, Turner & Cunningham, 1991). Coping strategies play an important role in the way individuals respond or react to stressful situations or life events. Conceptualised in

terms of emotion-focused, problem-focused and avoidance dimensions, coping appears to mediate between antecedent stressful events and their consequences, such as anxiety and depression (Billings & Moos, 1981).

The present study has used a bereavement model to understand parents' experience of adjustment to the birth of a child with cleft lip and palate. Whilst this approach has allowed the identification of potential resilience and risk factors relating to adaptation, models that focus on stress and coping in families, may provide an alternative framework for studying the processes of adjustment in parents of children with cleft.

Directions for Future Research

In response to questions that have been raised by the findings, and also the limitations of the study, areas of future research are proposed.

This is a new, and primarily exploratory study into the areas of parental adjustment and prenatal diagnosis of cleft lip and palate. A number of tentative results have emerged, such as the potential distinction between internal and external adjustment, that require further investigation before any firm conclusions can be drawn. Replication of this work with larger participant groups, would also provide greater clarity regarding the patterns of resolution specific to parents of children with clefts, and perhaps also in relation to prenatal diagnosis.

The dynamic process of adjustment calls for a prospective study of parental adaptation. This would allow closer investigation of the factors that may interact between initial diagnosis of cleft lip and palate, and later adjustment. The complexity of this process cannot be underestimated, however, prospective research would also allow identification of transition points and times of crisis, in addition to coping strategies and resources utilised by parents over time. Thus, parent, child and social contextual correlates or predictors of more or less successful adjustment, could be identified.

Although mothers are generally the primary caretakers, the supportive contribution of the father is also an important aspect of family functioning (Pederson, 1980). With the increasing recognition of the importance of fathers responses in adjustment to a child's

disability, and differences in the factors that affect responses of fathers and mothers (Sloper *et al.*, 1991), research into the adjustment and coping of fathers of children with cleft lip and palate is long overdue.

As we have seen, research suggests that the way parents are informed of the diagnosis of their child's disability, may be a vital factor in subsequent coping (e.g., Springer & Steele, 1980). The limited attention this area has received from the literature, reinforces the need for future evaluation of the relationship between the informing process and subsequent adaptation, particularly within the cleft lip and palate population.

Furthermore, theory suggests that poor psychological adjustment and unsuccessful resolution of grief may have a negative impact on the child, and on the parent - child relationship (e.g., Pianta & Marvin, 1993c). In order to draw conclusions about the long-term effects of more or less successful adjustment to the birth of a child with cleft lip and palate, investigation of the long-term outcome for the child, in relation to parental adjustment or resolution, and prenatal diagnosis, is required. An evaluation of attachment behaviour in these children would be of particular interest, given the apparent association between resolution and the mother - child attachment relationship (Pianta & Marvin, 1993c), and the observed relationship between attachment status and later psychosocial problems in children (Urban, *et al.*, 1991).

Implications for Professional Practice

It is important to consider the implications that the findings of the present study have for clinical practice. A number of issues relating to the delivery of healthcare services for parents of children with cleft lip and palate have emerged from this research.

Firstly, there are a number of concerns relating to the needs of parents during and after diagnosis of cleft lip and palate. Clearly, the diagnosis of any congenital abnormality, whether during pregnancy or at the birth, should be given to the parent in a straightforward and sensitive way, in order to promote coping and adjustment. However, the experiences of mothers reported here suggest a need for further training and support of health professionals on how this information is shared with parents. Given that immediate access to professionals on a specialist craniofacial team is not

always an option, greater emphasis needs to be placed on the availability of accurate information on cleft lip and palate for parents, in antenatal departments and maternity wards. It is also important that efforts are made to convey this information in a manner which meets the individual needs of the parent. Initial support following diagnosis may be as important for some parents as information about cleft. Health professionals therefore need to give greater recognition to the emotional impact that the diagnosis of a child with cleft lip and palate may have on a parent. Furthermore, there may be a valuable role for psychology services in the support parents following diagnosis, in terms of both direct clinical work with parents, and indirect consultation work with other health professionals.

