Parent-child bonding and attachment during pregnancy and early childhood following congenital heart disease diagnosis.

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ABSTRACT

Diagnosis and treatment of congenital heart disease (CHD) can present challenges to the developing parent-child relationship due to periods of infant hospitalisation and intensive medical care, parent-infant separations, child neurodevelopmental delay and feeding problems, and significant parent and child distress and trauma. Yet, the ways in which CHD may affect the parent-child relationship are not well-understood. We systematically reviewed the evidence on parental bonding, parent-child interaction, and child attachment following CHD diagnosis, according to a pre-registered protocol (CRD42019135687). Six electronic databases were searched for English-language studies comparing a cardiac sample (i.e., expectant parents or parents and their child aged 0-5 years with CHD) with a healthy comparison group on relational outcomes. Of 22 unique studies, most used parent-report measures (73%) and yielded mixed results for parental bonding and parent-child interaction quality. Observational results also varied, although most studies (4 of 6) found difficulties in parent-child interaction on one or more affective or behavioural domains (e.g., lower maternal sensitivity, lower infant responsiveness). Research on parental-fetal bonding, father-child relationships, and child attachment behaviour was lacking. Stronger evidence is needed to determine the nature, prevalence, and predictors of relational disruptions following CHD diagnosis, and to inform targeted screening, prevention, and early intervention programs for at-risk dyads.

Keywords: Attachment, bonding, congenital heart disease, parent-infant interaction, parent-child relationship, parenting.

Word Count: 196

INTRODUCTION

Early adversity is broadly conceptualised as any condition threatening a child's physical or emotional wellbeing [1-5]. Prenatally and throughout childhood, early adversity can initiate neurobiological adaptations influencing brain development, stress physiology and immune system functioning [1, 2, 6]. Caregiving environments may potentiate or mitigate the biobehavioural risks associated with early adversity [7-9], with secure attachment relationships potentially buffering the impact of adversity on child neurodevelopment [8, 10] and enhancing the efficacy of early developmental interventions [11]. Family-centred paediatric care is thus recommended in numerous clinical guidelines and position statements to support the development of children with critical or chronic illness, including congenital heart disease [12-16].

Despite this, limited research has examined how medical adversity may influence parent-child relationships. Studies of child attachment and parent-child interaction in high-risk medical populations, including amongst premature infants [17, 18], infants at high-risk for cerebral palsy [19], and children with various forms of critical or chronic illness [20], report inconsistent and contradictory findings. Few studies have evaluated the parent-child relationship following congenital heart disease (CHD) diagnosis, despite this being the most common birth anomaly, affecting approximately 1.35 million babies worldwide each year [21-23].

Although contemporary medical care has increased survival, complex CHD remains a leading cause of infant morbidity and mortality [24]. Parent-infant dyads may experience frequent and prolonged separations, invasive infant medical procedures, and exposure to aversive environmental stimuli during intensive care unit (ICU) admission [25-27]. Up to 50% of infants with CHD exhibit neurodevelopmental delay [28-30], associated with such factors as CHD complexity, prematurity, neurological injury, extended hospitalisation, and genetic factors [28, 31-34]. Neurodevelopmental delays may manifest as physiologic instability and self-regulatory difficulties, including sleeping, settling, and soothing difficulties [35, 36], behavioural disorganisation (e.g., fewer feeding cues) [27, 37], or atypical autonomic, state and motor organisation [33, 38]. Children with CHD are also at heightened risk of longer-term learning deficits, and emotion and behaviour regulation difficulties, including heightened rates of anxiety, autism and attention deficit hyperactivity disorder [28-30, 33].

Challenges such as these may adversely influence parents' capacity to identify and respond sensitively to children's cues, disrupting the dyadic interactive exchanges that underpin child attachment strategies [39-41].

Diagnosis and treatment of CHD is also associated with high parental psychological distress; a well-established risk factor for parent-child relational disruptions [42, 43]. Clinically-elevated symptoms of anxiety or depression are reported by up to 50% of parents of children with complex CHD, and 30-80% report severe psychological distress [16, 44]. For parents who receive their baby's CHD diagnosis during pregnancy, a sizeable proportion report prolonged periods of high distress, anxiety and depression [45-48], which may disrupt the developing parent-fetus bond [49] and parent-infant interaction postpartum [50, 51].

Factors outside of threats to child health can also influence relationship quality. Vulnerability to perinatal mental health difficulties, for example, may be heightened by socio-contextual risk factors, including adverse childhood experiences, limited social support, discordant couple relationships, and socio-economic hardship [52, 53]. Exposure to other stressful life events (e.g., bereavement) can also temporarily or permanently disrupt adult attachment patterns [54, 55], which is a core theorised pathway to parental sensitivity and responsiveness [56]. Higher attachment anxiety, for example, is associated with more threatening appraisals of motherhood and lower maternal self-efficacy following CHD diagnosis [55].

Despite these challenges, to our knowledge, no attempt has been made to systematically review the literature on parent-child relationships following CHD diagnosis. We thus aimed to identify, synthesise, and critically evaluate evidence on parent-child bonding, interaction and attachment following fetal or postnatal CHD diagnosis. We specifically focused on the first five years of childhood, during which caregiving relationships form a child's primary social context. We also aimed to determine biopsychosocial correlates and outcomes of these relational constructs, and use the findings to inform recommendations for future research, clinical care and health policy initiatives.

METHODS

The review protocol was prospectively registered with PROSPERO (CRD42019135687).The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [57] guidelines were followed to identify, extract and synthesise evidence from eligible articles (Supplementary Table 1).

Definition of Relational Measures

To facilitate comparison with previous literature, constructs of interest were defined as follows:

(1) *Parental bonding towards fetus or child:* Cognitive, affective, attitudinal or behavioural aspects of engagement by mothers and fathers towards their fetus or child [49]. Parental bonding (sometimes referred to as parental attachment, parental attachment feelings, or the parent-fetal or parent-child bond) is a function of the caregiving behavioural system which aims to protect and comfort the child, unlike child attachment which refers to the child's care-eliciting strategies under stress [58].

(2) *Parent-child interaction*: Includes (a) infant-initiated interactive behaviours in response to parental interactions related to an activity (e.g., engagement) or affect (e.g., positive or negative affect, vocalisations), (b) parent-initiated interactive behaviours in response to infant interactions related to an activity (e.g., proximity to infant) or affect (e.g., sensitivity, facial expressions), and (c) dyadic interaction, evaluated reciprocally from the parent and/or child's perspective (e.g., emotional attunement, behavioural synchrony, contingency). Both macro- and micro-level observational coding tools were considered eligible. At the macro-level, coding focuses on broad (or global) aspects of parent or child behaviour, usually over seconds to minutes [59]. Micro-level coding, on the other hand, focuses on fine-grained aspects of behavioural states often occurring rapidly, outside conscious awareness, over small time segments (e.g., coding video frames in <1 second intervals) [59]. No restrictions were placed on the assessment context (e.g., feeding, free or structured play).

(3) *Child attachment*: A child's expectancies regarding the nature and quality of care received from primary caregivers, and associated behavioural strategies for proximity and safety-

seeking during times of distress [60, 61].

Search Strategy

Six electronic databases were systematically searched (MEDLINE, PsycINFO, CINAHL, EMBASE, SCOPUS, Maternity and Infant Care) to identify relevant literature from inception to January 2020. Keywords were designed to capture the target population (i.e., congenital heart disease and specific diagnostic subgroups) and outcomes of interest (i.e., parent-child bonding, parent-child interaction, and child attachment patterns) (Supplementary Table 2). Ancestry searching and citation chaining were performed to identify additional literature. Searches were limited to human studies published in the English language in a peer-reviewed journal. Titles were screened by one reviewer and all remaining abstracts and full-texts were independently screened by two reviewers. Any discrepancies in screening were resolved through discussion and review of the article against inclusion and exclusion criteria. All final articles were approved by all authors.

Study Selection Criteria

Inclusion criteria were as follows: (1) assessed a relational outcome, including parent-child bonding, interaction, or child attachment classification using a parent-report or observational measure; (2) reported on mothers and/or fathers and their fetus or child (mean age ≤5 years) with CHD; and (3) included a healthy comparison group (i.e., an active or retrospectively-enrolled control sample without critical or chronic illness, or normative community data). Exclusion criteria included: (1) reported on expectant parents with a fetal CHD diagnosis who opted for pregnancy termination or comfort care; (2) non-peer reviewed or grey literature; (3) employed a qualitative or interventional methodology (unless pre-intervention data were reported); or (3) referenced no, or a medically-unwell, comparison group. Mixed samples were eligible if >60% of participants met inclusion criteria, or CHD outcome data was reported separately or provided on request.

Data Extraction

Data were initially extracted by one reviewer using a standardised pre-piloted form to collect: publication characteristics (e.g., authors, year), study design (e.g., cross-sectional), cardiac and comparison group characteristics (e.g., age at assessment, gender), outcome measures (e.g., assessment methods, scoring), main results, and correlates, predictors and consequences of relational outcomes. A second reviewer read all included articles and cross-checked all data extracted from each article for completeness and accuracy. Coding differences were resolved through discussion until consensus was reached. The level of agreement was extremely high (a total of three differences identified: all typographical errors). Methodological quality was independently assessed for each study by two reviewers using the QualSyst tool [62]. The QualSyst tool includes 14 items to appraise methodological rigour for quantitative research based on study design, methodology, data analysis, and outcome reporting. Scores are calculated based on whether the article meets criteria for each weighted item; 'Yes' (score of 2; criterion adequately addressed), 'Partial' (1; partially addressed), 'No' (0; not addressed), or not applicable. A total methodological rigour score is then calculated from all applicable items, by dividing the total score by the total possible score to provide a rating out of 1. Higher scores indicate greater methodological rigour (>0.8='Strong', 0.71-0.79='Good', 0.50-0.70='Adequate'; <0.50='Limited'). Both reviewers independently assessed all final full-texts based on the 14 QualSyst items, and coding discrepancies were resolved through discussion until consensus was reached. Agreement between the two reviewers was extremely high (96%; 12 discrepancies identified from 318 ratings; see Supplementary Table 3).

Data Synthesis and Analysis

Narrative summary was the primary reporting method, with findings grouped according to similar relational constructs. Statistical synthesis using meta-analysis was considered, but not possible due to considerable heterogeneity in study designs, outcome measures and sample characteristics.

RESULTS

Following screening of 6,459 unique titles, 266 articles remained for abstract review. Agreement between reviewers for abstract screening was 94% (Cohen's Kappa=0.82). Of the 89 articles

remaining for full-text review, agreement for the full-text screening strategy was 90% (Cohen's Kappa=0.80). In total, 32 articles representing 22 unique studies were identified as eligible and included in the final synthesis (Figure 1).

Sample Characteristics

Studies sampled parent-child dyads predominantly from the United States (*n*=9) and most articles (*n*=16) were published between 2010-2019 (Table 1). Assessments were conducted at various time-points; one study assessed parent-child relational outcomes during pregnancy [63], 11 in infancy at 0 to 12 months [64-79], two in toddlerhood at 12 to 36 months [80, 81], seven in early childhood at 36 months to 5 years [82-88], and one spanned multiple developmental phases [89-94] (Table 1 and 2). Mother-child relationships (*n*=16) were mostly assessed; fewer studies reported on the father-child relationship (*n*=5). Mean child age at assessment ranged from 17.1 days to 5.4 years and mean parent age at assessment ranged from 27.6 to 35.3 years. Cardiac anomalies ranged from mild (e.g., small ventricular septal defects) to more complex (e.g., hypoplastic left heart syndrome), although varied within and between studies. Most samples (*n*=18) included children who had undergone cardiac surgery. Other disease-related factors (e.g., time of diagnosis, age at surgery, length of hospitalisation) and social factors (e.g., race, ethnicity, socio-economic status) were not consistently captured (Supplementary Table 4).

