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Title	Longitudinal associations between family factors and the neurodevelopmental and psychosocial outcomes of children with congenital heart disease: A systematic review
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Publication date	2026-07-31
Original Citation	McCusker, C, Phillips, L & Murphy, S 2026, 'Longitudinal associations between family factors and the neurodevelopmental and psychosocial outcomes of children with congenital heart disease: A systematic review', Early Human Development, vol. 222, 106627, pp. 1-12. https://doi.org/10.1016/j.earlhumdev.2026.106627
Type of publication	Review
Link to publisher's version	https://www.scopus.com/pages/publications/105046176328 - 10.1016/j.earlhumdev.2026.106627
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Download date	2026-11-17 04:48:22
Item downloaded from	https://hdl.handle.net/10468/19203

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Early Human Development 222 (2026) 106627

Contents lists available at ScienceDirect

Early Human Development

journal homepage: www.elsevier.com/locate/earlhumdev

Longitudinal associations between family factors and the neurodevelopmental and psychosocial outcomes of children with congenital heart disease: A systematic review

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ARTICLE INFO

Keywords:
Congenital heart disease
Family processes
Child outcomes

ABSTRACT

Family factors have been gaining increased attention in understanding adverse neurodevelopmental and psychosocial outcomes for children with congenital heart disease (CHD). To clarify relevance, we undertook a systematic review of only longitudinal studies which assessed such associations. Comparisons with the contribution of disease/surgical factors were also made where included studies considered such. We included longitudinal studies which assessed dynamic family factors (e.g. parent mental health, attachment, family functioning) and later child outcomes. Searches were conducted across *CINAHL, Medline-Pubmed, PsychInfo and SCOPUS Web of Science*. The NIH Quality Assessment Tool was used to evaluate study quality and risk of bias. Eighteen studies, utilizing data from 11 study samples and 2109 participants, met inclusion criteria. These studies included samples from infancy, with follow-up periods stretching into young adulthood, and with various degrees of CHD severity. The quality of studies was "good" to "fair", with key limitations of attrition and limited sociocultural diversity in samples. Findings suggested that family factors predicted later child psychosocial outcomes and more consistently than severity of disease indicators. This contrasted with a much smaller number of studies examining family factors and child neurodevelopmental outcomes, where no reliable conclusions could be reached. Findings highlight the importance of screening and family focused interventions for this population.

1. Introduction

Children with congenital heart disease (CHD) are at elevated risk for adverse neurodevelopmental and psychosocial outcomes such as motor and language delays, executive function deficits, social difficulties, and behavioral difficulties [1,2]. Historically, most attention has been directed at examining outcomes for children with CHD as they relate to disease and surgical variables. For instance, neurological correlates, cyanosis, cardiopulmonary bypass and severity have all been negatively associated with neurodevelopmental and psychosocial outcomes in this population [1,3]. However, a systematic review by Drakouli et al. [3] suggested that family factors including parental support, sense of coherence and mental health, whilst implicated, had been less systematically investigated. Research in developmental psychology has long demonstrated the importance of family factors such as parent psychopathology, parenting style, parent-child attachment and family functioning for child psychosocial outcomes [4]. Such associations have also been reported for children with chronic illness in general [5,6] and so it may be expected that such are also implicated in outcomes for children with CHD.

In this review, we were specifically interested in those family factors which are dynamic and potentially modifiable (e.g. parenting style), rather than structural and more difficult to modify (e.g. socioeconomic status). Schleider and Weisz [7] define such family factors as: parent-level factors, (e.g. parental mental health); dyad-level factors, which are localised within parent-child relationships or sibling relationships (e.g. parenting stress and attachment); and family-level factors, which concerns a family's level of functioning as a collective whole (e.g family cohesion and family stability). These processes are themselves known to be adversely affected by childhood CHD, with elevated levels of distress, worry, anxiety, depression and post-traumatic stress features common among parents [8]. A 13-year longitudinal study by Ehrler et al. [9] also found that 25% of mothers of children with CHD reported chronically low mental health-related quality of life throughout their child's development. Additionally, elevated levels of behavioral and internalizing problems have been reported in siblings of children with CHD, and

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https://doi.org/10.1016/j.earlhumdev.2026.106627
Received 23 April 2026; Received in revised form 20 July 2026; Accepted 21 July 2026
Available online 24 July 2026
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general family processes such as cohesion, communication and financial strain have been reported to be negatively affected [10]. Such impacts might be expected to compromise coping and parenting capacity and thus have a negative impact on outcomes for the child with CHD.

Thus, McCusker et al. [11] found parenting style, marital status and maternal mental health difficulties were associated with child behavioral outcomes, with greater effect sizes than disease or surgical variables, although the data were cross-sectional. Capitello et al. [12] also found that measures of mother and father anxiety and depression predicted specific domains of health-related quality of life (e.g. treatment anxiety; communication difficulties) among children and adolescents with a CHD diagnosis. Parenting stress, when the child was 8 years old, predicted clinician ratings of global psychosocial functioning at age 16 [13]. Although research is limited, associations between parenting stress and mental health and neurodevelopmental outcomes have also been reported [2,11]. In a recent cross-sectional study of adolescents with CHD [14], better parental mental health and family functioning was identified among participants with a *"favourable"* neurodevelopmental profile (i.e. those scoring in the normal range across various cognitive outcomes) compared to those with a *"vulnerable"* profile. However, the consistency with which such family factors predict outcomes in longitudinal research designs remains equivocal.

Drawing conclusions about the relative contribution of disease factors versus family factors in outcomes for children with CHD has been complicated. The systematic inclusion of family factors in CHD outcome studies is relatively recent, and methodologies have varied in which constructs to measure, and how to measure them. Diagnostic samples have varied, control groups have often been lacking and the developmental stage of the cohort investigated has varied without sufficient attention to the relevance of such. Much of the research has been cross-sectional where the issue of respondent bias is significant. The *Cardiac Neurodevelopmental Outcome Collaborative* have called for more robust, theoretically grounded and longitudinal studies in this area, to elucidate which parent and family factors are associated with which child neurodevelopmental and psychosocial outcomes across time [15]. We sought to systematically review the existing base of prospective studies which have addressed these questions.

We included only longitudinal studies where there had been a gap (of at least 6 months) between measurement of family factors and later child outcomes. Our primary research question was: Do family factors predict neurodevelopmental and psychosocial outcomes for children with CHD across time, and which factors show the most consistent associations? Most of the included studies also examined the impact of disease factors, and our secondary question related to the relative contribution of both sets of factors, where such comparisons were possible. As far as we are aware, this is the first systematic review of this nature.

2. Methodology

We followed the methodological approach outlined by the *Preferred Reporting Items for Systematic Reviews & Meta-Analyses* [16]. A review protocol was developed a-priori and registered on PROSPERO (Prospero registration: CRD42022331024).

2.1. Study inclusion criteria

Studies were included in the review based on the following criteria:

- Studies used a prospective design. Studies included a minimum of two waves of data collection, with at least a six-month interval, and where dynamic family factors (as outlined above) were assessed at a wave prior to psychosocial and/or neurodevelopmental child outcomes. We chose six months as a likely reasonable period within which putative influences between predictor and outcome factors might be expected to operate.
- The sample was comprised of families with dependent children with clearly identified congenital heart disease (not acquired), who were under 18 years of age at the first wave of data collection.
- Studies examined the role of one or more dynamic family factors (i.e. parent-level factors, dyad-level factors, or family-level factors, as per Schleider and Weisz [7]). Parent-level factors included variables such as parent mental health and the quality of the marital relationship; dyad-level factors pertained to the parent-child relationship such as parenting stress and parenting style; family-level factors related to systemic processes within the family as a whole, such as family adaptability and cohesion. Such factors had to be identified as independent, predictor or moderator / mediator variables on dependent / outcome variables.
- Dependent variables included child neurodevelopmental (e.g. cognitive functioning; language skills; motor functioning) and psychosocial (e.g. behavioral adjustment; psychological wellbeing) outcomes.
- Predictors and outcomes were measured using standardized instruments with acceptable psychometric properties, reflected in reported reliability and validity analyses.
- Only studies written and published in English in a peer-reviewed academic journal were included.