Primarily used in research, the theory of resolution of parental reaction to diagnosis (Pianta & Marvin, 1992), may also have particular value in the clinical setting, particularly in terms of providing an understanding of how parents may adapt to the trauma of the diagnosis of their child's disability, and with respect to the emphasis that this model places on the identification of resilience and risk factors that may be involved in adjustment. The Reaction to Diagnosis Interview (Pianta & Marvin, 1992), may also lend itself well to clinical practice. A reliable and efficient assessment tool, the interview is short, and the questions follow a clinical-type format which could easily be incorporated into an initial assessment, and used to inform clinical intervention.

Finally, findings from this study indicate that prenatal diagnosis may facilitate parental adjustment to the birth of a child with cleft lip and palate. This begs the question of whether specific prenatal screening for cleft should be offered for all women, as part of routine antenatal care. Linked to this issue is the ethical concern that, where termination is an option, the implementation of such a screening programme might reflect a move within society to a position of greater intolerance of disability. Maternal support for prenatal screening of cleft is by no means universal. However, it is notable that all of mothers in this study who had previously experienced prenatal detection of cleft, advocated the diagnosis of cleft lip and palate during pregnancy, despite awareness of potential disadvantages of this procedure.

The impact that women's preferences with respect to prenatal diagnosis, may have on provision of services is unclear. However, on reflection of the many issues that have emerged in this study, it is apparent that, if correctly managed, prenatal diagnosis may

develop an increasingly valuable role in the support of parents of children with cleft lip and palate.

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APPENDICES

APPENDIX 1

ETHICAL APPROVAL

RIVERSIDE RESEARCH ETHICS COMMITTEE

CHELSEA & WESTMINSTER HOSPITAL
Lower Ground Floor Pharmacy Offices
369 Fulham Road London SW10 9NH
Tel: 0181 846 6855 Fax: 0181 846 6860

Ms Andrea Heverin
Clinical Psychologist in Training
Craniofacial Team
Chelsea & Westminster Hospital
369 Fulham Road
LONDON SW10 9NH

Dear Ms Heverin

RREC: 1148 An Investigation into the Impact of Prenatal Detection of Craniofacial Abnormality on Parental Adjustment to the Birth of a Child with Cleft Lip and Palate.

I am writing to inform you that approval for the above study has been considered and approved by Chairman's Action.

Please note the following conditions which form part of this approval:

[1] This approval is for one year only. For projects with an expected duration of more than one year, a letter from the principal investigator will be required in order to further extend consent. This will enable the Committee to maintain a full record of research.

[2] Any changes to the protocol must be notified to the Committee. Such changes may not be implemented without the Committee's approval.

[3] The Committee should be notified immediately of any serious adverse events or if the entire study is terminated prematurely.

[4] You are responsible for consulting with colleagues and/or other groups who may be involved or affected by the research, e.g., extra work for laboratories. Approval by the Committee for your project does not remove your responsibility to negotiate such factors with your colleagues.

Cont/2

RREC: 1148 An Investigation into the Impact of Prenatal Detection of Craniofacial Abnormality on Parental Adjustment to the Birth of a Child with Cleft Lip and Palate.

[5] You must ensure that nursing and other staff are made aware that research in progress on patients with whom they are concerned has been approved by the Committee.

[6] Pharmacy must be told about any drugs and all drug trials, and **must** be given the responsibility of receiving and dispensing any trial drug.

[7] The Committee must be advised when a project is concluded and should be sent one copy of any publication arising from your study, or a summary if there is to be no publication.

May I take this opportunity to wish you well in your research. However, if any doubts or problems of an unexpected nature arise, please feel free to contact me at any time.