Study Design and Outcome Measures

Studies predominantly used parent-report measures (*n*=16), although observational measures (*n*=4) and combined parent-report and observational measures were used (*n*=2) (Table 1 and 3). The Parenting Stress Index was used in fifteen studies [66-68, 75-79, 81-85, 87, 91, 95]. Parent-infant interaction was assessed in five observational studies, using various coding strategies, including: micro-analytic interval-based [86] and frequency-based coding systems [82], macro-analytic rating scales [69, 70, 80, 89, 93] and scoring checklists [73, 74], and combination methods [94]. No two studies used the same coding system and two used non-validated systems. Child attachment classifications were assessed in one study only [90] using Ainsworth's Strange Situation Procedure

(SSP) [96]; as analyses were conducted on the same sample longitudinally, we report on data from the largest sample [90, 92-94]. Most studies were cross-sectional (*n*=17); only five studies collected longitudinal data [63, 67-71, 76, 89, 90, 92, 93] (Table 3). Only one study tested a theoretical framework; Shore's theory of the development of child self-regulation [69-71]. Comparison groups included active control samples free of critical or chronic illness (*n*=13) and community-based normative data (*n*=9).

Assessment of Methodological Quality

Overall, methodological quality was 'strong' (*M*=0.80, *SD*=0.96), ranging from 0.55-0.95 (Supplementary Table 3). Of the included studies, 17 had 'strong' methodological rigour, 9 had 'good', and 5 had 'adequate' rigour based on the QualSyst scoring system. No clear patterns were found when results were examined according to methodological quality category or overall QualSyst score, for any relational construct (i.e., bonding, parent-child interaction, or child attachment pattern). Common problems included the use of small samples sizes and absence of power analyses, inappropriate or poorly described statistical analyses, inadequate descriptions of sample characteristics, and limited or no control for confounding factors.

Result Overview

Key findings are summarised in Table 4 and Figure 2. Results are grouped according to measure type and construct, and grouped based on whether the study demonstrated more positive, similar, or poorer outcome(s) when comparing cardiac dyads with a healthy referent group.

Parent-Reported Outcomes

Parent Bonding and Attachment Towards Fetus or Infant (8 studies)

Only one study assessed parental-fetal attachment [63]; expectant mothers who received a fetal CHD diagnosis reported higher global maternal-fetal attachment at ~27 weeks' gestation compared with women who received a healthy echocardiography scan, controlling for pre-scan attachment levels [63]. Two studies assessed parent-infant bonding postpartum. One found that mothers and fathers of

infants with CHD reported global attachment feelings comparable to an Australian community sample, within two months post-hospital discharge for cardiac surgery [65, 72]. Overall, 15% of these mothers and 19% of these fathers reported scores indicative of 'low attachment feelings' [65, 72]. Fathers also reported lower affection and pride towards their baby with CHD, and less pleasure, satisfaction and competence, but no difference in patience and tolerance, compared to community norms [65]. Maternal data on these domains was not reported [72]. The second study [64] found lower global attachment amongst mothers of 0-12 month old infants with moderate and complex CHD relative to mothers of typically-developing infants using the Maternal Attachment Inventory [97]. Five studies assessed parenting stress, in terms of how attached parents felt towards their child with CHD, using the Long-Form Parenting Stress Index (PSI); four found similar levels of attachment amongst parents of children with CHD and typically-developing children from age 3 months to 6 years [67, 79, 81, 87]. Only one study found a stronger parent-reported attachment towards their child with CHD (aged 3-6 years) relative to community norms [95].

Parenting Stress and Parent-Child Interaction (15 studies)

Six studies assessed parenting stress in terms of how positively reinforced parents' felt by interactions with their child, using the Long-Form PSI. Three studies found similar levels of positive reinforcement [67, 79, 87], one study higher reinforcement [95], and two studies found lower reinforcement [81, 91] amongst parents and their child with CHD compared with healthy referents.

Nine studies assessed parent perceptions of dysfunctional parent-child interaction; four examined between-group differences and all found that parents of children with CHD and typically-developing children reported similar levels of dysfunctional interaction from 10 months and 5 years [77, 78, 82, 83, 85]. Five studies examined the prevalence of 'at-risk' dysfunctional interactions reported by parents of children with CHD, which ranged from 8% (n=25) to 55% (n=48) across various ages and medical stages [66, 75, 76, 84, 85].

Observational Paradigms

Child Attachment (1 study)

Child attachment patterns were assessed longitudinally using the SSP in one study [96]. Categorised using Ainsworth's tripartite system [96], insecure-avoidant classifications were most prevalent amongst toddlers with CHD (28%) and cystic fibrosis (38%) at 12-18 months, and the frequency of secure attachment classifications was lowest amongst children with CHD (57%) and cystic fibrosis (55%), relative to healthy counterparts (14% and 73%, respectively) [90]. Re-categorised using Main and Solomon's four-classification system [98], disorganised attachment was highest amongst children with CHD (26%) and cystic fibrosis (33%) relative to healthy children (12%) [90]. When the two medical groups were combined, children with CHD or cystic fibrosis had a higher proportion of insecure-avoidant and disorganised attachment classifications, compared to healthy children [90].

Parent-Child Interaction (6 studies)

Results are described for parent, followed by child, and dyadically-focused constructs below. Findings are organised according to interactional context (i.e., feeding, separation and reunion, free- or structured play) as synthesis of common relational constructs was not possible across the different measurement and coding systems.

Parent Interactive Behaviour (6 studies)

Six studies examined parent interactive behaviour, of which two examined mother-infant feeding. Harrison et al. (2009, 2013, 2014) found that mothers of 2-week old infants with CHD (but not 2-month or 3-year old children) showed more optimal caregiving behaviours, including more support, attunement and warmth, more positive affective involvement, sensitivity and responsiveness, and greater regulation of negative affect and behaviour, compared with mothers of typically-developing infants [69-71]. Contrarily, Lobo et al. (1992, 1995) observed lower socioemotional growth fostering amongst mothers of 4-month old infants with CHD, but no differences in cognitive growth fostering, sensitivity to infant cues, or responsiveness to infant distress, compared with mothers of typically-developing infants [73, 74].

Three studies assessed maternal play behaviour. During a free play task, Laing et al. (2010) [80] found compromised maternal behaviour with their 18-36 month old toddler with CHD, including

lower sensitivity to child distress and non-distress cues, more intrusiveness and over-control, and less positive and more negative affect, relative to community norms [80]. Mothers were also more emotionally and physically detached and disengaged, lacked vocal and facial animation, and had greater difficulty fostering child cognitive development [80]. On a structured play task, Goldberg et al (1990a) found that mothers of 24-30 month old toddlers with CHD displayed a more directive parenting style and engaged in more task-structuring and limit-setting, but showed similar expressed hostility and teaching behaviour (i.e., supportive presence, respect for child autonomy, quality of assistance, and parenting confidence) compared with mothers of typically-developing children [93]. No between-group differences in maternal behaviour were observed on a structured play and clean-up task with 2-5 year old children [82].

Three other studies evaluated parental behaviour during separation and reunion tasks (two overlapping with the sample in Goldberg et al., 1990a). Madigan et al. (2011) found no differences in atypical and disrupted behaviour at 12-months or 7-years, comparing mothers of children with CHD, cystic fibrosis and healthy children [94]. One study examined fathers' behaviour with these same children at 12-14 months, and found no differences in positive paternal greeting and departure behaviours, positioning, sensitivity, elaborativeness, comfort and mutuality compared with fathers of typically-developing children [89]. One Brazilian study used a study-specific separation and reunion paradigm and observed that mothers asked fewer questions of their 4-5 year old child with CHD, but interacted more frequently than mothers of healthy children [86].

Child Interactive Behaviour (6 studies)

Infant interactive behaviour was assessed in the two feeding studies. At 2-weeks and 2-months, Harrison et al. (2009; 2013; 2014) found no differences in positive or negative affect (e.g., irritability, dysregulation), or communication and social skills, comparing infants with CHD and typically-developing children [69-71]. In contrast, Lobo et al. (1992, 1995) found 4-month old infants with CHD expressed more subtle distress behaviours (e.g., more hand-to-head behaviour, finger splaying), less clear cues, and lower responsiveness than typically-developing peers [72, 73].

Child interaction was examined in the three play studies. During free play, Laing et al. (2010) found that 18-36 month old toddlers with CHD showed less positive behaviour, including less positive mood, poorer sustained attention, lower responsiveness and social connectedness, and more physical activity compared with community norms [80]. Similarly, 24-30 month old toddlers with CHD showed more problematic behaviour during structured play, including less persistence and cooperation, less positive play, and greater maternal dependence than their typically-developing peers, but did not differ in affect, negativity, enthusiasm or task avoidance [93]. In contrast, during structured play, Carey et al. (2002) found no between-group differences in the frequency of positive behaviours (e.g., positive physical touch), negative behaviours (e.g., total criticisms, yells, whines, negative physical touch, destructive behaviour), compliant or non-compliant behaviours comparing 3-year old children with CHD and typically-developing peers [82].

Child interactive behaviours were examined in two separation and reunion tasks. In the father-child study, Darke and Goldberg (1994) found no differences in child prosocial behaviour, including expressions of positive affect, social responsiveness, or departure reactions comparing 12-14 month old toddlers with CHD and healthy same-age children [89]. Peçanha and colleagues (2015), however, described 4-5 year old children with CHD as less responsive to maternal questioning than healthy children, although no differences were found in engagement, compliance, interaction or independence [86].

Dyadic Interactive Patterns (2 studies)

Two studies examined dyadic constructs, both during mother-infant feeding. No between-group differences in dyadic mutuality, reciprocity and tension were found for mother-infant dyads at 2-weeks or 2- months [71]. Conversely, in another study, lower dyadic attunement, synchrony and contingency were found amongst mothers and their 4-month old infant with CHD compared with typically-developing dyads [73].

Factors Associated with Parent-Child Relational Outcomes for CHD Group

Sociodemographic factors

Four studies examined socio-demographic factors; no associations were found between parent age [65], educational attainment [65], socioeconomic status [65], maternal occupational status [72], or infant gender [90] and any relational outcome. Older children with CHD played more positively (i.e., positive affect, greater communication and social skills) than younger children in one study [71]. Mothers reported less dysfunctional interactions with first-born infants with CHD compared to those with older sibling(s)[78], but father-infant attachment did not differ based on birth order [65].

Medical and physiological factors

Three studies examined cardiac characteristics and treatment factors; no associations were observed between relational outcomes and type of cardiac anomaly [65], cyanosis [72], disease severity [92], age at diagnosis [92], delay in diagnosis [92], number or length of hospital admissions [65, 72, 92], cardiac surgery [92], surgery type (corrective vs. palliative) [65, 72], or timing of diagnosis [72], with two exceptions. Fathers who spent less time at home prior to their infant's cardiac surgery reported lower patience and tolerance, pleasure in father-infant interaction, and global attachment post-operatively, and fathers who received a fetal (relative to postnatal) cardiac diagnosis reported lower patience and tolerance [65]. Pregnancy intendedness was examined in two studies; one found stronger maternal-reported attachment to a planned baby [63]; the other found no association between these two variables [64].

Four studies examined associations with infant physiology. No associations between maternal or infant behaviour and infant heart or respiratory rate were found [74]; however, 3-year old children with CHD showed greater parasympathetic recovery when their mothers displayed greater regulation of anxiety, lower intrusiveness, and more positive affective involvement [69-71] compared with typically-developing children. Expectant mothers with higher blood pressure and those who imagined greater childbirth pain also reported stronger prenatal attachment towards their unborn baby with CHD [63].

Psychological factors

Two studies examined parental depression and relational outcomes. Mothers who endorsed higher scores (i.e., above community and clinical cut-offs) on the Edinburgh Postnatal Depression Scale reported weaker attachment towards their infant (aged 3 months) in one study [72]. Less positive play behaviour was also observed amongst fathers with greater parenting-related depression, and where fathers and mothers reported greater stress [89]. No associations were found between parental stress and infant attachment classifications [92].

CHD was associated with lower quality of child and maternal interaction at 24-30 months (e.g., more maternal limit-setting, lower infant affection) compared to healthy referents; however, when neurodevelopmental status at 12-18 months (assessed using the Bayley Scales of Infant Development (BSID-II) Mental Development Index) was included as a covariate, these differences were no longer present [90]. No association was found between child attachment classification (assessed using the SSP) and neurodevelopment (assessed using the BSID-II) or temperament [92], nor were relational outcomes associated with receiving CHD-related information or maternal perceptions of CHD aetiology [64].