Qualitative studies and studies that did not report on direct associations between family factors and child outcomes were excluded. Studies of children with genetic syndromes (e.g. Down Syndrome) or diagnosed neurodevelopmental co-morbidity (e.g. Autism Spectrum Disorder) were also excluded as such co-morbid conditions would likely also influence outcomes. Conference proceedings and academic dissertations were not included.

2.2. Search strategy

CINAHL, Medline-Pubmed, PsychInfo and SCOPUS Web of Science were searched. Search terms, developed through a scoping literature review, are outlined in Table 1. The title and abstract fields of our chosen databases were searched using these terms. No limits were applied to publication dates. Reference lists of included studies were manually screened for studies not detected during database searches. Searches

Table 1
Boolean search terms.

Population	Child* OR Teenage* OR Adolescen* OR Youth* OR Pediatric* OR Pediatric* OR infant* AND "congenital heart disease" OR "congenital heart defect*" OR CHD OR "tetralogy of Fallot" OR "transposition of the great arteries" OR "hypoplastic left heart syndrome" OR "cyanotic" OR "ventricular septal defect" OR "atrial septal defect" OR "patent ductus arteriosus" OR "atrioventricular septal defect" OR "coarctation of the aorta" OR "tricuspid atresia"
Phenomena	"family function*" OR "family factor*" OR parent* OR maternal OR mother* OR paternal OR father* OR sibling* OR "family environment" OR famil* OR "family cohesion" OR "family system*" OR "family process*" OR "family predictor*" OR "family context" OR "parent child relations*" OR "parent child interaction*" OR "family relations*" OR "family interaction*" OR "parent stress*" OR "parent mental health" OR "parent infant relations*" OR "family impact" OR "parenting style*"
Outcome	Neurodevelopment* OR "cognitive outcome*" OR OR "intelligence" OR "IQ" OR "executive function*" OR "language" OR "memory" OR "motor" OR "quality of life" OR "health related quality of life" OR "psychological outcome*" OR adjustment OR behav* OR emotion* OR "psychosocial outcome*" OR psychopathology OR psychiatric OR internalis* OR Externalis* OR "mental health" OR anx* OR depress* OR "psychological wellbeing" OR externaliz* OR internaliz* OR "externalizing problem*" OR "internalizing problem*" OR "emotional outcome*" OR "behavior problem*" OR "well-being"
Design	prospective OR longitudinal

were conducted in July 2026.

2.3. Data screening

A total of 384 records were retrieved when duplicates were removed (see Fig. 1). The first and third authors screened all titles and abstracts at the first stage (CMC; SM). A total of 314 records were excluded at this stage. Full texts of the remaining 70 studies were reviewed by all authors. There was 95% consensus, and disagreements were resolved by discussion. Eighteen papers met inclusion criteria. Some papers relied on the same baseline datasets, reporting various longitudinal waves in different papers. Thus, although 18 papers were identified, there were 11 study samples included for review. Excluded studies from the full-text review stage, and the reason for exclusion, are documented in Fig. 1. Most papers excluded did not operationalize family factors as predictor variables or did not assess child outcomes.

2.4. Quality assessment

Quality assessment of included studies was conducted using the 14 item *NIH Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies* [17]. This tool evaluates the internal validity of studies and includes various indicators of risk of bias – e.g. of selection bias, information bias, measurement bias or confounding bias [17]. Item 12 was removed as it only pertained to intervention studies. The total maximum score for each study was therefore 13. For this review, a score of 11–13 was rated as "good", a score of 8–10 was rated as "fair" and a score of 7 or below was rated as "poor". Quality assessment of studies was conducted for all papers by all authors independently and there was a concordance rate of 71%, with divergence ranging from 1 to 3 points only. Discrepancies were addressed through discussion and consensus was reached.

2.5. Data extraction and synthesis

Relevant data pertaining to child and family characteristics (e.g sample size, mean participant age, racial and ethnic groups, disease severity), study informants, designs, family and child measures and key findings were extracted using a bespoke data extraction form. Data related to the 3 levels of family factors (parent, dyad and family processes), as defined above, were typically reported by parents, although some direct assessment occurred in some studies (e.g. of the attachment relationship). Data related to the 2 domains of child outcomes (psychosocial and neurodevelopmental) were based on parent, child and teacher reports as well as direct assessment (e.g. of neurodevelopmental functioning). In the data extraction process, predictor and outcome measures were extracted if the authors determined that these measures, as used in the original studies, fell into the above family factor and child outcome domains. The specific measures used across studies are noted in the legend to Table 3. As noted above, although disease severity was not a search criterion, almost all studies included disease factors in their analyses. Therefore, these data points were also extracted from the studies. Definitions of disease severity varied from clinical ratings of severity, diagnostic groupings (e.g. corrected -v- palliated conditions) or even overall CHD -v- control group comparisons. Study data were extracted by the second and third authors (LP; SM) and validated by the first author (CMC). Due to the heterogeneity of family and child measures, study designs, and statistical data across the included studies, a meta-analysis was not appropriate. For instance, statistical data related to associations varied in terms of univariate, multivariate or interactional effect sizes, and in the use of categorical or interval data associations. A narrative synthesis organised by child outcomes (i.e. psychosocial and neurodevelopmental) is thus provided and supported by reference to statistical information such as effect sizes (as available or discernible).

3. Results

3.1. Study characteristics

In terms of quality, the included studies were rated as either "good" ($n = 9$) or "fair" (n = 9) with none falling in the "poor" range. The NIH Quality Assessment scores for each study, and a breakdown of these scores, are outlined in Table 2.

Authors generally provided detailed account of population samples, inclusion and exclusion criteria, described some effect sizes and provided sufficient time between exposure and outcomes. In terms of key limitations, most studies did not report power calculations. Additionally, attrition rates greater than 20% were evident in 9 of the 18 studies, and we deemed insufficient attention was given to potential confounding variables in 7 of the 18 studies.

Table 3 provides a summary of study features, key outcomes and

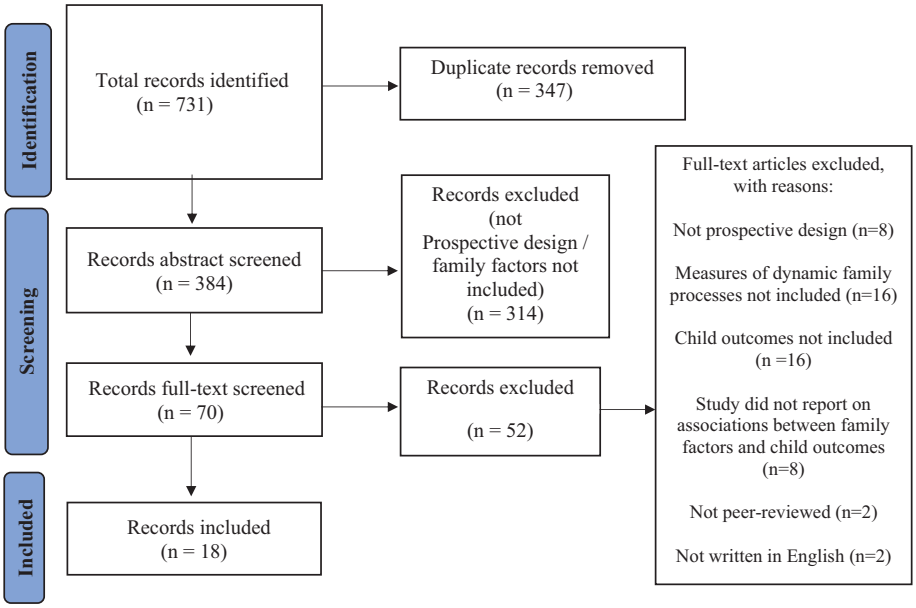

Fig. 1. Review Flow Chart.

Table 2
NIH quality assessment scores.