Yours sincerely

J.N Harcourt-Webster

Dr J N Harcourt-Webster MD FRCPath
Chairman - RREC

Seen and Approved	
Submission	Signed: J.N.H.W. Initials:
Protocol May 1996	Signed: J.N.H.W. Initials:
Information Sheet with Letter to Parents	Signed: J.N.H.W. Initials:
Consent Form For audio-tape.	Signed: J.N.H.W. Initials:
Questionnaires	Signed: J.N.H.W. Initials:
Letter of Indemnity	Signed: Initials: —
CTX/DDX/Licence	Signed: Initials: —

Great Ormond Street Hospital for Children NHS Trust

and the Institute of Child Health
(University College London Medical School)

The child first and always

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London WC1N 1EH

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20 December 1996

Ms A Heverin
Clinical Psychologist in Training
Department of Child Psychology
Chelsea & Westminster Hospital
369 Fulham Road
London
SW10 9NH

Dear Ms Heverin

96NR25 **An investigation into the impact of prenatal diagnosis of craniofacial abnormality on parental adjustment to the birth of a child with cleft lip and palate.**

Notification of ethical approval

The above research has been given ethical approval after review by the Great Ormond Street Hospital for Sick Children NHS Trust / Institute of Child Health Research Ethics Committee subject to the following conditions.

1. Your research must commence within twelve months of the date of this letter and ethical approval is given for a period of 6 months from the commencement of the project. If you wish to start the research more than twelve months from the date of this letter or extend the duration of your approval you should seek Chairman's approval.
2. You must seek Chairman's approval for of proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature, ie. using the same procedure(s) or medicinal product(s). Each research project is reviewed separately and if there are significant changes to the research protocol, for example in response to a grant giving bodies requirements you should seek confirmation of continued ethical approval.
3. It is your responsibility to notify the Committee immediately of any information which would raise questions about the safety and continued conduct of the research.
4. Specific conditions pertaining to the approval of this project are:

- The use of the enclosed standard consent forms for research. A copy of the signed form must be kept by you with the research records as our insurers may demand access to them.

Yours sincerely

Anna Jenkins

Anna Jenkins
Secretary to the Research Ethics Committee

enc

cc Mr B Sommerlad
Consultant Craniofacial Surgeon
Plastic and Reconstructive Surgery
GOS Trust

Mount Vernon and
Watford Hospitals
NHS Trust

The Clock Tower
Mount Vernon Hospital
Rickmansworth Road
Northwood
Middlesex HA6 2RN
Telephone 01923 826111

Fax.01923.844786

LOCAL RESEARCH ETHICS COMMITTEE

Chairman: Mrs. A. M. Rhodes 01923.844794
Secretary to Committee: 01923.844601

31st October 1996
ar/lc/ec96051

Ms. Heverin
Craniofacial Unit
Mount Vernon Hospital

Dear Ms. Heverin,

Re: EC96051: An Investigation into the Impact of Prenatal Detection of Craniofacial Abnormality on Parental Adjustment to the Birth of a Child with Cleft Lip and Palate.

Thank you for your letter of 10th October 1996 enclosing the amended letter and parents' response form. The conditions for full ethical approval of this study are now satisfied.

I am also now able to confirm that Chairman's Action was ratified at the meeting of the Ethics Committee on October 28th 1996.

Yours sincerely,

Anne Rhodes

Mrs. A. M. Rhodes
Chairman

RICHMOND TWICKENHAM & ROEHAMPTON HEALTHCARE NHS TRUST

From: Niall Johnston
Chair, Local Research Ethics Committee

Roehampton House,
Roehampton Lane,
London SW15 5PN
Telephone 0181 789 6611
Fax 0181 780 1089

Andrea Heverin
9 Tregothnan Road
London
SW9 9JU

30 October 1996

Dear Andrea,

AN INVESTIGATION INTO PARENTS' EXPERIENCE OF THE BIRTH OF A CHILD WITH CLEFT LIP AND PALATE

Ref: 961204/01

I am pleased to be able to inform you that the Local Research Ethics Committee has granted ethical approval for your research project within this Trust's area of responsibility.