DISCUSSION

Family-centred care is recommended to support child development in medical settings, including fetal and paediatric cardiology [12-16]. Yet, the ways in which CHD may impact the parent-child relationship is not well-established. We identified 22 controlled studies of the parent-child relationship during pregnancy or early childhood (birth to five years) following CHD diagnosis. Most relied on parent-report measures (73%), focused on the parent-infant relationship (50%), and yielded mixed results when comparing parental bonding and parent-child interaction between parents of children with CHD and parents of typically-developing children. Observational data were scant and also yielded mixed results, although most studies (4 out of 6) found difficulties in mother-child interactions following cardiac diagnosis (e.g., lower maternal sensitivity, lower infant responsiveness). Wide variation in methodological approach was found and may contribute to the mixed findings across the included studies. No consistent pattern of results was found based on the level of methodological rigour or QualSyst score. Research on maternal and paternal bonding with the

fetus after prenatal cardiac diagnosis, father-child relationships, and child attachment behaviours was extremely limited, as was detailed observational analysis of dyadic interaction.

Interpretation of Key Findings

Greater maternal-fetal bonding was found following prenatal CHD diagnosis in the one study carried out during pregnancy identified in our review [63]. In contrast, some [99] but not all studies within the broader literature [100] have found decreased maternal bonding during pregnancy following diagnosis of various other fetal malformations (e.g., gastrointestinal and renal anomalies). Given the high attrition (91%) in the single CHD study identified, robust research is needed to better understand the prenatal bonding process after fetal cardiac diagnosis. Postpartum, five studies in our review found that parents reported global attachment feelings towards their infant with CHD comparable to community-based norms; however, we also found evidence of distortions in bonding processes normally assumed to underpin attachment relationships. Fathers, for example, reported lower affection, pride, pleasure, satisfaction and competence parenting their infant with CHD [65]. Some studies in our review also found that parents reported weaker attachment and less positive reinforcement in interactions with their child with CHD compared with community norms. Whilst these findings provide some evidence of the robustness of the attachment-caregiving system and its evolutionary capacity for adaptation and compensation under stress, including childhood illness [9], overall, the mixed pattern of relational outcomes highlights the need for further investigation. Strikingly, not a single study identified in our review examined the link between parent-report and observational measures. Meta-analytic data show low convergence between the two (r=0.17) [101]; for example, within the broader literature, a study by Zaslow et al. (2006) including low-income families found no relationship between maternal-reported controlling behaviour and actual harsh child treatment [102]. Self-report of relational dynamics is influenced by numerous factors, including individual attachment representations, childhood experiences, reporting bias, and capacity for insight [103-105]. Given that parent-report measures appear to be a poor proxy of attachment-related behaviours, caution is needed when interpreting the present results. A deeper understanding of specific bonding processes (e.g., parental reflective functioning, mentalization, emotion regulation)

and their predictors following CHD diagnosis is also needed [106, 107]. Clinically, targeted, developmentally-sensitive education and care should be offered to parents reporting bonding difficulties. Interventions focused on supporting the parent-child relationship and buffering parental psychological distress have demonstrated efficacy in increasing maternal-reported attachment in the neonatal ICU and may have translational benefit in the cardiac setting [108].

Patterns of parent-child interaction were also mixed. Four of six observational studies identified in the present review found compromised interactive patterns amongst mothers of children with CHD in at least one domain; for example, lower maternal sensitivity and responsiveness, and more disruptive and intrusive behaviours compared to healthy referents. Similarly, in four of six observational studies, infants and children with CHD displayed lower interactional quality, including lower responsiveness and clarity of cues, from 4-weeks to 4-years of age. These findings were observed across various interactional contexts (e.g., feeding, free and structured play, and separation and reunion). Parent-reported rates of relational dysfunction also varied widely across studies included in our review with estimates ranging from 8% to 55%. Overall, these results suggest that a proportion of mother-child dyads experience relational disruptions following CHD diagnosis. One possible explanation is that infants with CHD share common relational challenges with other hospitalised infants. Premature infants, for example, have been described as less alert, attentive, and responsive compared with full-term infants, and their mothers as more active, over-stimulating, and affectively disengaged [28-30]. This has been attributed to prolonged early separations, high maternal distress, and poorer infant signalling and communication of cues due to delayed neurological maturation [17]. For mothers of premature infants (and possibly also mothers of infants with CHD), patterns of over-stimulation may represent an appropriate response to compensate for their infant's poor signalling [17], underscoring a need to better understand why interactional difficulties emerge in the CHD context.

In contrast, we also identified several studies that found no difference, or more positive maternal and child behaviours, amongst cardiac dyads relative to healthy referents. For example, one study found no difference in maternal sensitivity to infant cues during feeding between those with CHD and typically-developing infants [73]. Father-child interaction also did not differ between

groups in the one available observational study of paternal involvement in play with their 3-year old toddler with CHD [89]. Infant attachment patterns were assessed in only one study, which found no difference in the proportion of secure and insecure attachment classifications between toddlers with CHD aged 12-18 months and healthy referents. In the broader literature, many studies have not found higher rates of insecure attachment in medically-fragile infants, including premature infants [109], infants with cystic fibrosis [110], and children with physical and developmental impairments [111] compared with community samples, though it is not uncommon for these studies to be small and underpowered [90].

Overall, this systematic review found a heterogenous pattern of parent-infant interaction and attachment following CHD diagnosis. Methodological variation may contribute to the mixed findings. Parent-report measures were often used despite susceptibility to defensive reporting and impression management [112], whilst observational data was limited. The few available observational studies employed various micro-analytic (e.g., Dyadic Parent-Child Interaction Coding System [113]) and macro-analytic coding systems (e.g., Parent-Child Early Relational Assessment [114]), with limited consistency in the constructs assessed and interactional contexts.

Surprisingly, the potential role of disease- and treatment-related factors were rarely examined, despite the general appreciation that factors such as prolonged hospitalisation and neurodevelopmental delay may increase the risk of lower parent-child interaction quality and insecure or disorganised child attachment [19, 115-117]. Only three studies investigated these associations, of which one found fathers who received a fetal CHD diagnosis and spent fewer days at home before their infant's cardiac surgery reported lower patience and tolerance, pleasure in interactions. and global attachment quality [64, 65, 72, 92]. Further investigation of the relationship between birth-, disease- and treatment-related factors and parent-child relationship quality is therefore strongly recommended. Parental psychological factors were also seldom evaluated, despite the well-documented high levels of maternal and paternal distress in neonatal, paediatric and cardiac ICU settings [44, 118, 119], and known links with parental sensitivity [42, 43]. Two of three studies identified in this review found a higher risk of poorer relational outcomes given maternal and paternal postpartum stress and depression [72, 89]; however, the impact of parental mental health, attachment

styles, and other protective factors (e.g., social and marital support) remains unclear. The potential role of sociodemographic factors also warrants further study [120].

Overall, notwithstanding these methodological issues, we did not find evidence of widespread disruptions in early caregiving relationships following CHD diagnosis. Rather, it appears a proportion of dyads are vulnerable to interactional difficulties. Patterns of results did not appear to vary based on methodological quality, indicating a need for further research to quantify the rates of attachment-related difficulties, and identify factors contributing to relational risk and resilience.

Limitations of the captured studies and areas for future research

Eco-bio-developmental frameworks highlight the importance of early protective and risk factors in shaping child neurodevelopmental trajectories [121, 122]. Yet, only one of the studies identified in the present review explicitly tested a theoretical framework [69-71]. Multi-determinant frameworks are needed to better understand predictors, mediators and moderators of relational outcomes. Belsky's Model of Parenting Determinants [123] focuses on child, socio-contextual and parental psychological factors, and is one example. Contemporary attachment and mentalization-based approaches may also have utility here; for example, Fonagy's Theory of Mentalization [124] is another well-substantiated model linking parental reflective function (i.e., a parent's understanding of their own and their child's mental states) to child attachment strategies and mental health [125, 126]. Longitudinal research is also needed to investigate the 'parental buffering hypothesis'; that is, whether secure parent-child attachment relationships can mitigate the high rates of neurodevelopmental, socio-emotional and behavioural difficulties found amongst children with CHD [4].

Transactional models take into account the dynamic interplay between parent and infant as active partners in a non-linear, reciprocal system [127]. Parental sensitivity, for example, is influenced by both child (e.g., temperament, medical illness) and parent characteristics (e.g., personality, childhood experiences of caregiving) [128, 129]. Yet, in our review, only two studies incorporated dyadic measures of the parent-child relationship and yielded conflicting findings [71, 73]. Paradigms incorporating dyadic constructs alongside discrete parental and child constructs, such as Crittenden's (2006) Child Adult Relational Experimental (CARE) Index [130] are recommended to assist in

differentiating the observed content of a behaviour (e.g., maternal smiling) from its functional significance in the relationship (i.e., what the smile signals or communicates to the child; see Provenzi et al., 2018 for a relevant review of dyadic constructs [40]). Validated microanalytic measures, such as Tronick et al.'s (1978) Still Face Paradigm [131], and representational measures, such as the Working Model of the Child Interview [132], may also have utility in studying parent-child representational processes and communication disturbances after cardiac diagnosis, especially given that few studies identified in our review explicitly assessed 'relational risk' [39, 133].

Family systems approaches espouse the importance of the family unit in adapting to stressors [134]. Despite this, only five studies in our review examined the father-infant relationship, echoing critiques across paediatric psychology research in general, and paediatric cardiology research specifically [135]. Traditionally, research has suggested that fathers adopt a protector and playmate role [136], while mothers play an instrumental role in early caregiving. This warrants investigation of potential gender differences in CHD. More culturally and ethnically diverse samples are also needed, given the under-representation of non-Caucasian ethnicities, including Indigenous and minority populations in this review [60, 137].

Other methodological limitations of the identified studies include small samples, absent power calculations, inappropriate or poorly described statistical analyses, and inadequately described sample characteristics. Limitations of this review include exclusion of non-English literature and qualitative studies, which may restrict exploration of cultural differences in parenting behaviours and potential explanations for the inconsistent relational outcomes identified. Exclusion of grey literature may also bias the present results; for example, published research is more likely to contain statistically significant findings and is associated with larger effect size estimates, compared with unpublished research [138, 139]. Further, despite an a priori decision to exclude studies including children with significant comorbidities, many articles described minimal sample characteristics, making it difficult for the authors to identify potential differences in attachment and bonding outcomes based on health-related factors.

Clinical Implications

Identifying dyads at risk of relational difficulties represents an important health, societal and economic priority, as secure attachment relationships are foundational to child health, development, and wellbeing [8, 10, 11, 140]. From an economic perspective, evidence shows that family-centred early childhood screening and intervention is cost-effective and generates favourable long-term economic returns [141-144]. Routinely offering parents and their infants an opportunity for bonding and attachment assessment using clinical interviewing and observational paradigms is thus strongly recommended as part of CHD care, alongside mental health and neurodevelopmental evaluation. While stronger evidence is critically needed on the efficacy of parent-infant psychotherapy interventions in CHD [16, 145], preliminary evidence supports the utility of tailored psycho-education, family-centred developmental care during hospital admissions, opportunities for promoting parent emotional expression and parent-child bonding, support in fostering parenting and coping skills, and problem-solving strategies [16, 146]. Various therapeutic approaches also have a strong evidence-base for improving parent-child relationships within the general community (e.g., attachment-based psychotherapy programs, such as Watch, Wait, and Wonder [147] and the Circle of Security [148]), and should be considered as early as possible. For example, the Watch, Wait, and Wonder attachment-based program is a child-led psychotherapeutic approach that directly draws on the child's spontaneous activity and free play to enhance parental sensitivity and responsiveness, prompt reflection on child and parent needs, and strengthen the parent-child attachment relationship [147]. Further, evidence shows that treating maternal psychiatric disorder, in particular perinatal anxiety and depression, is necessary but not sufficient for healing parent-child relational difficulties, which necessitates focussed attention on the parenting role and dyadic interactions between parent and child [149].