	Berant et al. [18]	Brandlistuen et al. [19]	Ehrler et al. [20]	Goldberg et al. [21]	Goldberg et al. [22]	Goldberg et al. [23]	Heller et al. [24]	Herrup et al. [25]	Landolt et al. [26]	Lang et al. [27]	Luyckx et al. [28]	Luyckx et al. [29]	Majnemer et al. [30]	McCusker et al. [31]	Oris et al. [32]	Rassart et al. [33]	Visconti et al. [34]	Werninger et al. [35]
1. Was the research question or objective in this paper clearly stated?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
2. Was the study population clearly specified and defined?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
3. Was the participation rate of eligible persons at least 50%?	1	1	1	1	1	1	1	1	0	1	1	1	0	1	1	1	1	1
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period?)	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
5. Was a sample size justification or power description provided?	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
6. For the analyses in this paper, were the predictors of interest measured prior to the outcomes being measured?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
7. Was the time frame sufficient?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
8. for the predictors that can vary in level, did the study examine different levels of the exposure as related to the outcomes?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
9. Were the predictor measures clearly defined, valid, reliable and implemented consistently across participants?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
10. Were the predictor(s) assessed more than once over time?	1	0	0	1	1	0	0	0	1	1	1	1	0	1	0	1	1	0
11. Were the outcome measures clearly defined, valid, reliable and implemented consistently across participants?	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
12. Was loss to follow-up after baseline 20% or less (for at least one follow up measurement?)	1	1	1	1	0	0	1	0	1	0	1	1	0	0	0	0	1	0
13. Were key potential confounding variables measured and adjusted statistically?	0	1	1	1	0	0	0	1	0	0	1	1	1	1	1	1	0	1
Total scores	11	11	11	12	11	9	10	10	10	10	12	12	9	11	10	11	11	10

Table 3
Study characteristics and synthesis of findings.

Authors; country; study design	Sample characteristics	Family / child factors assessed	Main findings	Disease impact / additional findings
Berant et al. [18]; Israel; 2 waves separated by 7 years; No Control Group.	60 dyads; All married parents of Jewish origin. Mean age at baseline = 3 months; Mean age at follow up = 7 years; CHD mixed severity (e.g. VSD, TGA).	Family: Mother's attachment style[1] Child: Self-concept[2] Child emotional wellbeing[3]	Mother's avoidant-attachment and attachment anxiety predicted child self-concept [avoidance ($\beta = -0.29$, $p < .01$); anxiety ($\beta = -0.38$, $p < .01$)] @ 7 years. Mother's avoidant-attachment also predicted child self-image ($\beta = -0.45$, $p < .01$), dominant affect ($\beta = -0.44$, $p < .01$), and low-level defences ($\beta = 0.29$, $p \leq 0.05$) @ 7 years. Mother's attachment anxiety predicted child interpersonal anxieties ($\beta = 0.39$, $p < .01$) @ 7 years.	CHD severity did not independently predict child self-concept. However, the impact of mother's attachment anxiety was amplified in those dyads with more severe CHD.
Goldberg Studies (3)				
Goldberg et al. [21]; Canada; 2 waves separated by 12–18 months; Healthy control group.	42 CHD dyads; Largely middle-class parent sample with mean of 13 years in education; Mean child age at baseline = 1.7 months; Mean child age at follow up = 13.7 months; Mixed severity CHD (e.g. VSD, Coarctations, TOF).	Family: Parenting stress[4] Psychological distress[5] Child: Attachment style[6] Mental & motor development [7] Infant temperament[8]	Parenting stress or distress at child age 1.7 months did not predict infant outcomes 12–18 months later, with respect to infant attachment, temperament or mental & motor development.	CHD group less likely to be securely attached than controls ($p < .01$).
Goldberg et al. [23]: Canada; 3 waves spanning 3 years (12, 24 and 36 months); 3 comparison groups; CHD, CF$^{\Delta}$ and healthy controls.	145 dyads; CF$^{\Delta}$ ($n = 40$), CHD ($n = 54$), healthy ($n = 51$); Largely middle-class parent sample with mean of 13 years in education; Mean child age at baseline = 12 months Mean child age at final follow up = 3 years.	Family: Infant-mother attachment[6] Child: Child problem behavior[9]	In CHD sample, an avoidant infant-mother attachment relationship at 12 months was associated with higher externalizing (Cohen's d = 0.60 $p < .05$) and internalizing child behavior scores (Cohen's d = 1.13, $p < 01$) @ 3 years.	Compared to other groups, CHD group had higher scores on internalizing subscale of CBCL but not externalizing or total behavior problem scores.
Goldberg et al. [22]; Canada; 4 waves spanning 4 years (12–48 months); 3 comparison groups; CHD, CF$^{\Delta}$ and healthy controls	137 dyads - CF$^{\Delta}$ ($n = 47$) or CHD ($n = 48$), healthy ($n = 42$); Largely middle-class parent sample with mean of 13 years in education; Mean age at baseline = 12 months; Mean age at final follow-up = 4 years	Family: Attachment[6] Parenting Stress[4] Child: Child Problem Behavior[9]	Parenting stress significantly predicted both internalizing and total child behavior scores in each teacher year and at new 4 year follow-up. Combined scores (R^2 range 0.15–0.29) at $p < .01$. At 4 years, parenting stress from baseline was the only significant predictor of total behavior problems ($R^2 = 0.23$, $p < .001$). Parent-child attachment at Baseline did not statistically contribute to child behavioral outcomes at 4 years.	Illness severity at Baseline predicted internalizing scores at 24 months ($R^2 = 0.04$, $p < .05$) and in combined scores 4 years ($R^2 = 0.04$, $p < .01$). Child temperament at Baseline predicted internalizing scores at 24 months and 4 years.
Heller et al. [24]; Canada; 2 waves separated by 12 months; 3 comparison groups - Cleft Pallet (n = 42), CHD ($n = 38$), hearing impairment ($n = 50$).	130 dyads; 32% parents educated beyond high school; 66% "medium or low" socioeconomic status; Mixed CHD severity (specific diagnoses not specified); Reported age range at baseline 4–13 years. No reported age range at follow up	Family: Family Functioning[10] Child: Child Problem Behavior[9] Child Psychological Symptoms[11]	No statistically significant association between family functioning scores at Baseline and child behavior outcomes / psychological symptoms 12 months later.	CHD group higher levels of behavior maladjustment.
Lang et al. [27]; Germany; 2 waves spanning 7 years from primary school age to adolescence; Healthy control group	24 dyads; Mean age at Baseline = 7.3; Mean age at follow-up = 12.4; All children with an isolated VSD; 33.3% of mothers and 29.2% of fathers were highly educated.	Family: Maternal Anxiety[23] Maternal Proactive Parenting Behaviour[33] Child: Cognitive Performance[34] Language Competencies[34] Depressive symptoms[35] Anxiety[35] ADHD[35] Health-Related Quality of Life[36]	Higher maternal anxiety at Baseline was significantly associated with higher depression symptoms ($r = 0.645$, $p < .005$) and higher anxiety symptoms ($r = 0.475$, $p < .05$) at follow-up. No statistically significant associations found between maternal factors at Baseline and cognitive performance / language competencies / ADHD / HRQOL at follow-up.	Disease related factors were not included in models or statistically controlled.
iDetach Studies (3)				
Luyckx et al. [28]; Belgium; 2 waves separated by 9 months; No control group	429 adolescents with CHD; 23% lone parents; Mixed severity CHD (classified as simple – complex); Mean age at Baseline = 15.8 years; Mean age at follow up = 16.6 years	Family: Parental Support [12] Child: Quality of Life[13] Perceived Health Status[14] Coping[15]	Parental support at Baseline predicted quality of life ($r = 0.13$, $p < .01$) and adolescent coping ($r = 0.09$, $p < .05$) 9 months later, while controlling for disease complexity.	Adolescents with more complex disease pathology scored lower on quality of life at Baseline. Perceived health status at Baseline predicted quality of life 9 months later ($r = 0.12$, $p < .05$).