Approval is granted subject to the proviso that the LREC must be informed, in writing:

- on completion of the project;
- if the project will over-run its estimated completion time;
- before any alterations are made to the treatment or protocol which might have affected ethical approval being granted;
- if any subject in the study reports adverse experiences.

Additionally, you should ensure that a positive response is received from parents whether or not they wish to participate in the study and the initial contact letter should make clear that non-participation will not prejudice future treatment.

You should note that the granting of ethical approval does not imply the consent of the Chief Executive of the Trust for the research to actually take place. This consent must be obtained before you commence the research.

Please ensure that we receive one copy of your final report.

With every good wish for your research.

Yours sincerely,

APPENDIX 2

CORRESPONDENCE TO PARENTS

Introductory Letter

An Investigation into Parents' Experience of the Birth of a Child with Cleft Lip and Palate

Dear *Parent*

I am carrying out a research study into parents' experience of having a child with cleft lip and palate. *X*, Consultant Plastic Surgeon, suggested that I contact you to ask whether you would like to take part in this study.

The main aim of this research is to investigate the feelings and worries that parents may have when their child is born with a cleft, and to look at how parents come to terms with this. It is hoped that by conducting this study, it will be possible to understand more fully parents' experience of the birth of a child with cleft lip and palate, and as a result, be able to meet parents' needs more effectively.

If you wish to take part in the study, this will involve a single interview lasting approximately one hour. The interview is confidential and would be carried out by myself at a time and place that is most convenient for you. Taking part in the research is voluntary, which means you can withdraw from the study at any time. I am happy to answer any questions that you may have about this research, and can be contacted on 0181 746 8972. Please indicate on the attached form whether you wish to be contacted, and return the form using the pre-paid envelope provided.

Thank you for taking the time to read this letter, and I look forward to receiving your reply.

Yours sincerely,

Andrea Heverin
Clinical Psychologist in Training

An Investigation into Parents' Experience of the Birth of a Child with Cleft Lip and Palate

Please indicate whether or not you wish to be contacted:

I **do / do not** wish to be contacted regarding participation in this study.

signed: .....

print name: .....

telephone number: .....

date: .....

Thank you letter

An Investigation into Parents' Experience of the Birth of a Child with Cleft Lip and Palate

Dear Parent,

Thank you for taking part in this study. Your contribution to this research into parents' experience of having a child with cleft lip and palate has been extremely valuable. As we've already discussed, all of the information you provided is completely confidential. Once the study is completed, I will send you a summary of the findings. However, if you have any queries about the research in the meantime, please do not hesitate to contact me on 0181 746 8972.

Best wishes.

Yours sincerely,

Andrea Heverin
Clinical Psychologist in Training

APPENDIX 3

INFORMATION SHEET AND CONSENT FORMS

AN INVESTIGATION INTO PARENTS' EXPERIENCE OF THE BIRTH OF A CHILD WITH CLEFT LIP AND PALATE

INFORMATION SHEET

You have already been contacted by the researcher, Andrea Heverin about this study. This information sheet describes the research in more detail.

1. The aim of the study

The main aim of this study is to investigate the feelings and worries that parents may have when their child is born with a cleft lip and palate, and to look at how parents come to terms with this.

2. Why is the study being done?

It is hoped that by conducting this research, it will be possible to understand more fully parents' experience of the birth of a child with cleft lip and palate, and as a result, be able to meet parents' needs more effectively.

3. How is the study to be done?

In this study, parents are interviewed about their experience of having a child with cleft lip and palate. They are also asked to complete three short questionnaires about their health, relationships, and the support they receive.

Should you decide to take part in the study, you will be interviewed once by the researcher at a time and place that is most convenient for you. The interview lasts approximately one hour. You will then be asked to complete the three questionnaires. If you are agreeable, the interview will be audiotaped so that it can be studied in detail at a later time. The tape will be held by the researcher until the end of the study, at which point it will be destroyed. All the information you give is confidential.

4. What are the risks and discomforts?

There are no anticipated risks or discomfort involved in taking part in this study.