Research is also accumulating on the benefits of telehealth for parent-child psychotherapeutic intervention, with preliminary meta-analytic evidence of efficacy. For example, Internet-delivered Parent-Child Interaction Therapy [150] and other behaviourally-based parenting interventions [151] have demonstrated improvements in parent mental health, family interactions and functioning, and reductions in child behavior problems. Technology-assisted observational assessments may also provide another pathway to identifying parent-child dyads at risk of relational difficulties, with

evidence that the Child-Adult Relationship Experimental (CARE)-Index can be distance-delivered with high feasibility and acceptability [152]. Distance-delivered assessment and intervention may thus provide a promising and cost-effective solution to scaling evidence-based practices and may increase the reach and accessibility of early family intervention services for those in need, including families from 'harder to reach groups', such as minorities and those from regional and remote areas. Integrated, stepped care approaches may also assist in improving the efficiency of resource use and cost, with the intensity of evidence-based interventions tailored to the level of need [153, 154]. An embedded model of stepped mental health care has been effectively implemented at the Heart Centre for Children in Sydney, Australia, to facilitate coordinated, collaborative care, and has the potential to significantly reduce the burden of care on clinical psychologists and other mental health providers [155]. We also recommend clinicians utilise existing, community-based referral pathways to perinatal mental health services, child protection and social services, or domestic violence organisations, as indicated.

Family-centred developmental care is also strongly recommended to manage acute stressors in the hospital setting. Within the neonatal [156] and cardiac ICU [27], models of individualised family-centred care have been implemented, incorporating a focus on parental engagement, cue-based care, and creating a nurturing and supportive environment. These programs have shown efficacy in improving parent-infant bonding, parent mental health and caregiver confidence, as well as preliminary evidence of improved infant neurodevelopment and feeding [146, 157, 158]. Interdisciplinary teamwork is also strongly recommended, involving psychology, medical, nursing, and allied health providers to facilitate a collaborative, team-based approach to supporting parent-child bonding across the continuum of care.

Conclusions

While parent-child relationships do not appear universally disrupted by CHD diagnosis and treatment, some dyads are at higher risk of interactional and attachment difficulties. Stronger empirical evidence is needed on the nature, prevalence and consequences of parent-child relational disruptions in the

context of CHD. Use of multidimensional conceptual models is strongly recommended to assist in identifying factors that contribute to relational risk and resilience, and to facilitate targeted screening, assessment and intervention for at-risk dyads in paediatric medical settings, including congenital cardiac care. Family-centred care remains foundational to paediatric care and specific policies and best-practice recommendations are needed to support parent-child relationships following diagnosis of critical and chronic childhood medical illness.

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TABLES AND FIGURES

Table 1. Summary of Included Study Characteristics.

Study Characteristics	Number of Unique Studies (*n*=22)
Study Design	
Cross-sectional	17
Longitudinal	5
Year of Publication	
Prior to 2010	6
2010-2019	16
Sample Size	
<50	9
50-99	5
>100	8
Child Developmental Stage	
Fetus: ~27 weeks gestation	1
Infancy: 0-12 months	11
Toddlerhood: 12-36 months	2
Early childhood: 3-5 years	7
Mixed ages	1
Relational assessment Tool	
Observational	4
Parent-report	16
Both	2
Type of Parent-Report Measure	
Parental attachment towards fetus	1
Parental attachment towards child	7
Parent-child interaction	15
Type of Observational Measure	
Child attachment pattern	1
Parent-child interaction	6
Parent Gender	
Mother	11
Father	0
Mothers and fathers	8
Not reported	3
Control Group	
Healthy control	13
Community norms	9

Table 2. Study Sample Characteristics.

Citation	Sample *(N)*	Child Age at Assessment (*Mean, SD, where provided*)	Child Sex *(% female)*	Cardiac Anomaly	Cardiac Complexity	Parent Age *(Mean, SD)*	Participation and Retention Rates *(P, R)*	Power Calculation
Parent-report measures								
Boztepe et al. 2016	200 mother-infant dyads • 50 CHD • 50 cleft lip and palate • 100 healthy controls	1-4 months (36%), 5-8 (40%), 9-12 (24%) 1-4 months (36%), 5-8 (36%), 9-12 (28%) 1-4 months (36%), 5-8 (28%), 9-12 (26%)	48% 58% 51%	-	Moderate: 70% Severe: 30%	19-30 years 46% 60% 75%	P: - R: 100%	Yes
Bright et al. 2013	63 father-infant dyads (CHD)	*M*=2.7 ±1.3 months (range 1.4-6.7 months)	46%	CoA (19%), HLHS/Norwood procedure (17%), VSD (13%), complex TGA (11%), TGA with intact ventricular septum (9.5%), ToF/PA with VSD (9.5%), PA with intact ventricular septum (6%), other (15%)	-	*M*=34.3 ±5.6 years (range 19.6-48.5)	P: 69% R: 72%	No
Jordan et al. 2014	97 mother-infant dyads (CHD)	*M*=3.4±1.4 months	47%	CoA (17%), HLHS/Norwood procedure (14%), TGA (12%), VSD (11%), TGA with intact VSD (10%), ToF/PA with VSD (9%), interrupted aortic arch/VSD (5%), PA with intact ventricular septum (5%), PDA (4%), TAPVD (4%), TA (2%), other (7%)	-	*M*=32.9 ±4.9 years	P: 84% R: 80%	No
Brosig et al. 2007	26 parent-child dyads (CHD)	*M*=4.7 years ±10 months (range 3-6 years)	27%	HLHS (50%), TGA (50%)	-	-	P: 45% R: 100%	No
Caris et al. 2016	459 parent-child dyads (CHD)	*M*=4.9±5.2 years	37%	HLHS (100%)	-	*M*=35.3 ±7.2 years	P: - R: 81%	No
De Stasio et al. 2018	100 mother-child dyads • 40 CHD • 34 preterm • 26 matched healthy controls	*M*=12 months of corrected age	46%	Atrioventricular canal defect, CoA, pulmonary stenosis, TGA, ToF, VSD	-	At birth: *M*=36 ±5.8 years *M*=35.2 ±4.2 years *M*=36 ±0.35 years	-	No
Fonseca et al. 2014	14 parent-infant dyads (CHD)	6 months	50%	-	-	Mother: *M*=33.3 ±4.9 years Father: *M*=35.9 ±5.7 years	P: 73% R: 82%	Yes
Golfenshtein et al. 2017, 2019	129 parent-infant dyads • 66 CHD • 63 healthy controls	3, 6, 9, 12 months	33% 37%	Single ventricle physiology post–operatively: 51%	Complex: 100%	-	P: 33% R: 54%	Yes
Hill et al. 2014	8 parent-infant dyads (CHD)	*M*=36 ± 23 months	63%	HLHS (n=4), PA with intact ventricular septum (n=2), AVSD (n=1), tricuspid atresia with interrupted aortic arch (n=1)	-	-	-	No
Majnemer et al. 2006	131 parent-child dyads (CHD)	*M*=5.4 years ±11.3 months	-	ToF (31%), TGA (27%), septal defects (12%), aortic arch anomaly (10%), other (e.g., DORV, TAPVD, SVCHD) (20%)	-	-	P: - R: 37% (Baseline) 49% (Follow-up)	No
Montis & Tumbarello 2011	54 parent-infant dyads (CHD) & matched healthy controls (-)	*M*=10 months (range 0-24)	-	-	Complex: 100%	Mother: *M*=34.5 years Father: *M*=38.5 years	-	No
Phipps & Drotar 1990	90 mother-infant dyads • 30 CHD • 30 home apnoea monitoring • 30 matched healthy controls	*M*=5.7 months ±=3.0 *M*=4.9 months ±=2.2 *M*=5.8 months ±=2.4	53% 57% 57%	Arrhythmia, pulmonary stenosis, ToF, VSD	Mild to moderate: 100%	*M*=28.9 ±5.1 years *M*=26.1 ±6.2 years *M*=27.8 ±4.5 years	P: 96%, 95%, - R: 64%, 68%	No
Re et al. 2018	25 mother-infant dyads (CHD)	*M*=2 months (range 2-5 months)	-	Single ventricle (7/22), biventricular (15/22)	Mild: 5% Moderate: 55% Severe: 40%	-	P: 73% R: 93%	No

Citation	Sample *(N)*	Child Age at Assessment *(Mean, SD)*	Child Gender *(% female)*	Cardiac Anomaly	Cardiac Complexity	Parent Age *(Mean, SD)*	Participation and Retention Rates *(P, R)*	Power Calculation
Parent-report measures								
Ruschel et al. 2014	197 expectant mothers • 96 with fetal cardiac diagnosis • 101 with clear fetal echocardiography scan	*M*=26.7±4 weeks gestation, and 30 days post-fetal echocardiography	-	VSD (47%), HLHS (8%), arrhythmia (7%), DORV (8%), ToF (4%), other (26%)	-	*M*=29.0 ±6.9 years *M*=27.6 ±6.4 years	P: 100% R: 8%	No
Sarajuuri et al. 2012	83 parent-child dyads • 37 CHD • 46 matched healthy controls	*Median*=18.3 months (range 17.2-23.1)	HLHS: 21% UVH: 54% Control: 35%	HLHS (62%), other functionally UVH defect (38%)	-	HLHS *Median*=29 years (range 18-40) UVH *Median*=30 years (range 17-40) Control *Median*=31 years (range 19-42)	P: 97% (HLHS), 65% (UVH) R: Mothers: 79-87% Fathers: 61-71%	No
Smith et al. 2017	48 parent-infant dyads (CHD)	*Median*=7.4 months (range 1.4-20.9)	-	ASD (37.5%), AVSD (22.5%), PDA (10%), CoA (7.5%), DORV (7.5%), aortic stenosis (2.5%), TGA (2.5%), TAPVD (2.5%), HLHS (2.5%), ToF (2.5%), TA (2.5%)	-	*Mother Median*=30 years (range 16-43)	P: - R: 83% (Baseline) 52% and 46% (3- and 6-month Follow-up)	No
Torowicz et al. 2010	129 mother-infant dyads • 69 CHD • 60 healthy controls	3 months	35%	Single ventricle physiology (48%), biventricular physiology (52%)	Complex: 100%	*M*=32.6 ±4.8 years (range 20-44)	-	No
Observational measures								
Carey et al. 2002	60 mother-child dyads • 30 CHD • 30 matched healthy controls	*M*=3.4 years ±0.9 (range 2-5 years)	43%	ToF (20%), HLHS (13%), aortic stenosis (7%), other (e.g., bicuspid aortic valve, tricuspid atresia, VSD) (60%).	Moderate: 7% Marked: 67% Severe: 26%	*M*=33.5 ±4.5 years *M*=32.1 ±4.2 years	P: 77% R: -	No
Darke & Goldberg 1994, Goldberg et al. 1990a, 1990b, 1991, 1995, Madigan et al. 2011	145 mother-child dyads[1] • 54 CHD • 40 cystic fibrosis • 51 matched healthy controls	3-6 months, 12-18 months, 24-30 months, and 7 years[1]	34%[3]	CoA, TGA, ToF, VSD	-	*Mother:* *M*=28.2 ±4.4 years *M*=27.4 ±5.0 years *M*=31.0 ±4.8 years	P: 70% (CHD), 85% (Cystic fibrosis) R: -	No
Harrison 2009, 2013, Harrison & Ferree 2014	31 mother-infant dyads • 15 CHD • 16 matched healthy controls	*M*=17.1 ±=3.9 days and 59.1 ±=4.4 *M*=17.4 ±=4.0 days and 59.3 ±=4.3 Whole sample: 3.1 years ±1.0 month	39-45% across timepoints	TGA (100%); additional VSD (n=6)	-	*M*=28.7 ±5.0 years *M*=29.7 ±5.8 years	P: - R: 65% (3 year Follow-up)	No
Laing et al. 2010	50 mother-child dyads (CHD)	18-36 months	30%	-	-	-	P: 57% R: 79%	No
Lobo 1992, Lobo & Michel 1995	20 mother-infant dyads • 10 CHD • 10 matched healthy controls	*M*=15.8 weeks ±=11.0 *M*=16.8 weeks ±=11.7	55%	VSD (30%), VSD with pulmonary stenosis (10%), VSD + congestive heart failure (10%), VSD + CoA (10%), ToF (10%), DORV + CoA (10%), TGA/hypoplastic right ventricle PA (10%), TGA/pulmonary stenosis (1%)	-	*M*=28.1 ±5.6 years *M*=30.3 ±5.0 years	-	No
Peçanha et al. 2015	37 mother-child dyads • 9 CHD • 17 asthma • 11 healthy controls	*M*=4.6 years ±=11 months *M*=4.2 years ±=10 months *M*=4.3 years ±=11 months	49%	-	-	*M*=34 ±5.5 years (range 29-40)	-	No

Note. [1]Total sample size and timing of assessment varied across the included papers. Numbers included refer to largest sample size included in Goldberg et al. (1995).
Abbreviations: ASD = Atrial Septal Defect, AVSD=Atrioventricular Septal Defect, CHD= Congenital Heart Disease, CoA= Coarctation of the Aorta, DORV = Double Outlet Right Ventricle, HLHS=Hypoplastic Left Heart Syndrome, NR= Not reported, PA=Pulmonary Atresia, PDA=Patent Ductus Arteriosus, SVCHD= Single Ventricle Congenital Heart Disease, TA=Tricuspid atresia, TAPVD = Total Anomalous Pulmonary Venous Drainage, TGA=Transposition of the Great Arteries, ToF=Tetralogy of Fallot, UVH= Univentricular Heart, VSD=Ventricular Septal Defect.