(*continued on next page*)

Table 3 (*continued*)

Authors; country; study design	Sample characteristics	Family / child factors assessed	Main findings	Disease impact / additional findings
Luyckx et al. [29]; Belgium 3 waves spanning 18 months from Baseline (15.8 years); No control group	429 adolescents with CHD; 23% lone parents; Mixed severity CHD (classified as simple – complex). Mean age at Baseline = 15.8 years Mean age at final follow = 17.3 years	Family: Parental Support [12] Child: Quality of Life[13] Perceived Health Status[14] Depression[16] Loneliness[17]	Paternal support at Baseline (15.8 years) predicted increases in perceived health status ($r = 0.07$) and, quality of life ($r = 0.08$) at 17.3 years. Paternal support at Baseline predicted decreases in depressive symptoms ($r = -0.06$) and decreases in loneliness ($r = -0.08$) at 17.3 years, while controlling for disease complexity.	Adolescent depressive symptoms predicted decreases in perceived health & quality of life. Perceived health status at Baseline predicted quality of life at second, 9-month follow-up ($r = 0.16$).
Oris et al. [32]; Belgium; 4 waves spanning 36 months from Baseline (15.8 years); Included healthy control group	398 adolescents with CHD; 25% lone parents; Mean age at baseline = 15.8 years; Mean age at final follow-up = 18 years.	Family: Parental Support [12] Child: Depression[16] Wellbeing[13, 18]	High scores on parental support at 15.8 years significantly associated with lower depressive symptoms scores at 18 years.	No associations between child outcomes and disease complexity were noted.
Rassart et al. [33]; Belgium; 4 waves spanning 36 months from Baseline (16.3 years); No control group	429 adolescents with CHD; Mixed severity CHD (classified as simple-complex); Mean age at baseline = 16.3 years; Mean age at follow-up not reported.	Family: Parenting Style [12] Child: Perceived Health[18] (i.e. physical, emotional, social, and school-related)	Trajectories of perceived health among adolescents were significantly predicted by both maternal [X^2 (2) = 43.43, $p < .005$] and paternal [X^2 (10) = 40.65, $p < .005$] parenting styles. Adolescents classified in the democratic parenting class reported the most favourable perceived health outcomes over time. Adolescents classified in the authoritarian, overprotective, and psychologically controlling parenting classes displayed an increased risk for persistently poor perceived health.	No significant associations found between disease complexity and perceived health.
Majnemer et al. [30]; Canada; 2 waves separated by 5 years; No control group	94 dyads; Mixed CHD severity (e.g. VSD, TGA excluding hypoplastic left heart syndrome); Mean age at baseline not reported. Mean age at follow up = 64.2 ± 11.3 (months)	Family: Parenting Stress[4] Child: Child Problem Behavior[9] Cognitive Performance [19] Receptive Language [20] Functional Outcomes [21,22]	Parenting stress at Baseline significantly associated (R^2 range 0.12–0.23; $p \leq .01$) with lower IQ scores, poor receptive language skills, socialization and adaptive behavior at 5 year follow-up.	Disease and surgical factors not associated with later behavioral difficulties. Disease factors significantly associated with cognitive & language outcomes ($p < .04$). There were also some specific associations between cyanosis, and time of surgery, in this sample with children who underwent later surgery and who had acyanotic lesions having poorer developmental outcomes.
McCusker et al. [31]; UK; 2 waves separated by 6 years; Sibling control group for some analyses.	40 dyads; Mean age at Baseline = 2.8–8.6 months; Mean age at follow up = 7.7 years; Mixed CHD severity (e.g. VSD, TGA, TOF, ASD).	Family: Maternal Mental Health[23] Maternal Worry[24] Family Composition Family Functioning[25] Child: Child Problem Behaviors[9] Child Competencies[9]	Maternal mental health ($\beta = 0.357$), family cohesion ($\beta = -0.162$) & lone parent status ($\beta = -0.0282$) at Baseline predicted total child behavior problems at 7 years. Adjusted $R^2 = 0.331$, F (5.79), $p = .004$. *Teacher reported outcomes:* Family cohesion at Baseline predicted behavior problems at 7 years ($\beta = -0.453$). Maternal mental health at Baseline predicted child adaptive functioning at 7 years ($\beta = -0.355$)	Child "Competence" outcomes predicted by disease & surgical factors - length of time on bypass ($\beta = -0.524$), Adjusted $r^2 = 0.396$, F (3.46) $p = .046$. *Teacher reported outcomes:* Open heart surgery (β presence of syndrome (β = -0.273) at Baseline, predicted child behavior problems at 7 years.
MoBa Studies (2)				
Brandlistuen et al. [19]; Norway; 3 waves – Baseline (age 6 months), 18 and 36 month follow-ups. Healthy control Group	175 dyads in the CHD group; Mothers' mean years in education = 14.5; Mean age at baseline = 6 months Mean age at final follow up = 36 months; Mixed range of CHD severity – defined as "mild – moderate" or "severe".	Family: Psychological Distress[26] Child: Communication[27] Pro-Social Impairments[28]	Psychological distress in mothers at child aged 6 months was not predictive of level of developmental impairments at 18 and 36 months.	Severe CHD had x2 higher odds for any developmental impairment compared to controls, whereas mild/moderate group did not differ significantly to controls. Surgical variables *NS for developmental impairments. Levels of developmental impairments consistent across each time periods.
Landolt et al. [26]; Norway; 3 waves – Baseline at age 6 months, 18 and 36 month follow-ups; Healthy Control Group.	408 dyads; Mothers' mean years in education = 14.5; Mean age at baseline = 6 months; Mean age at final follow up = 36 months. Mixed range of CHD severity from "simple" to "severe".	Family: Psychological Distress[26] Child: Child Behavior[9, 29]	Maternal distress at 18 months significantly predicted child's internalizing problems at 36 months ($\beta = 0.10, p < .05$) but not vice versa.	The CHD group, compared to the healthy control group, showed higher levels of internalizing behavior at all assessment points.
Herrup et al. [25]; USA; 2 waves spanning 20 years from Baseline (4 years) to	202 dyads; Mean age at Baseline = 4 years; Mean age at follow-up = 22.3 years; All children with CHD who underwent cardiopulmonary bypass	Family: Parenting Stress[4] Child Mental Health Related	Parenting stress at Baseline predicted poorer resilience among young adults at follow-up ($R^2 = 0.28$; $p < .05$). No association was	Cardiac diagnosis was statistically controlled for in model, but no significant association was reported.

(*continued on next page*)

Table 3 (*continued*)

Authors; country; study design	Sample characteristics	Family / child factors assessed	Main findings	Disease impact / additional findings
young adulthood; No control group	surgery, with or without deep hypothermic circulatory arrest; Child participants were 54% male; 80% White.	Quality of Life[37] Resilience[38]	found between parenting stress and mental health-related quality of life among young adults at follow-up.	
Visconti et al. [34]; USA; 2 waves separated by 4 years; Compared to standardized normative data.	158 dyads; 90% married; 89% white; Good spread in levels of educational attainment and socioeconomic status; Mean age at baseline = 1 year; Mean age at follow up = 4 years. Only TGA diagnoses of CHD,	Family: Social Support[30] Parenting Stress[4] Child: Child Behavior[9]	Correlational analysis - Parenting stress at Baseline associated with total behavior problems $r = 0.31$, $p \leq .001$, internalizing scores $r = 0.23$ $p \leq .01$ and externalizing scores r = 0.23, p ≤ .01 at 4 years. Cross-sectional associations between same pairs stronger in final regression model.	Illness related factors not included in models or statistically controlled.
Zurich Studies (2) Werninger et al. [35]; Switzerland; 2 waves separated by 6 years No control group	125 dyads with CHD only; Mean age at Baseline = 4 years; Mean age at follow up = 10.2 years; Sample underwent cardiopulmonary bypass surgery.	Family: Maternal Mental Health[23] Child: Behavior[28] Social Responsiveness[31] ADHD symptoms[32]	Maternal distress at Baseline significantly predicted behavior difficulties, social responsiveness and ADHD symptoms problems at 10 years. R^2 range from 0.17 to 0.23 ($p < .01$).	Disease and surgical factors did not predict behavior difficulties at 10 years.
Ehrler et al. [20]; Switzerland; 4 waves – Baseline at 4 years. 6, 10, and 13 years follow-ups; Compared to matched normative data.	137 dyads; Mean age at Baseline = 4 years; Mean age at final follow-up = 13.7 years; All children with complex CHD who underwent cardiopulmonary bypass surgery; Child participants 64% male	Family: Maternal Mental Health[23] Child: Child Behavior[28]	Better maternal mental health at baseline significantly predicted normal behavior across ages ($R^2 = 0.21$; $p < .01$).	Disease and surgical factors did not predict behavioral trajectories across ages.