5. What are the potential benefits?

It is unlikely that this study will bring any immediate benefits to you. However, it is hoped that by increasing our understanding of parents' experience of having a child with cleft lip and palate, services may be able to meet parents' needs more effectively in the future.

6. Who will have access to the case / research records?

The researcher and a representative of the Research Ethics Committee will have access to the information collected during the study.

7. Do I have to take part in the study?

If you decide, now or at a later stage, that you do not wish to participate in this study, that is entirely your right and will not in any way prejudice any present or future treatment.

8. Who do I speak to if problems arise?

If you have any complaints about the way in which this research project has been conducted, please, in the first instance, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact the Chairman of the Research Ethics Committee by post via the Research and Development Office, Institute of Child Health, 30 Guilford Street, London, WC1N 3EH, or if urgent, by telephone on 0171 242 9789 x 2620, and the Committee administration will put you in contact with him.

9. How to contact the researcher

You can contact the researcher to discuss this project at the following address:

Andrea Heverin
Clinical Psychologist in Training
Craniofacial Team, Chelsea and Westminster Hospital
369 Fulham Road, London SW10 9NH
0181 746 8972

Thank you for taking the time to read this information sheet

An Investigation into Parents' Experience of the Birth of a Child with Cleft Lip and Palate

CONSENT FORM

Please read and then tick the appropriate answer:

Have you read the information sheet?

Yes / No

Have you had the opportunity to ask questions and discuss this study? **Yes / No**

Have you received satisfactory answers to all of your questions ? **Yes / No**

Have you received enough information about the study? **Yes / No**

Do you understand that you are free to with draw from the study at any time, without having to give a reason, and without affecting your future medical care? **Yes / No**

Do you agree to take part in this study? **Yes / No**

Signed: Date:

Name in block capitals:

An Investigation into Parents' Experience of the Birth of a Child with Cleft Lip and Palate

AUDIOTAPE CONSENT FORM

Please read carefully and then sign and date.

I hereby agree to have this interview, conducted by Andrea Heverin, audiotaped. I understand that the tape will be held in her possession until the study is completed and that the information contained on it will only be accessible to researchers working on this study. On completion of the study the tape will be destroyed.

Signed: Date:

Name in block capitals:

APPENDIX 4

REACTION TO DIAGNOSIS INTERVIEW

1. When did you first realise that (child) had a problem (probe for detail) ?

2. What were your feelings at the time of this realisation?

3. How have these feelings changed over time?

4. Tell me exactly what happened when you learned of your child's cleft (lip / palate) Where were you, who else was there, what were you thinking and feeling at the time?

5. Parents sometimes wonder or have ideas about why they have a child with a medical problem. Do you have anything like that, that you wonder about?

   (Prompt if necessary: For example, some parents feel that they might have done something to contribute to their child's condition, others believe God must have had a reason for giving them this child. What do you wonder about?)

APPENDIX 5

INDICATORS OF PARENTAL ADJUSTMENT TO CONGENITAL DISFIGUREMENT

1. **Feelings about the baby's condition**
   1 = no distress at all
   2 = upset about it, but this gradually passed as time went by
   3 = still upset when talking or thinking about the birth

2. **Taking the baby out**
   1 = took the baby out with little anxiety
   2 = took the baby out, but found it worrying , and sometimes covered the part
   3 = could only take the baby out if the part was covered

3. **Taking of photographs**
   1 = took photos and displayed them
   2 = took some photos, but did not display them
   3 = did not take any photos

4. **Attitude to surgery**
   1 = could completely accept part before surgery
   2 = accepted part, but uncomfortable until surgery
   3 = could not completely accept things until after surgery

5. **Effect on decision to have more children, even if family complete**
   1 = completely unaffected
   2 = cautious approach, and would need scans and assurances, or wanted another child immediately
   3 = would not have any more children

6. **Effect on parental relationship**
   1 = drew together in mutual support
   2 = the situation caused some strains between parents
   3 = the situation contributed to the breakdown of the relationship