Table 3. Study Design Characteristics.

Citation	Country	Eligibility and Recruitment Setting		Study Design[1]	Measure(s)	Child Age at Assessment *(Mean, SD)*	Parent Informant/ Observed	Observational Setting[2]
		Cardiac Group	Control Group					
Parent-report measures								
Boztepe et al. 2016	Turkey	Infants pre- or postnatally diagnosed with moderate or severe CHD without any other congenital anomalies or acquired diseases, receiving treatment in a university hospital aged ≤ 12 months. Recruited from outpatient clinics of Hacettpe University Ihsan Dogramici Children's Hospital (Oct 2014-Oct 2015).	Healthy infants	CS	Maternal Attachment Inventory	*CHD:* 1-4 months (36%), 5-8 (40%), 9-12 (24%) *Control:* 1-4 months (36%), 5-8 (28%), 9-12 (26%)	Mother	-
Bright et al. 2013 Jordan et al. 2014	Australia	Infants were consecutive admissions for open or closed cardiac surgery to correct CHD within first 3 months of life, were medically stable, and within 2-months [65] or 4 weeks [72] post-discharge. Premature infants and those with chromosomal abnormalities excluded. Recruited from a tertiary paediatric hospital (Feb 2005-Sept 2006).	Community norms	CS	Paternal Postnatal Attachment Scale Maternal Postnatal Attachment Scale	2.7 months±1.3 (range: 1.4-6.7 months) 3.4 ±1.4 months	Father Mother	-
Brosig et al. 2007	USA	Consecutive surgical admissions for the Norwood procedure for HLHS or other CHD forms with significant obstruction to blood flow within the past 10 years, and a contemporary cohort of children who had an arterial switch operation for TGA, aged 3-6 years. Recruited from Children's Hospital of Wisconsin (1996-1999).	Community norms	CS	PSI-LF: Child Reinforces Parent and Parent Attachment subscales	4.7 years ±10 months (range: 3-6 years)	Parent	-
Caris et al. 2016	USA	Parents of infants with HLHS recruited through various Listservs and social network sites.	Community norms	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	4.9±5.2 years	Mothers (87%) & fathers (13%)	-
De Stasio et al. 2018	Italy	Infants with CHD had a favourable outcome, no known developmental delays, major neurological or neurosensory impairment (e.g., cerebral palsy), genetic syndromes, or further surgery planned, and were 12-months old. Recruited from neonatal ICU at a major tertiary neonatal unit in Rome, Bambino Gesu Children's Hospital	Healthy children born at term from two childcare units in Rome.	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	12 months of corrected-age	Mother	-
Fonseca et al. 2014	Portugal	Infants pre- or postnatally diagnosed CHD, without perinatal death, approximately one month after the disclosure of the diagnosis of CHD. Recruited from the Paediatric Cardiology Service of Pediatric Hospital at Centro Hospitalar de Coimbra (Sept 2009-Feb 2012).	Community norms	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	6 months	Mothers & fathers	-
Golfenshtein et al. 2017, 2019	USA	Infants had complex CHD requiring corrective or palliative cardiac surgery within the first 6 weeks of life, no other congenital anomalies or genetic syndromes (except 22q deletion or DiGeorge syndrome), born >35 weeks gestation and >2000 grams. Recruited from cardiac ICU of a children's hospital in mid-Atlantic America.	Healthy infants from hospital primary care practices.	L	PSI-LF: Child Reinforces Parent and Parent Attachment subscales	3, 6, 9, 12 months*	Parent	-
Hill et al. 2014	USA	Children with single ventricle CHD presenting to the Feeding, Swallowing and Nutrition Centre at the Children's Hospital of Wisconsin.	Community norms	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	36±23 months	Parent	-

Citation	Country	Eligibility and Recruitment Setting		Study Design[1]	Measure(s)	Child Age at Assessment *(Mean, SD)*	Parent Informant/ Observed	Observational Setting[2]
		Cardiac Group	Control Group					
Parent-report measures								
Majnemer et al. 2006	Canada	(i) Newborns with CHD requiring neonatal ICU admission and open-heart surgery in the first weeks of life, or (ii) infants <2 years old with CHD who had surgical repair in infancy, at Montreal Children's Hospital. Excluded infants with known risks for neurodevelopmental delay including: prematurity (<37 weeks gestation), small for gestational age, perinatal asphyxia, congenital brain malformations or genetic syndromes, previous open-heart surgery, or HLHS.	Community norms	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	5.4 years ±11.3 months	Mothers (82%) & fathers (18%)	-
Montis & Tumbarello 2011	Italy	Children had complex CHD, corrected only in some cases, for which they were placed under the prevention of syncytial virus.	Healthy children	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	10 months (range: 0-24)	Mothers (52%) & fathers (48%)	-
Phipps & Drotar 1990	USA	Infants had serious CHD warranting medical attention but not surgery prior to study entry. Outpatient cardiology clinics at cardiology division of Rainbow Babies and Children's Hospital and paediatric comprehensive care division of Cleveland Metropolitan General Hospital.	Healthy infants from ambulatory clinics at routine well-child visits and paediatric practices.	CS	PSI-LF: Child Reinforces Parent and Parent Attachment subscales	*CHD/Home Apnoea Monitoring/Control:* 5.7 months ±=3.0 4.9 months ±=2.2 5.8 months ±=2.4	Mother	-
Re et al. 2018	Australia	Infants had serious CHD, were clinically stable, born >36 weeks gestation, and had no known chromosomal, genetic, or syndromal disorder. Infants were admitted for cardiac surgery at the Royal Children's Hospital over a 10-month period. Mothers were aged 16-45 years.	Community norms	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale	2 months*	Mother	-
Ruschel et al. 2014	Brazil	All pregnant women who received a positive screen for fetal heart disease at the Fetal Cardiology Unit of Institute of Cardiology of Rio Grande do Sul (May 2008-Sept 2010), unless screening indicated a diagnosis with potential improvement of fetal status or diseases incompatible with life.	All pregnant women with a clear fetal echocardiography at the Fetal Cardiology Unit of Institute of Cardiology of Rio Grande do Sul (May 2008-Sept 2010).	L	Maternal-Fetal Attachment Scale	26.7±4 weeks gestation, and 30 days post-fetal echocardiography	Mother	-
Sarajuuri et al. 2012	Finland	Nation-wide sample of patients with HLHS or UVH who underwent paediatric cardiac surgery in Finland at the Children's Hospital of Helsinki University Central Hospital from Aug 2002-Feb 2005. Children had no known suspected chromosomal abnormalities.	Healthy newborns from low-risk deliveries	CS	PSI-LF: Child Reinforces Parent and Parent Attachment subscales	Median=18.3 months (range: 17.2-23.1)	Mothers & fathers	-
Smith et al. 2017	South Africa	Children with CHD <30 months. Excluded neonates, children who were critically ill, and those who had previous or emergency cardiac surgery. Recruited via consecutive sampling at Universitas Academic Hospital Paediatric Cardiology Unit over 17-month period.	Community norms	L	PSI-SF: Parent-Child Dysfunctional Interaction subscale	Median=7.4 months (range: 1.4-20.9)	Mothers & fathers	-
Torowicz et al. 2010	USA	Infants with CHD recruited from Cardiac ICU of the Children's Hospital of Philadelphia and were: full term (≥ 36 weeks gestation), ≥2500 grams at birth, had palliative or corrective surgery in first 6 weeks of life, and no other major congenital anomalies. Infants had no congenital or acquired anomalies (e.g., trisomies, gastrointestinal disorders, orofacial clefts, or neurological impairment).	Healthy infants	CS	PSI-LF: Child Reinforces Parent and Parent Attachment subscales	3 months*	Mother	-

Citation	Country	Eligibility and Recruitment Setting		Study Design[1]	Measure(s)	Child Age at Assessment *(Mean, SD)*	Parent Informant/ Observed	Observational Setting[2]
		Cardiac Group	Control Group					
Observational Measures								
Carey et al. 2002	USA	Infants had moderate to severe CHD aged 2-5 years, and were consecutive admissions from a large paediatric cardiology clinic at a children's hospital.	Infants had no chronic illness, significant health conditions, birth anomalies, were aged 2-5 years, and recruited in paediatric medical practices and school settings.	CS	PSI-SF: Parent-Child Dysfunctional Interaction subscale Dyadic Parent-Child Interaction Coding System	3.4 years ±0.9 (range: 2-5 years)	Mother	- Home Parent-directed play Child-directed play Clean-up
Darke & Goldberg 1994, Goldberg et al. 1990a, 1990b, 1991, 1995, Madigan et al. 2011	Canada	Infants had VSD, TGA, ToF, or CoA with a good prognosis either requiring surgical repair or spontaneous recovery expected, and no daily therapeutic responsibilities. Infants had no other handicaps, were expected for a 1-year follow-up, were <6-12 months at study entry, and had been discharged from hospital to home for at least 1 month. Recruited from inpatient and outpatient wards of the Hospital for Sick Children (1986-1990).	Healthy children from paediatric practices and medical walk-in clinics at the Hospital for Sick Children (1986-1990).	L	*1*. PSI-LF: Child Reinforces Parent and Parent Attachment subscales *2*. Author-developed greeting and play task *3*. Strange Situation Paradigm *4*. Puzzle tasks *5*. Atypical Maternal Behaviour Instrument for Assessment and Classification (AMBIANCE)	3-6 months 12-18 months 12-18 months 24-30 months 12-18 months and 7 years	Mother & father Father Mother Mother Mother	1. - 2. Laboratory Free play 3. Laboratory Free play Distress-inducing Separation and reunion 4. Laboratory Experimenter-induced play Problem-solving task 5. Laboratory Distress-inducing task Separation and reunion
Harrison 2009, 2013, Harrison & Ferree 2014	USA	Infants were born full-term, diagnosed prenatally or at birth with TGA and had no known co-morbidities, and completed assessments 7-14 days post-surgery, 6 weeks later, and at 3-years. Recruited from three major metropolitan children's hospitals over a 15-month period.	Full-term, healthy infants born at a large birth centre over a 15-month period.	L	Parent-Child Early Relational Assessment (P-CERA)	*Timepoints:* 17.1-17.4 days±4.0 59.1-59.3 days±4.4 3.1 years ±1 month	Mother	Hospital Feeding Problem-solving task Experimenter-directed play
Laing et al. 2010	Australia	Infants were admitted to neonatal ICU at the Children's Hospital at Westmead (June 2002-July 2004) for major life-saving surgery for CHD within the first 28 days of life, born > 33 gestational, and had no known neurological defects or genetic syndrome with developmental delay. Families resided within 200 km from the hospital.	Community norms	CS	National Institute of Child Health and Human Development (NICHD) Qualitative Scales of the Observational Ratings of Mother-Child Interaction	18-36 months*	Mother	Hospital Free play Teaching
Lobo 1992, Lobo & Michel 1995	USA	Infants <6 months, born full-term, lived at home, had no prolonged hospitalisation, and were being cared for in a paediatric cardiac clinic of a large children's hospital.	Full-term infants, <6 months, no major illness or surgery. Recruited through pediatric clinics and general community.	CS	Nursing Child Assessment Feeding Scale (NCAFS)	*CHD/Controls* 15.8 weeks±11.0 16.8 weeks±11.7	Mother	Laboratory Feeding
Peçanha et al. 2015	Brazil	Children had heart surgery at least 1 year prior, a clinically stable condition and no current or other medical problems. Recruited through hospital outpatient clinic.	Children with no chronic medical history, allergies, motor, physical or mental symptoms, or psychological care, from community paediatric clinic.	CS	Author-developed social interaction paradigm	*CHD /Controls* 4.6 years ±=11 months4.3 years ±=11 months	Mother	Laboratory Free play Distress-inducing paradigm Separation and reunion

Note. [1]*Study Design:* CS= Cross-sectional, L=Longitudinal.
[2]*Measures*: PSI-SF = Parenting Stress Index (Short-Form). PSI-LF= Parenting Stress Index (Long-Form).