[1] Adult-Attachment Style Scale [40].
[2] Tennessee Self-Concept Scale [41].
[3] Children's Apperception Test [42].
[4] Parenting Stress Index [37].
[5] Rutter Health Questionnaire [43].
[6] The Strange Situation Test [36].
[7] Mental Development Index & Psychomotor Development Index from Bayley Scales of Infant Development-II [38].
[8] Revised Infant Temperament Questionnaire [44].
[9] Achenbach Child Behavior Check List [39].
[10] Family Functioning Index [45].
[11] Children Self-Report Psychiatric Scale [46].
[12] Child Report of Parent Behavior Inventory -responsiveness subscale [47].
[13] Quality of Life Linear Analogue Scale [48].
[14] Perceived Health Status Linear Analogue Scale [28].
[15] Orientation to Life Questionnaire [49].
[16] Center for Epidemiologic Studies Depression Scale [50].
[17] UCLA Loneliness Scale [51].
[18] Pediatric Quality of Life Inventory [52].
[19] Wechsler Preschool & Primary Scale of Intelligence [53].
[20] Peabody Picture Vocabulary Test [54].
[21] Vineland Adaptive Behavior Scale [55].
[22] Peabody Developmental Motor Scales [56].
[23] Brief Symptom Inventory [57].
[24] Maternal Worry Scale [58].
[25] Family Environment Scale [59].
[26] Hopkins Symptom Checklist [60].
[27] Ages & Stages Questionnaire [61].
[28] Strengths & Difficulties Questionnaire [62].
[29] Infant Characteristic Questionnaire [63].
[30] Social Support Network Inventory [64].
[31] Social Responsiveness Scale [65].
[32] Conners-3 Short form [66].
[33] Alabama Parenting Questionnaire [67].
[34] Wechsler Intelligence Scale for Children - Fifth Edition [68].
[35] Diagnostic System for Psychiatric Disorders [69].
[36] Revised Quality of Life Questionnaire [70].
[37] Short-Form Health Survey [71].
[38] Resilience Scale [72].
[*] NS = non significant.
[Δ] CF = cystic fibrosis.

some secondary findings of relevance. The contributions of severity of disease / illness and family factors to child outcomes were included in most studies and are summarized in Table 3. Whilst associations between both sets of factors and child outcomes are reported, it was not always possible to determine which had the greater or lesser impact in any given study.

Studies originated from Europe and North America. Study completion dates ranged from 1985 to 2023. Sample sizes ranged from 40 to 429 with total $n = 2109$. Racial, ethnic and socioeconomic characteristics of the samples are reported in Table 3, where these were evident in the original papers. Data related to such sample characteristics were typically sparse, but where available, suggested limited demographic diversity in samples. The time between the collection of predictor and outcome measures ranged from 6 months to 11 years. Ten studies used two waves of data collection, four studies used three waves of data collection, and four studies had four waves. The age range of participants with CHD ranged from infancy to young adulthood.

Fourteen studies used parent report measures, and four studies used adolescent reports of family factors and child outcomes. However, three studies also used observational protocols to assess family factors (e.g. *Strange Situation Procedure* [36]). Four studies used direct measurement of child outcomes (e.g neurodevelopmental assessment) and one study included teacher reports of child outcomes.

Measures of family factors varied throughout studies as outlined in Table 3, however, parenting stress as measured by the *Parenting Stress Index* [37] and measures of parental psychological distress were used most frequently. Child outcome measures included standardized and validated neurodevelopmental measures (e.g. *Bayley Scales of Infant Development* [38]), as well as measures of psychosocial and behavioral functioning (e.g. *Child Behavior Check List* [39]).

3.2. Overview of outcomes

Of the 18 studies, which examined longitudinal associations between family factors and child outcomes, 15 reported statistically significant associations with effect sizes ranging from small to large. Table 3 summarizes detailed findings for each study. In Table 4, we present a categorical synthesis of data to enable patterns to be more apparent. For each study, Table 4 categorises the relevant predictor and outcome variables in relation to the three family factor domains (i.e. parent-level; dyad-level; family-level) and two child outcome domains (i.e. psychosocial; neurodevelopmental) defined in our review. Using the Goldberg et al. [23] study as an example, infant-mother attachment has been categorised as a dyad-level family factor, and child problem behavior has been categorised as a child psychosocial outcome. Table 4 then indicates whether each of the three levels of family factors, and severity of disease factors, were or were not associated with each of the two levels of child outcomes when such were included in analyses.

3.3. Psychosocial outcomes

Seventeen studies explored prospective associations between family factors and child psychosocial outcomes (see Tables 3 and 4). Of these studies, 15 demonstrated statistically significant direct effects of family factors for child psychosocial outcomes, with small to large effect sizes as per Cohen [73] criteria.

Six studies examined the relationship between parent level factors and later child psychosocial outcomes. All found significant associations with effect sizes ranging from small to medium [18,20,27,30,32,35]. Thus, for example, the mother's attachment style (measured when the child was aged one) predicted scores on a child emotional well-being scale and child self-concept scale at age seven years [18]. Although disease severity did not independently contribute to child psychosocial outcomes in this study, the authors noted that the effect of maternal attachment style was greatest in children with more severe disease. This suggests interactional effects between the factors studied. Among younger populations, maternal psychological distress, measured when the child was 18 months, contributed to child internalizing difficulties at age 3 years [26]. Similar trends were observed among older cohorts where maternal mental health, when the child was 4 years, predicted specific behavior problems at age 10, including social interaction difficulties and ADHD related symptoms [35]. The McCusker et al. [31] study also demonstrated that psychosocial outcomes in middle childhood were primarily associated with family factors (maternal mental health and family cohesion) assessed in infancy, rather than severity of disease. In this study, the authors reported that family cohesion accounted for 33% of the variance in later behavioral outcomes. These findings included both teacher and parent reports of child behavioral outcomes.

Eleven studies examined prospective associations between dyad-level factors and child psychosocial outcomes. Nine of the eleven studies reported significant associations, with small to large effect sizes. Thus, the parent-child relationship was shown to be associated with later behavioral problems, where infants with CHD who demonstrated an avoidant attachment style with the caretaker at 12–18 months, displayed greater externalizing and internalizing scores on the Child Behavior Checklist at age three years compared to controls [23]. Four papers reported significant associations between parenting stress and later child behavioral outcomes [22,25,30,34]. Higher levels of

Table 4
Summary of prospective associations between family processes, disease factors and child outcomes.

Study (date) and child outcome(s) measured	Parent level factors	Dyad level factors	Family level factors	Disease factors
Berant et al. (2008) [18]				
- Psychosocial	✓	N/A	N/A	X
Brandlistuen et al. (2011) [19]				
- Neurodevelopmental	X	N/A	N/A	✓
Ehrler et al. (2026) [20]				
- Psychosocial	✓	N/A	N/A	X
Goldberg et al. (1991) [21]				
- Psychosocial	X	X	N/A	✓
- Neurodevelopmental	X	X	N/A	X
Goldberg et al. (1995) [23]				
- Psychosocial	N/A	✓	N/A	✓
Goldberg et al. (1997) [22]				
- Psychosocial	N/A	✓	N/A	✓
Heller et al. (1985) [24]				
- Psychosocial	N/A	N/A	X	✓
Herrup et al. (2026) [25]				
- Psychosocial	N/A	✓	N/A	X
Landolt et al. (2014) [26]				
- Psychosocial	✓	N/A	N/A	✓
Lang et al. (2022) [27]				
- Psychosocial	✓	X	N/A	N/A
- Neurodevelopmental	X	X	N/A	N/A
Luyckx et al. (2012) [28]				
- Psychosocial	N/A	✓	N/A	N/A
Luyckx et al. (2014) [29]				
- Psychosocial	N/A	✓	N/A	N/A
Majenmer et al. (2008) [30]				
- Psychosocial	N/A	✓	N/A	X
- Neurodevelopmental	N/A	✓	N/A	✓
McCusker et al. (2013) [31]				
- Psychosocial	✓	N/A	✓	✓
Oris et al. (2016) [32]				
- Psychosocial	N/A	✓	N/A	X
Rassart et al. (2014) [33]				
- Psychosocial	N/A	✓	N/A	X
Visconti et al. (2002) [34]				
- Psychosocial	N/A	✓	N/A	N/A
Werninger et al. (2020) [35]				
- Psychosocial	✓	N/A	N/A	X

✓ = was found to predict later child outcomes; X = was measured but was not found to predict later child outcomes; N/A = was not measured.

parenting stress, reported when the infant was aged one, were associated with increased behavior problems reported when the child was 4 years [34]. Goldberg and colleagues [22] also found that parenting stress at age one significantly predicted child behavioral outcomes at age 4. Indeed, whilst both parenting stress and measures of illness severity (as rated by the study clinician) predicted later child behavior outcomes, the variance explained by parenting stress was greater than that of illness severity in this study (see Table 3).