Abbreviations: ASD = Atrial Septal Defect, AVSD=Atrioventricular Septal Defect, CHD= Congenital Heart Disease, CLP = Cleft lip and palate, CoA= Coarctation of the Aorta, DORV = Double Outlet Right Ventricle, HLHS=Hypoplastic Left Heart Syndrome, ICU=Intensive Care Unit,, NR= Not reported, PA=Pulmonary Atresia, PDA=Patent Ductus Arteriosus, TAPVD = Total Anomalous Pulmonary Venous Drainage, TGA=Transposition of the Great Arteries, ToF=Tetralogy of Fallot, VSD=Ventricular Septal Defect, UVH= Univentricular Heart.

Table 4. Summary of Study Results Comparing Cardiac Sample with Healthy Referents.

		Stronger or Better Outcomes in Cardiac Sample	No Difference in Outcomes between Cardiac and Control Sample	Weaker or Poorer Outcomes in Cardiac Sample
Parent-Report Measures	**Parental Attachment Towards Child**	• More positive maternal attachment following fetal CHD diagnosis [62] • More positive parental attachment to child with CHD aged 3-6 with HLHS [95].	• Maternal attachment towards infant at 3 months [72]. • Paternal attachment towards infant at 3 months [65]. • Parental attachment to child reported by: parents of infants at 3-, 6-, 9- and 12-months [67], mothers of 3-month [79] and 6-month-old infants [87], and mothers and fathers of toddlers at 18-months [81].	• Weaker maternal attachment towards 0-12 month old infant [64]. • Weaker paternal affection and pride, and pleasure in interaction at 3 months [65].
	Parenting Stress	• More positive reinforcement in interactions with their child aged 3-6 with transposition of the great arteries [95].	• Positive reinforcement in interaction, reported by: parents of infants at 3-, 6-, 9- and 12-months [67], mothers of 3-month [79] and 6-month old infants [87], mothers of 18-month old toddlers with univentricular heart defects, and fathers of 18-month old toddlers with univentricular heart defects and hypoplastic left heart syndrome [81]. • Dysfunctional parent-child interaction reported by: mothers and fathers of 10-month-old infants [77], mothers of infants at 12-months [78] and 3-years [82], and parents of 5-year old children [85].	• Positive reinforcement in interactions reported by: parents of toddlers aged 12-18 months [91], and mothers of toddlers with HLHS aged 18-months [81]. • Clinically concerning parent-child dysfunctional interaction reported by: parents of 5-year-old children [83], 8% of mothers of 2-month old infants [75], 15% of parents with 5-year-old children [85], 21% of mothers and 29% of fathers of 6-month old infants [66], 50% of parents of 3-year old children [84], and 55%, 44%, and 41% of parents of infants prior to cardiac surgery, at 3- and 6-months post-surgery [76].
Observational Measures	**Child Attachment**		• Prevalence of infant attachment patterns at 12-18 months including: secure, insecure-resistant, insecure-avoidant and insecure- disorganised strategies [93].	
	Parent-Child Interaction (Parent)	• (1) Higher maternal support, attunement and warmth, (2) positive affective involvement, sensitivity and responsiveness, and (3) better regulation of negative affect and behaviour, with their 2-week old but not 2-month old infant [69, 71]. • More maternal interaction in play with their 4-year-old child [86].	• Maternal sensitivity to cues, response to child distress, cognitive growth fostering, or total score whilst feeding their 4-month-old infant [73]. • Disrupted atypical maternal interaction in play at 12-18 months and 7-years [94]. • Positive father interaction (i.e., greeting position, greeting behaviour and latency, departure reaction, positioning, elaborativeness, sensitivity, comfort, mutuality) in play with their 12-18 month old child [89]. • Quality of maternal teaching (i.e., supportive presence, respect for autonomy, quality of assistance, confidence), or expressed hostility in play with child at 24-30 months [93]. • Positive (i.e., total number of questions, praise, physical positive behaviours) and negative maternal behaviour, (i.e., total criticisms, yells, whines, physical negative, destructive behaviours), or maternal commands in play with their 2-5-year-old [82]. • Maternal positive affective involvement, sensitivity and responsiveness, regulation of negative affect and behaviour, and anxiety and intrusiveness in play with 3-year-old [70]. • Maternal praise, responding to interaction, facilitating play, controlling play, and directiveness in play with their 4-year-old child [86].	• Lower maternal social-emotional growth fostering feeding their 4-month old infant [73]. • Lower maternal sensitivity and responsiveness to child distress and non-distress cues, lower stimulation of cognitive development, and less positive regard for their 18-36 month old child. Higher intrusiveness and more over-controlling behaviours, detachment and disengagement (i.e., lack of emotional or physical involvement), negative regard for the child, and more flatness of affect [80]. • More maternal task structuring and limit-setting in play with their 24-30 month old [93]. • Fewer maternal questions in play with their 4-year-old child [86].
	Parent-Child Interaction (Child)		• Potent distress cues (i.e., gaze aversion, back arching, time spent) during mother-infant feeding at 4 months [73]. • Positive affect, communication and social skills, and dysregulation or irritability during mother-infant feeding at 2 weeks or 2 months [71]. • Prosocial behaviour (i.e., infant social responsiveness, departure reaction, positive affect) in father-child play at 12-14 months [89]. • Negative mood in mother-child play at 18-36 months [80]. • Task involvement (i.e., enthusiasm, avoidance) or interaction with mother (i.e., affection, negativity) in mother-child play at 24-30 months [93]. • Positive (i.e., total questions, praise, physical positive behaviours) and negative (i.e., total criticisms, yells, whines, physical negative, destructive behaviours) behaviours, compliance or non-compliance in mother-child play at 2-5 years-old [82]. • Engagement, compliance, interaction and independent play with mother at 4-5 years [86].	• Lower clarity of cues and responsiveness to parent, and more subtle distress cues (i.e., hand to head behaviour, finger splaying) during mother-infant feeding at 4-months [73]. • Less positive mood, lower capacity for sustained attention with objects and non-social activities, less responsiveness and social connectedness with mother, and higher physical activity amongst children aged 18-36 months [80]. • Lower task involvement (i.e., persistence, compliance, positive experience). Greater dependence on mother in play at 24-30 months [93]. • Responded less to maternal questions in play at 4-years of age [86].
	Parent-Child Interaction (Dyadic)		• Dyadic mutuality and reciprocity, and dyadic tension between mother-infant dyads at 2 weeks or 2 months [71].	• Lower attunement, dyadic synchrony and contingency amongst mother-infant dyads at 4 months [73].

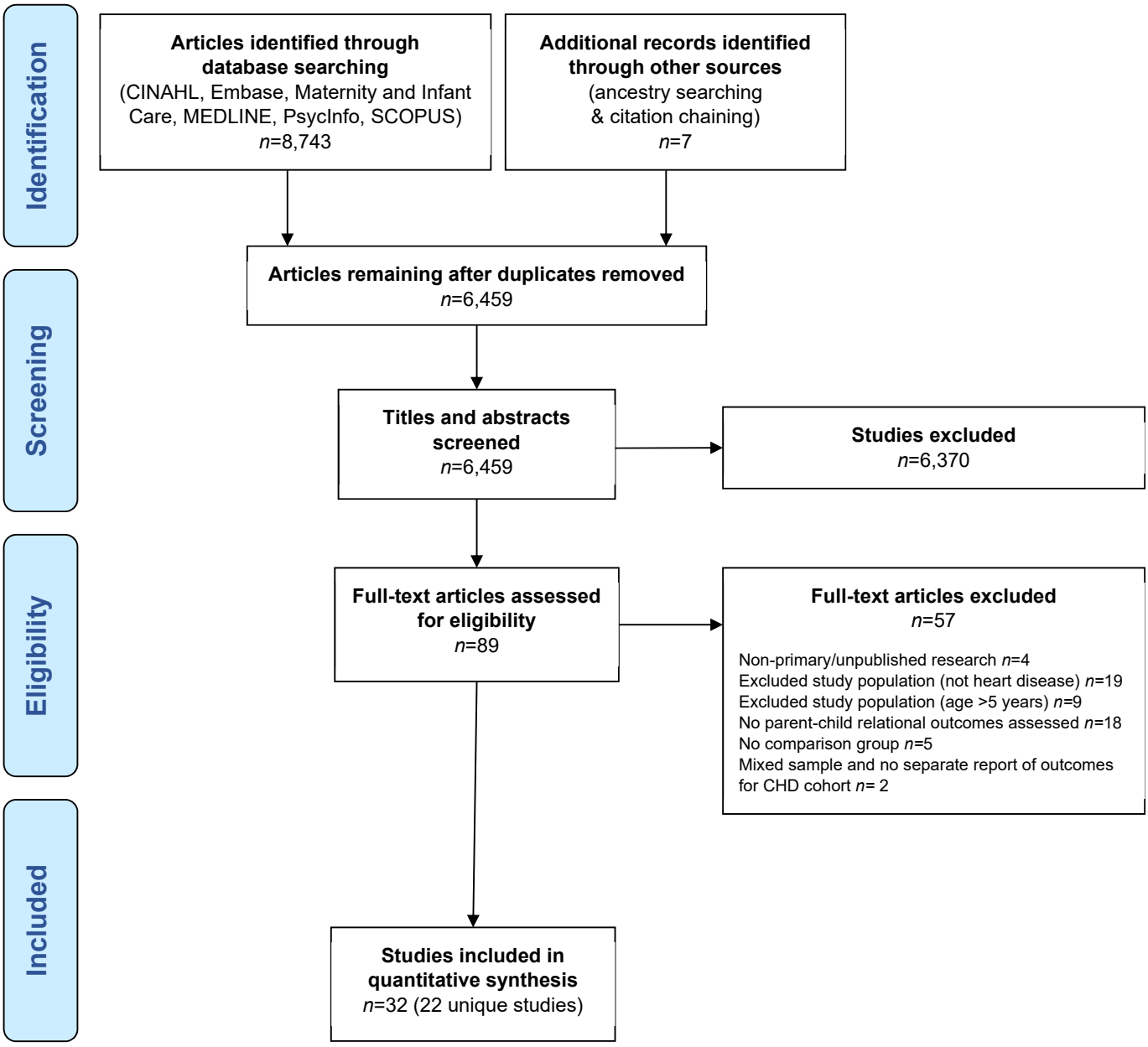


Figure 1. PRISMA Diagram of Systematic Review Process.

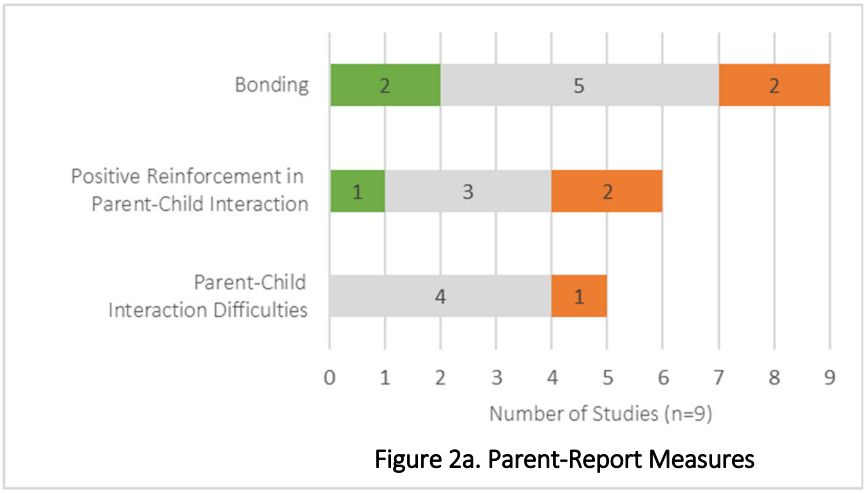


Figure 2a. Parent-Report Measures

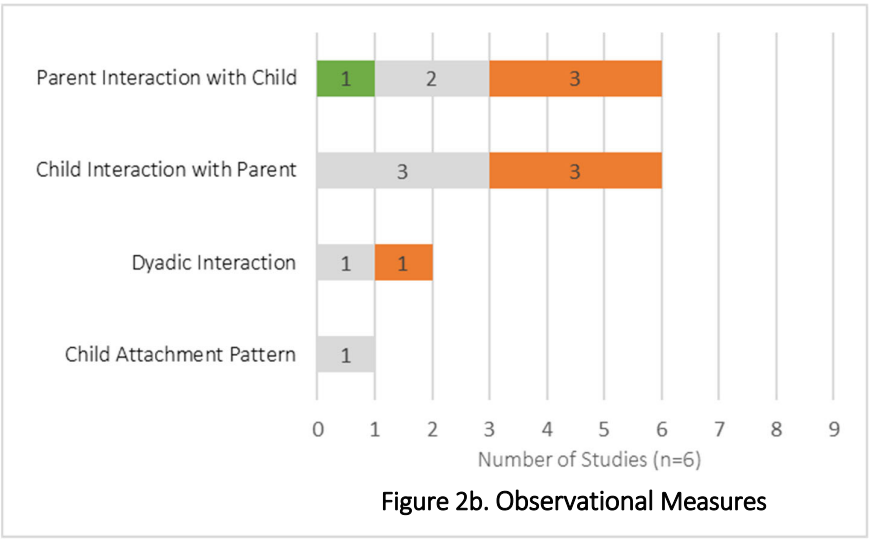
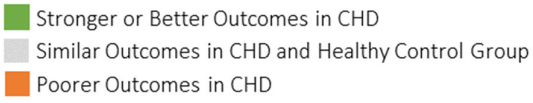


Figure 2b. Observational Measures



Figures 2a-b. Summary of relational outcomes reported for cardiac dyads across studies, compared with a healthy control group.

Supplementary Table 1. PRISMA Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	3
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	5-7
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	7
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	7
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	8-10
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	9
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Supplementary Table 2
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	9
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	9-10
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	8-9
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	10
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	10
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	-

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	-
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	-
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	10
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	Table 1, 2 and 3
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Table 2
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	Table 4
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	-
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	Table 3
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	-
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	18-23
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	23-24
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	18-27
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	2

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit: **www.prisma-statement.org**.

Supplementary Table 2. Literature search strategy used for the Medline database.

Topic	Keyword/MeSH Terms
Parent-fetal or parent-child relationship	exp Child Behavior/ OR exp Infant Care/ OR exp Maternal Behavior/ OR exp Maternal Deprivation/ OR exp Object Attachment/ OR exp Parent-Child Relations/ OR exp Paternal Behavior/ OR exp Paternal Deprivation/ OR exp Parenting/ OR exp Reactive Attachment Disorder/ OR ["attachment adj (avoidan* or behavio* or disorgani* or disord* or insecur* or pattern or resistan* or secur* or strateg*).mp] OR [((ante natal or antenatal or baby or babies or child* or dyad* or father* or infan* or mother* or maternal* or parent* or paternal* or pre natal or prenatal or post natal or postnatal or post partum or postpartum or toddler*) adj2 (attachment or bond* or communication or behavio* or interaction* or relation* or dyad* or attunement or contingency or coordination or matching or mirroring or mutuality or reciprocity or reparation or responsiv* or synchrony or sensitivity)).mp.] OR [(Strange situation) or (Attachment Q-Set) or (Attachment Q-Sort) or (AQS).mp]
Congenital heart disease	exp Heart Defects, Congenital/ OR exp Heart Diseases/cn OR exp Heart Valve Diseases/cn OR [(cardiac or heart) adj3 (anomal* or abnormalit* or congenital* or cyanotic or defect* or malform* or "single ventricle").mp] OR [(cardiac or heart) adj3 (fetal or foetal or foetus or fetus or prenatal or pre natal or prepartum or pre partum or ante natal or antenatal or newborn or neonat* or pediatric or paediatric or baby or babies or infan* or toddler* OR child).mp] OR congenital cardiovascular malformation*.mp OR exp Aortic Valve Stenosis/ OR aortic adj2 stenosis.mp OR Bicuspid aortic valve.mp OR (aortic or aorta) adj3 coarct*.mp OR Complete Atrioventricular Canal defect.mp OR exp Double Outlet Right Ventricle OR double adj3 ventricle.mp OR Ebstein's anomaly.mp OR interrupted aortic arch.mp OR exp Mitral Valve/ OR exp Pulmonary Valve Stenosis/ OR pulmonary adj2 stenosis.mp OR exp Pulmonary Atresia/ OR pulmonary atresia.mp OR (septal or septum) adj defect*.mp OR hypoplastic adj3 heart.mp OR tricuspid atresia.mp OR (tetralogy or trilogy) adj3 Fallot.mp OR total anomalous pulmonary venous connection.mp OR transpos* adj4 (arteries or artery or vessel*).mp OR exp Truncus Arteriosus/ OR truncus arteriosus.mp OR exp Cardiac Surgical Procedure/

Note. Similar search terms were used for all database searches.

Supplementary Table 3. Itemized and Total Methodological Rigour Scores, Presented by Study.

Study Citation	*QualSyst Items*											*Total Sum/ Total Possible Sum*	*Methodological Rigour Score and Category*	
	1. Objective sufficiently described	*2. Study design evident and appropriate*	*3. Method of subject/ comparison group selection described and appropriate*	*4. Subject and comparison group sufficiently described*	*8. Outcome measures well-defined and robust to measure-ment bias*	*9. Sample size appropriate*	*10. Analytic measures justified and reported*	*11. Estimate of variance reported for main results*	*12. Control for confounds*	*13. Results reported in sufficient detail*	*14. Conclusions reported*			
Boztepe et al. 2016	2	2	1	1	2	2	2	2	0	2	2	16/22	0.73	Good
Bright et al. 2013	2	2	2	2	1	2	1	2	1	2	2	19/22	0.86	Strong
Brosig et al. 2007	2	2	2	1	2	1	2	1	1	2	2	18/22	0.82	Strong
Carey et al. 2002	2	2	2	2	2	1	1	2	2	2	2	20/22	0.91	Strong
Caris et al. 2016	2	2	0	2	2	2	N/A	N/A	1	2	2	15/18	0.83	Strong
Darke & Goldberg, 1994	2	2	1	1	1	1	1	0	2	2	2	15/22	0.68	Adequate
deStasio et al. 2018	2	2	2	1	2	2	2	1	1	2	2	19/22	0.86	Strong
Fonseca et al. 2014	2	2	2	1	2	0	N/A	N/A	1	2	2	14/18	0.78	Good
Goldberg et al. 1990a	2	2	1	1	2	1	1	1	2	0	2	15/22	0.68	Adequate
Goldberg et al. 1990b	2	2	2	1	2	1	1	0	2	2	2	17/22	0.77	Good
Goldberg et al. 1991	2	2	2	1	2	2	1	1	2	1	2	18/22	0.82	Strong
Goldberg, 1994	2	2	2	1	2	2	1	1	2	2	2	19/22	0.86	Strong
Goldberg, 1995	2	2	2	1	2	1	2	0	1	2	2	17/22	0.77	Good
Golfenshtein et al. 2017	2	2	2	2	2	2	2	2	0	2	2	20/22	0.91	Strong
Harrison, 2009	2	2	1	2	1	0	2	1	1	2	2	16/22	0.73	Good
Harrison & Ferree, 2013	2	2	1	2	1	0	2	2	1	2	2	17/22	0.77	Good
Harrison, 2014	2	2	1	2	2	1	2	1	0	2	2	16/22	0.73	Good
Hill et al. 2014	2	2	1	0	2	0	N/A	N/A	1	2	2	12/18	0.67	Adequate
Jordan et al. 2014	2	2	2	2	2	2	2	2	1	2	2	21/22	0.95	Strong
Laing et al. 2010	2	2	2	1	2	2	2	2	1	2	2	20/22	0.91	Strong

Reference	*QualSyst Items*											*Total Sum/ Total Possible Sum*	*Methodological Rigour Score and Category*	
	1. Objective sufficiently described	*2. Study design evident and appropriate*	*3. Method of subject/ comparison group selection described and appropriate*	*4. Subject and comparison group sufficiently described*	*8. Outcome measures well-defined and robust to measure-ment bias*	*9. Sample size appropriate*	*10. Analytic measures justified ana reported*	*11. Estimate of variance reported for main results*	*12. Control for confounds*	*13. Results reported in sufficient detail*	*14. Conclusions reported*			
Lobo, 1992; Lobo & Michael, 1995	2	2	2	2	2	0	1	1	1	2	1	16/22	0.73	Good
Madigan et al. 2011	2	2	2	1	2	2	1	1	2	2	2	19/22	0.86	Strong
Majnemer et al. 2006	2	2	2	1	2	2	N/A	N/A	1	2	2	16/18	0.89	Strong
Montis & Tumbarello, 2011	1	2	0	1	2	1	0	1	1	2	2	13/22	0.59	Adequate
Pecahna et al. 2015	2	2	0	1	0	0	1	2	1	1	2	12/22	0.55	Adequate
Phipps & Drotar, 1990	2	2	2	1	2	1	2	2	1	2	2	19/22	0.86	Strong
Re et al. 2018	2	2	2	2	2	0	N/A	N/A	1	2	2	15/18	0.83	Strong
Ruschel et al. 2014	2	2	0	2	2	2	0	1	1	2	1	16/22	0.73	Good
Sarajuuri et al. 2012	2	2	2	1	2	1	2	1	1	2	2	18/22	0.82	Strong
Smith et al. 2017	2	2	2	1	2	2	N/A	N/A	1	2	2	16/18	0.89	Strong
Torowicz et al. 2010	2	2	1	1	2	2	2	2	1	2	2	19/22	0.86	Strong

Note. QualSyst Items 5–7 are excluded from this summary table, as these items are relevant for interventional studies only. Scores are calculated based on whether the article meets criteria for each weighted item; 'Yes' (score of 2; criterion adequately addressed), 'Partial' (1; partially addressed), 'No' (0; not addressed), or not applicable. Higher scores indicate greater methodological rigour (>0.8='Strong', 0.71-0.79='Good', 0.50-0.70='Adequate'; <0.50='Limited').

Supplementary Table 4. Additional Sample Medical and Demographic Characteristics for Cardiac Group only.