Additionally, in a study by Herrup et al. [25], higher parenting stress reported at age four significantly predicted lower levels of resilience among participants in young adulthood (mean age = 22.3 years old). Among cohorts of adolescents with CHD, perceived parental support, as rated by the adolescents, significantly predicted their scores on measures of coping, wellbeing, and quality of life 9 months later, while statistically controlling for disease severity [28]. Stability coefficients were reported as "moderate". Similarly, paternal support significantly predicted later loneliness, depression and quality of life among this cohort at an 18-month follow-up period, and more so than perceived health status and internalizing difficulties [29]. Additionally, Lang et al. [27] reported that higher maternal anxiety at age 4 predicted higher depression and anxiety symptoms among adolescents, while Ehrler et al. [20] found that better maternal mental health at age 4 predicted fewer behavioral difficulties across childhood and adolescence. At the family-level, teacher reports of behavioral outcomes in the school setting at 7 years were predicted by parent reports of cohesive family functioning in the first year of life, with a moderate effect size [31].

When disease factors were explored in relation to psychosocial outcomes, conclusions were more variable. Fourteen studies examined these associations, but only 6 reported significant associations (see Tables 3 and 4). Goldberg et al. [22] for example, found that illness severity (assessed in infancy) significantly predicted internalizing difficulties at age 4, although effect sizes were smaller than the family factors also included in this study (see Table 3). The direction of effects of illness severity warrants noting, as those with less severe illness demonstrated greater behavioral problems in early years. By contrast, disease and surgical factors were found to be non-significant in in their associations with child behavioral outcomes at age five [30] and age ten [35].

3.4. Neurodevelopmental outcomes

Four studies explored prospective associations between family factors and neurodevelopmental outcomes (see Tables 3 and 4). Of these, only one reported a significant association. Majnemer and colleagues [30] found that parenting stress was significantly associated with lower IQ scores, poor receptive language skills, socialization and adaptive behavior among 5-year-olds with CHD. However, Goldberg et al. [21] found that parenting stress and parent-child attachment in infancy were not significantly associated with child developmental outcomes in a 12-month follow-up of children with CHD. Similarly, maternal distress was found to be non-significant in predicting developmental attainments of 3-year-olds with severe CHD [19]. Among adolescents, Lang et al. [27] found that maternal factors at age 4 were not significantly associated with either cognitive performance or language skills at follow-up (mean age = 12.4).

Disease (CHD severity) and surgical (hypothermic circulatory arrest time) factors however, were associated with later neurodevelopmental impairments related to communication and language, motor functioning, intellectual ability and functional behavior [19,30].

4. Discussion

A striking finding from this systematic review was that only 18 studies met our inclusion criteria, across several decades of research into outcomes for children with CHD. Within longitudinal study designs, attention to family factors in determining psychosocial and neurodevelopmental outcomes lags behind attention to the impact of disease severity and other medical factors. This was especially apparent with respect to objectively measured neurodevelopmental outcomes. This supports calls in the literature for concerted focus on these factors and in prospective studies [3,15].

The primary finding from studies reviewed was that of 18 longitudinal studies, 15 reported significant associations between at least one domain of family functioning and psychosocial and neurodevelopmental outcomes for children with CHD. Such associations were particularly evident across the studies with respect to psychosocial outcomes. Only four studies examined associations between family factors and child neurodevelopmental outcomes. However, three of these studies yielded insignificant results, suggesting that family factors could be less commonly implicated in neurodevelopmental outcomes.

These findings suggest that among children with CHD, family factors warrant further attention if we want to improve psychosocial outcomes for this population. Even when illness related variables were included in predictive models, family factors often manifested greater effect size associations for later psychosocial outcomes [18,20,22,31,35]. This is noteworthy given the traditional emphasis on disease / surgical determinants of psychosocial and neurodevelopmental outcomes [3]. However, this is consistent with research in other pediatric conditions, where the importance of family factors and, by implication, family-focused interventions has become increasingly emphasized [5].

In terms of which specific family factors were important, in the current systematic review, parenting stress, perceived parental support, maternal mental health, maternal worry and the attachment relationship were documented as important predictors of psychosocial outcomes including emotional wellbeing [18,27,29,32], resilience [25], coping [28] and adolescent-reported quality of life [28,29]. This was often demonstrated in studies where disease severity was controlled for [28,29,32] or where effect sizes were compared [18,22,26,31,35]. These associations appeared to be greatest in cohorts of later childhood / adolescent samples and/or with an increasing passage of time.

Conversely, we saw less consistent evidence for associations between disease / surgical factors and psychosocial outcomes. Within 11 of the 14 studies that included both, the effects of disease/surgical factors were either non-significant or had smaller effect sizes than family factors (see Tables 3 and 4). Our review did suggest some caveats to this conclusion. Disease factors appeared to be more commonly associated with psychosocial outcomes in younger cohorts. However, the direction of the association was not always as expected - as in Goldberg's [22] study, where those with less severe disease ratings demonstrated greater behavioral difficulties. This highlights that the impact of disease severity may not be linear. Further, our review has suggested that disease factors may sometimes interact with family factors in predicting psychosocial outcomes.

Although the number of studies which examined it in a longitudinal design was small, there was less convincing evidence for an association between family factors and neurodevelopmental outcomes. Three of the four prospective studies which examined this relationship did not find such associations. Rather, disease / surgical factors appeared more important for later neurodevelopmental outcomes [19,21,30]. However, it must be acknowledged that as only four studies (published across a period of three decades) examined these associations, firm conclusions cannot yet be drawn from this research. Furthermore, there is a particular dearth of literature examining associations between family factors and objective measures of neurodevelopmental outcomes, as only two studies in the corpus assessed these outcomes through direct neurodevelopmental assessments [27,30]. Therefore, further investigation of these specific associations would appear warranted, given likely links established in other pediatric populations between parent mental health (and other family factors) on later neurodevelopmental outcomes [74].

4.1. Strengths and limitations of studies reviewed

The studies included in this review demonstrated a number of

strengths – prospective designs (up to 11 years between predictor and outcome assessment), robust sample sizes, standardized study measures, and frequent use of control groups. Studies did not solely rely on parental informants, with a number of studies incorporating teacher reports and direct assessment of attachment behaviors and neurodevelopmental functioning.

A common limitation of such longitudinal research relates to attrition rates, which may introduce bias as well as reducing statistical power to detect associations. Some studies in this review addressed attrition rates by highlighting the absence of any attrition bias and using statistical methods to estimate missing values [28,29,32]. However, such approaches to attrition were not reported across all studies included in our review.

Furthermore, as the majority of studies included in this review relied on parent-report measures, the potential impact of shared method variance [75] must be considered in relation to the research findings. As this use of parent-report measures by the researchers has meant that the independent and dependent variables were assessed using a single source of information, this may have contributed to the relevant associations identified between family factors and child outcomes within these studies. However, the additional use of teacher reports [31] and direct assessment [21,23,27,30] by certain researchers mitigates the risk of shared method variance in these cases.