Citation	MEDICAL CHARACTERISTICS			PARENT AND FAMILY SOCIO-DEMOGRAPHIC CHARACTERISTICS		
	Birth Factors	**Cardiac Diagnosis, Comorbidities and Other Neurological Events**	**Treatment and Hospitalisation**	**Parental Education, Income, Socio-Economic Status and Employment**	**Marital Status**	**Other**
Self-Report Measures						
Boztepe et al. 2016	● Physical problems in pregnancy: 24% ● Psychological problems in pregnancy: 28%	●Time of diagnosis: Prenatal (20%), during birth (36%), postnatal (44%)	–	● ≤High school: 80% ●Job, working: 80% ●Household income <$300: 48%	–	● Nuclear family: 66%, ● Father support for infant care, always: 58%
Bright et al. 2013	*Paternal study:* ●Primiparous: 33%	*Paternal study* ● Acyanotic illness (57%), cyanotic (43%) ● Time of diagnosis: Prenatal (43%), postnatal (57%)	*Paternal study:* ● LOS, days: 1–14 (27%), 15–28 (32%), 29–42 (16%), 43+ (25%) ● Surgery: Corrective (65%), palliative (35%) ● Time home prior to first admission: *M*=10.3 ±20.9 days	*Paternal study* ●≤Secondary school: 64% ●University: 30% ●Main source of income, government benefit: 5%	*Paternal study:* ●NR	
Jordan et al. 2014	*Maternal study:* ● Primiparous: 41%	*Maternal study* ● Cyanotic illness: 46% ● Prenatal diagnosis: 45%	*Maternal study:* ● Age at discharge, days: Median=38, Range: 26–68 ● LOS: Median=25, Range: 15–43 ● Surgery type: Corrective (68%), palliative (32%)	●SES: *M*=4.4±1.1 (Range 1–7) *Maternal study* ●SES: *M*= 4.4±1.03	*Maternal study:* ● Married: 73%	*Maternal study:* ● Ethnicity, Australian–born: 82% ● English spoken at home: 96%
Brosig et al. 2007	–	–	*HLHS/TGA Median* ● Age at first surgery, days: 4 (range=2–15), 5 (range 1–64) ● CPB time, mins: 282 (range 143–673), 200 (range 168–259) ● LOS, first surgery: 26 (range 13–46), 16.5 (range 9–34) ● Deep hypothermic circulatory arrest time, mins: 65 (range 54–93), 14 ● Number of open–heart operations: 3 (range 1–4), 1 ● Surgery: Fontan (92% HLHS), arterial switch operation (100% TGA) ● Weight at first surgery, kg: 3.09 (range 1.8–4.6), 3.6 (range 2.4–4.4)	●Mean SES: Middle class	●Married: 92%	●Child ethnicity, Caucasian: 96%
Caris et al. 2016	●Prematurity, <37 weeks gestation: 7%	● Prenatal diagnosis: 70% ● Developmental or behavioral issues: 4% ● Gastric issues: 5% ● Genetic syndrome: 6% ● Pulmonary issues: 4% ● Other neurologic issues: 4% ● Seizures/stroke: 12%	● Gastronomy or nasogastric tube: 34% ● Home monitoring program: 71% ● Surgery: None (1%), Stage 1/Norwood/hybrid (9%), Stage II/Glenn/ comprehensive Stage II (44%), Stage III/Fontan (42%), transplant (4%) ● Tracheostomy: 2%	●High school: 26% ●Technical school: 7% ●College/university: 50% ● Graduate/professional: 16% ●Other: 1%	●Married: 83%	● Caregiver gender: father (13%), mother (87%) ● Race, White: 88.5% ● Children in family: *M*=2.3±=1.2
De Stasio et al. 2018	●Birth weight, grams: M=3076 ±631.5 ●Gestational age at birth: M=38 ±1.7 weeks ●Primiparous: 58%, 74%, 31%		● Length of hospitalisation, days: *M*=34 ±25.2	● High–school diploma: 40% ● University–degree: 38% ● Middle-school diploma: 22% ● SES: M=2.5, 2.0, 1.8	–	
Fonseca et al. 2014	● Primiparous: 21%	●Time of diagnosis: Prenatal (21%), postnatal (79%)	●Surgery: 64%	*Maternal* ● Education, ≤secondary: 36% ● Employed: 86% *Paternal* ● Education, ≤secondary: 100% ● Employed: 85%	● Married/ partnered 93%	
Golfenshtein et al. 2017, 2019	● Birth weight, grams: *M*=3299 ±475, ● Gestational age, weeks: *M*=38 ±1.23 ● Head circumference at 3–months (z–score): *M*=–.69 ±1.32 ● Length at 3–months (z–score): *M*=–.91 ±1.38 ● Weight at 3–months (z–score): *M*=–1.26 ±1.34	● Prenatal diagnosis: 75% ● Single ventricle physiology post–operatively: 51%	● Device–assisted feeding at discharge (nasogastric or gastric tube): 37% ● Device–assisted feeding at 3 months old: 10% ● LOS: Median=15 days (Range 2–159)	●College: 42%	–	● Mostly mothers ● Child ethnicity, non–Hispanic: 70% ● Child race, White: 89%
Hill et al. 2014	–	● Weight for age (z–score): *M*=–1.4±0.9	● Surgery: Post–Fontan (n=6), Glenn procedure (n=2) ● Gastronomy tube: 89% ● Choking, gagging or vomiting: 100%	–	–	–

Citation	MEDICAL CHARACTERISTICS			PARENT AND FAMILY SOCIO-DEMOGRAPHIC CHARACTERISTICS		
	Birth Factors	Cardiac Diagnosis, Comorbidities and Other Neurological Events	Treatment and Hospitalisation	Parental Education, Income, Socio-Economic Status and Employment	Marital Status	Other
Self-Report Measures						
Majnemer et al. 2006	● Primiparous: 50%	● Cyanotic lesion: 69%	● Age at surgery, months: M=2.6 ±4.2, range: 0.3–26.9, neonatal (41%) ● CPB time: M=151.6 mins ±50.3, range 76–301 ● Days in ICU: M=11.0 ±6.9, range 2–33 ● LOS: M=23.4 ±18.5, range 5–115 ● Deep hypothermic circulatory arrest: M=18.9 mins ±23.5, range 0–74 ● Open–heart surgery: 100% ● Oxygen saturations at preoperative neurological examination <85: 29.5% ● Surgery type: Corrective (94%), palliative (88%) ● Time since open–heart procedure: M=61.5 months ±10.1	–	–	● Foster parent (2%), biological parents (98%) ● Parent gender, mother: 82%
Montis & Tumbarello 2011	–		–	●High school: 45% ●Mean SES: Middle class	●Married: 99%	●Parent gender, mother: 52%
Phipps & Drotar 1990	●Primiparous: 50%	● Severity: Mild to moderate 100%	–	●≤ High school: 30% ●SES: I–III (57%), IV–V (44%)	–	●Race, White: 83%
Re et al. 2018	–	●Severity: Mild (1/22), moderate (12/22), severe (9/22) ●Physiology: Single ventricle (7/22), biventricular (15/22)	●Alarm Distress Baby Scale ≥ 5 (infant withdrawal): 11/22 ●Surgical outcome: Excellent (9/22), guarded (2/22), poor/palliative (11/22).	–	–	–
Ruschel et al. 2014	● Gestational age, weeks: M=26.7±4.2 ● Primiparous: 37% ● Loss of child: 21% ● Planned pregnancy: 48%	–	–	●≤ High school: 88% ●Income, minimum wages 3–6: 22%	● Partnered: 89%	
Sarajuuri et al. 2012	*HLHS/UVH Defect Medians* ● Birth weight, kgs: 3.52 (Range 2.61–4.29), 3.54 (Range 2.52–3.93) ● Maternal age at delivery: 29 (Range 18–40), 30 (Range 17–40)	● Neurological abnormality: HLHS 47%, UVH 3% ● Resuscitation: HLHS 11%, UVH 8%	● CPB time, mins: HLHS (Median=275, Range 221–497), UVH (Median=211, Range 65–351) ● Deep hypothermic cardiac arrest time, mins: HLHS (Median=5, Range 2–20), UVH (Median=8) ● Norwood I operation: HLHS=100%, UVH=38% ● Number of operations: HLHS (Median=2, Range 2–4), UVH (Median=2, Range 1–3)	● Maternal occupation: Median=4 (Range 2–9), Median=3 (Range 1–5)	–	–
Smith et al. 2017	–	● Severity: Mild (2.5%), moderate (75%), moderate to severe (20%), severe (2.5%) ● Down's syndrome: 25%	● Age at first cardiac surgery, months: Median=7.5 (Range 1.4–10.9) ● CPB: 68% ● Surgery: Definitive correction (74%), staged correction (13%), palliation (13%)	●M=Grade 9–11 ●Employed outside of home: 21% ●Low SES: 73%	–	●Primary caregiver, mother: 98%
Torowicz et al. 2010	–	● Severity: Complex CHD ● Physiology: single ventricle (48%), biventricular (52%)	● Growth failure: SV (45%), BV (22%) ● <5 medications: SV (67%), BV (89%) ● Repeated hospitalisation prior to follow–up visit: SV (33%), BV (22%) ● Surgery: 100% within first 6 weeks of life	●High school education: 94%	–	● Child ethnicity: Caucasian (72%), non–Hispanic (67%)
Observational Measures						
Carey et al. 2002	● First–born: 23%	● Severity: Moderate (7%), marked (67%), severe (26%) ● Time of diagnosis, first month of life: 87%	● Taking cardiac medications: 47% ● Total hospitalisations: M=2.4±1.15 (Range 0–5) ● Total days in hospital: M=30.6±22.1 (SD=0–106) ● Outpatient visits since diagnosis: M=12.5±4.8 (Range 4–28) ● Cardiac operations: M=2.1±1.15 (Range 0–5) ● Cardiac catheterization: M=1.6±1.1 (Range 0–5)	●≤High school: 23% ●Mother's employment, working outside home: 63%	●Married: 83%	● Child's ethnicity, Caucasian: 83% ● Parenting classes attended: 23% ● Social support, medium to high: 97%
Darke & Goldberg 1994, Goldberg et al. 1990a,1990b, 1991, 1995, Madigan et al. 2011	●Primiparous: 37%	–	● Heterogenous illness severity, treatment and prognosis ● Most infants had experienced planned surgeries at study entry	● Maternal education, years: *M*=13.7±2.8 ●Mother's occupation: *M*=6.5 ● Paternal education, years: *M*=13.8±3.1 ●Father's occupation: *M*=2.6-4.6	●Parent years together: M=4.8±3.1	–

Citation	MEDICAL CHARACTERISTICS			PARENT AND FAMILY SOCIO-DEMOGRAPHIC CHARACTERISTICS		
	Birth Factors	Cardiac Diagnosis, Comorbidities and Other Neurological Events	Treatment and Hospitalisation	Parental Education, Income, Socio-Economic Status and Employment	Marital Status	Other
Observational Measures						
Harrison 2009, 2013, Harrison and Ferree 2014	●Primiparous: 40%	–	● CPB time: M=175.9 mins ±48.8 ● Cross clamp time: M=102.3± 38.4 mins ● LOS: M=20.1 ±6.1 ● Days ventilated: M=4.5±1.2 ● Surgery timing, days: M=6.8 ±3.0, Range: 2–11 ● Subsequent hospitalisations by 3–years: 25%	●< Bachelor's degree: 53% ●Household income, $50,000+: 50%	●Married/ partnered: 100%	● Race/ethnicity, non–Hispanic White: 93%
Laing et al. 2010	● Singleton pregnancy: 96% ● Primiparous: 38% ● Full-term: 86% ● Small for gestational age: 2% ● APGAR 1, 8-10: 63% ● APGAR at 5 minutes, 8-10: 90% ● Gestational age: M=38.8 (SD=1.74, range: 34-41) ● Birth weight, grams: M=3390.22 (SD=668.45, range: 1695-5020)	● Developmental delay: 42%	●Age at time of first operation, days: M=5.06 (SD=5.3, range: 0-20) ● Open heart operation (60%), closed (40%) ● Number of operations: M=1.68 (SD=.82, range: 1-4) ● Hours on ventilation: M=106.1 (SD=76.68, range: 0-404) ● Days on oxygen: M=7.48 (SD=5.83, range: 0-25) ● Total length of stay, days: M=19.08 (SD=11.52, range: 1-64)	● Maternal education, HSC/TAFE: 56% ● Paternal education, HSC/TAFE: 48% ● Paternal occupation, professional: 55%	–	● Previous pregnancy losses: 94% ● Ethnicity, Caucasian: 90%
Lobo 1992, Lobo & Michel 1995	●Birth weight, grams: 3882.5±686.8	–	● Medications: Digoxin (60%), furosemide (10%), furosemide + metaproterenol sulphate (60%), vitamins (20%) ● Surgery or catheterisation: 33% ● Feeding: 6/10 bottle-fed, 2/10 solids, 1/10 breastfed, 1 bottle & solids.	●*M*=13.9 years ±2.1 ●Employed, yes: 40%	●Married, years: *M*=10	–
Peçanha et al. 2015	–	–	● Surgery at least one year prior: 100%	●*M*=15 years ●SES, middle income: 100%	–	–

Note. Abbreviations: APGAR= Appearance, Pulse, Grimace, Activity, and Respiration Score, ASD = Atrial Septal Defect, AVSD=Atrioventricular Septal Defect, BV = Biventricular physiology, CHD= Congenital Heart Disease, CoA= Coarctation of the Aorta, CPB=Cardiopulmonary bypass, DORV = Double Outlet Right Ventricle, HLHS=Hypoplastic Left Heart Syndrome, LOS=Length of hospital stay, Med= Median, NR= Not reported, PA=Pulmonary Atresia, PDA=Patent Ductus Arteriosus, SES= Socio-economic status, SV=Single ventricle physiology, TAPVD = Total Anomalous Pulmonary Venous Drainage, TGA=Transposition of the Great Arteries, ToF=Tetralogy of Fallot, VSD=Ventricular Septal Defect, UVH= Univentricular Heart.