A variable approach to the measurement of disease severity, or defining included diagnostic categories, was evident. This compromises the basis on which the impact of disease or illness factors can be understood. Additionally, a range of statistical models, including linear regressions, ANCOVAs, and latent class growth analysis, were implemented across the studies included in this review. This variation in statistical approaches may have contributed to the differential impacts of family versus disease factors across these studies.

Other limitations included the small number of studies which evaluated family factors in relation to neurodevelopmental outcomes, and the lack of sociodemographic diversity in sample characteristics when such were reported. Moreover, parent level factors tended to be maternal factors, with few studies reporting on paternal mental health or paternal parenting stress, for example.

4.2. Limitations of the current review and future recommendations

Given the relatively small number of studies reviewed we did not conduct certainty analyses or consider confidence in findings with respect to whether a study was rated as “good” or “fair”. Table 2 highlights that the potential key problems were with attrition greater than 20% (in 9 / 18 studies), insufficient attention to confounding factors (7 / 18 studies) and the absence of power calculations in almost all studies. Additionally, as only peer-reviewed journal articles were included in this review, we acknowledge that the file drawer problem may have contributed to bias in the review process, where unpublished insignificant research findings have not been considered.

Future work here should examine the relative significance of non-disease, child, factors (e.g. temperament and early cognitive deficits) in understanding outcomes. Moreover, most of the studies were not anchored in explicit theoretical models. Thus, explicit mechanisms of action are not clear. We might suggest, for example, that parenting stress and distress compromises the effective parenting strategies required to facilitate child adjustment, social functioning and independence, but further research, informed by theoretical frameworks, is required to evaluate such suggestions. Related, how family factors interact with disease factors, as suggested by the Berant et al. [18] study, should also be considered.

Our review suggests that studies to date have largely focused on family risk factors. It will be important that we also look at the etiological significance of protective factors. Factors such as family resilience, adaptability and cohesion may offer another lens through which to consider the family's impact on child psychosocial outcomes, and there have been calls in the CHD literature for future research to incorporate such concepts [15].

4.3. Conclusions and clinical implications

In incorporating only longitudinal studies, which reduces the directionality and respondent bias concerns of cross-sectional research, we have advanced the case for the relevance of family factors in understanding outcomes for children with CHD. Although disease factors did appear more reliably associated with psychosocial outcomes in infancy and early childhood, this association diminished in favour of family factors with older developmental cohorts.

Evidence from the current review has implications for clinical practice. Routine neurodevelopmental screening of children, with significant CHD markers, has been recommended by the American Heart Association since 2012 [1] and again most recently in their updated 2024 statement [76]. Our findings suggest that similar screening for parent and family risk factors is warranted. Research related to the timing, feasibility, acceptability and validity of such screening has been called for [15].

Findings also suggest family focused interventions, as part of pediatric care, are warranted. Encouraging evidence already exists regarding the positive impacts of relatively brief family-focused interventions among this cohort [31,77,78]. Strengthening family resilience, relationships and communication patterns, as well as supporting parental mental health may serve to reduce behavioral and emotional problems observed in this group.

CRediT authorship contribution statement

Chris McCusker: Writing – review & editing, Writing – original draft, Supervision, Methodology, Formal analysis, Conceptualization. **Lucy Phillips:** Writing – original draft, Project administration, Methodology, Investigation, Formal analysis. **Stephanie Murphy:** Writing – review & editing, Formal analysis.

Declaration of competing interest

The authors report no conflicts of interest. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Acknowledgements

This work was supported by Ireland's *Health Service Executive* as the sponsor of the pre-doctoral training post to Lucy Phillips. The research was conducted independently of the sponsoring agency and the sponsoring agency had no role in the conception, design, analysis or write up of this study.

Data availability

The data underlying this article are available in the article.

References

[1] B.S. Marino, P.H. Lipkin, J.W. Newburger, G. Peacock, M. Gerdes, J.W. Gaynor, K. A. Mussatto, K. Uzark, C.S. Goldberg, W.H. Johnson, J. Li, S.E. Smith, D. C. Bellinger, W.T. Mahle, Neurodevelopmental outcomes in children with congenital heart disease: evaluation and management, Circulation 126 (2012) 1143–1172, https://doi.org/10.1161/CIR.0b013e318265ee8a.

[2] S.D. Roberts, V. Kazazian, M.K. Ford, D. Marini, S.P. Miller, V. Chau, M. Seed, L. G. Ly, T.S. Williams, R. Sananes, The association between parent stress, coping and mental health, and neurodevelopmental outcomes of infants with congenital heart disease, Clin. Neuropsychol. 35 (2021) 948–972, https://doi.org/10.1080/13854046.2021.1896037.

[3] M. Drakouli, K. Petsios, M. Giannakopoulou, E. Patiraki, I. Voutoufianaki, V. Matziou, Determinants of quality of life in children and adolescents with CHD: a

systematic review, Cardiol. Young 25 (2015) 1027–1036, https://doi.org/10.1017/S1047951115000086.
[4] A.M. Groh, R.P. Fearon, M.J. Bakermans-Kranenburg, M.H. van IJzendoorn, R. D. Steele, G.I. Roisman, The significance of attachment security for children's social competence with peers: a meta-analytic study, Attach Hum. Dev. 16 (2014) 103–136, https://doi.org/10.1080/14616734.2014.883636.
[5] J. Leeman, J.L. Crandell, A. Lee, J. Bai, M. Sandelowski, K. Knafl, Family functioning and the well-being of children with chronic conditions: a Meta-analysis, Res. Nurs. Health 39 (2016) 229–243, https://doi.org/10.1002/nur.21725.
[6] S. Thomas, N.P. Ryan, L.K. Byrne, C. Hendrieckx, V. White, Psychological distress among parents of children with chronic health conditions and its association with unmet supportive care needs and children's quality of life, J. Pediatr. Psychol. 49 (2024) 45–55, https://doi.org/10.1093/jpepsy/jsad074.
[7] J.L. Schleider, J.R. Weisz, Family process and youth internalizing problems: a triadic model of etiology and intervention, Dev. Psychopathol. 29 (2017) 273–301, https://doi.org/10.1017/S095457941600016X.
[8] S.E. Woolf-King, A. Anger, E.A. Arnold, S.J. Weiss, D. Teitel, Mental health among parents of children with critical congenital heart defects: a systematic review, J. Am. Heart Assoc. 6 (2017) e004862, https://doi.org/10.1161/JAHA.116.004862.
[9] M. Ehrler, C. Wettach, I. Beck, E.R. Valsangiacomo Buechel, B. Latal, M.A. Landolt, Mental health-related quality of life in mothers of children with surgically repaired congenital heart disease: a 13-year longitudinal study, Qual. Life Res. 32 (2023) 2975–2986, https://doi.org/10.1007/s11136-023-03440-y.
[10] A.C. Jackson, E. Frydenberg, R.P.-T. Liang, R.O. Higgins, B.M. Murphy, Familial impact and coping with child heart disease: a systematic review, Pediatr. Cardiol. 36 (2015) 695–712, https://doi.org/10.1007/s00246-015-1121-9.
[11] C.G. McCusker, N.N. Doherty, B. Molloy, F. Casey, N. Rooney, C. Mulholland, A. Sands, B. Craig, M. Stewart, Determinants of neuropsychological and behavioural outcomes in early childhood survivors of congenital heart disease, Arch. Dis. Child. 92 (2007) 137–141, https://doi.org/10.1136/adc.2005.092320.
[12] T. Grimaldi Capitello, C. Correale, G. Amodeo, M. Balsamo, L. Carlucci, C. Fiorilli, Childhood heart disease and parental emotional wellbeing: a predictive model to explain the perception of quality of life in children and adolescents, Health Qual. Life Outcomes 23 (2025) 1, https://doi.org/10.1186/s12955-024-02328-w.
[13] D.R. DeMaso, M. Labella, G.A. Taylor, P.W. Forbes, C. Stopp, D.C. Bellinger, M. J. Rivkin, D. Wypij, J.W. Newburger, Psychiatric disorders and function in adolescents with d-transposition of the great arteries, J. Pediatr. 165 (2014) 760–766, https://doi.org/10.1016/j.jpeds.2014.06.029.
[14] M. Ehrler, R. O'Gorman, F.M. Wehrle, A. Speckert, A. Jakab, O. Kretschmar, B. Latal, Learning from those who thrive: protective factors and neuroimaging markers in adolescents with complex congenital heart disease and with a favorable neurodevelopmental profile, Child Neuropsychol. J. Norm. Abnorm. Dev. Child. Adolesc. 31 (2025) 613–634, https://doi.org/10.1080/09297049.2024.2419048.
[15] E. Sood, A.J. Lisanti, S.E. Woolf-King, J. Wray, N. Kasparian, E. Jackson, M. R. Gregory, K.N. Lopez, B.S. Marino, T. Neely, A. Randall, S.C. Zyblewski, C. L. Brosig, Parent mental health and family functioning following diagnosis of CHD: a research agenda and recommendations from the cardiac neurodevelopmental outcome collaborative, Cardiol. Young 31 (2021) 900–914, https://doi.org/10.1017/S1047951121002134.
[16] M.J. Page, J.E. McKenzie, P.M. Bossuyt, I. Boutron, T.C. Hoffmann, C.D. Mulrow, L. Shamseer, J.M. Tetzlaff, E.A. Akl, S.E. Brennan, R. Chou, J. Glanville, J. M. Grimshaw, A. Hróbjartsson, M.M. Lalu, T. Li, E.W. Loder, E. Mayo-Wilson, S. McDonald, L.A. McGuinness, L.A. Stewart, J. Thomas, A.C. Tricco, V.A. Welch, P. Whiting, D. Moher, The PRISMA 2020 statement: an updated guideline for reporting systematic reviews, BMJ 372 (2021) n71, https://doi.org/10.1136/bmj.n71.
[17] National Institutes of Health, National Institutes of Health Quality Assessment tool for Observational Cohort and Cross-Sectional Studies. https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools, 2021.
[18] E. Berant, M. Mikulincer, P.R. Shaver, Mothers' attachment style, their mental health, and their children's emotional vulnerabilities: a 7-year study of children with congenital heart disease, J. Pers. 76 (2008) 31–66, https://doi.org/10.1111/j.1467-6494.2007.00479.x.
[19] R.E. Brandlistuen, K. Stene-Larsen, H. Holmstrøm, M.A. Landolt, L.T. Eskedal, M. E. Vollrath, Occurrence and predictors of developmental impairments in 3-year-old children with congenital heart defects, J. Dev. Behav. Pediatr. 32 (2011) 526, https://doi.org/10.1097/DBP.0b013e318222eb5a.
[20] M. Ehrler, J. Fervença Ramos, F.M. Wehrle, E.R. Valsangiacomo Buechel, I. Beck, B. Latal, M.A. Landolt, Longitudinal behaviour of children with complex congenital heart disease from preschool to adolescence, Pediatr. Res. (2026) 1–9, https://doi.org/10.1038/s41390-026-04839-2.
[21] S. Goldberg, R.J. Simmons, J. Newman, K. Campbell, R.S. Fowler, Congenital heart disease, parental stress, and infant-mother relationships, J. Pediatr. 119 (1991) 661–666, https://doi.org/10.1016/S0022-3476(05)82425-4.
[22] S. Goldberg, M. Janus, J. Washington, R.J. Simmons, I.a.N. MacLUSKY, R. S. Fowler, Prediction of preschool behavioral problems in healthy and pediatric samples, J. Dev. Behav. Pediatr. 18 (1997) 304.
[23] S. Goldberg, A. Gotowiec, R.J. Simmons, Infant-mother attachment and behavior problems in healthy and chronically ill preschoolers, Dev. Psychopathol. 7 (1995) 267–282, https://doi.org/10.1017/S0954579400006490.
[24] A. Heller, S. Rafman, I. Zvagulis, I.B. Pless, Birth defects and psychosocial adjustment, Am. J. Dis. Child. 139 (1985) 257–263, https://doi.org/10.1001/archpedi.1985.02140050051021.
[25] E.A. Herrup, A.J. Shillingford, J.W. Gaynor, C.F.B. Driscoll, L. Hampton, B. Zhang, K. Baxelbaum, N. Burnham, Association of Parenting Stress in early childhood with emotional health and resilience of young adults with congenital heart disease, J. Pediatr. 294 (2026) 115110, https://doi.org/10.1016/j.jpeds.2026.115110.
[26] M.A. Landolt, E. Ystrom, K. Stene-Larsen, H. Holmstrøm, M.E. Vollrath, Exploring causal pathways of child behavior and maternal mental health in families with a child with congenital heart disease: a longitudinal study, Psychol. Med. 44 (2014) 3421–3433, https://doi.org/10.1017/S0033291713002894.
[27] L. Lang, J. Gerlach, A.-C. Plank, A. Purbojo, R.A. Cesnjevar, O. Kratz, G.H. Moll, A. Eichler, Becoming a teenager after early surgical ventricular septal defect (VSD) repair: longitudinal biopsychological data on mental health and maternal involvement, J. Clin. Med. 11 (2022) 7242, https://doi.org/10.3390/jcm11237242.
[28] K. Luyckx, L. Missotten, E. Goossens, P. Moons, Individual and contextual determinants of quality of life in adolescents with congenital heart disease, J. Adolesc. Health 51 (2012) 122–128, https://doi.org/10.1016/j.jadohealth.2011.11.007.
[29] K. Luyckx, E. Goossens, J. Rassart, S. Apers, J. Vanhalst, P. Moons, Parental support, internalizing symptoms, perceived health status, and quality of life in adolescents with congenital heart disease: influences and reciprocal effects, J. Behav. Med. 37 (2014) 145–155, https://doi.org/10.1007/s10865-012-9474-5.
[30] A. Majnemer, C. Limperopoulos, M. Shevell, C. Rohlicek, B. Rosenblatt, C. Tchervenkov, Developmental and functional outcomes at school entry in children with congenital heart defects, J. Pediatr. 153 (2008) 55–60.e1, https://doi.org/10.1016/j.jpeds.2007.12.019.
[31] C.G. McCusker, M.P. Armstrong, M. Mullen, N.N. Doherty, F.A. Casey, A sibling-controlled, prospective study of outcomes at home and school in children with severe congenital heart disease, Cardiol. Young 23 (2013) 507–516, https://doi.org/10.1017/S1047951112001667.
[32] L. Oris, I. Seiffge-Krenke, P. Moons, L. Goubert, J. Rassart, E. Goossens, K. Luyckx, Parental and peer support in adolescents with a chronic condition: a typological approach and developmental implications, J. Behav. Med. 39 (2016) 107–119, https://doi.org/10.1007/s10865-015-9680-z.
[33] J. Rassart, K. Luyckx, E. Goossens, S. Apers, P. Moons, A closer look at the developmental interplay between parenting and perceived health in adolescents with congenital heart disease, J. Behav. Med. 37 (2014) 1202–1214, https://doi.org/10.1007/s10865-014-9569-2.
[34] K.J. Visconti, K.J. Saudino, L.A. Rappaport, J.W. Newburger, D.C. Bellinger, Influence of parental stress and social support on the behavioral adjustment of children with transposition of the great arteries, J. Dev. Behav. Pediatr. 23 (2002) 314.
[35] I. Werninger, M. Ehrler, F.M. Wehrle, M.A. Landolt, S. Polentarutti, E. R. Valsangiacomo Buechel, B. Latal, Social and behavioral difficulties in 10-year-old children with congenital heart disease: prevalence and risk factors, Front. Pediatr. 8 (2020), https://doi.org/10.3389/fped.2020.604918.
[36] M.D.S. Ainsworth, M.C. Blehar, E. Waters, S. Wall, Patterns of Attachment: A Psychological Study of the Strange Situation, Lawrence Erlbaum, Oxford, England, 1978.
[37] R. Abidin, Parenting Stress Index: Professional Manual, Psychological Assessment Resources, 1995.
[38] N. Bayley, Bayley Scales of Infant Development, 2nd ed., The Psychological Corporation, Harcourt Brace & Company, 1991.
[39] T.M. Achenbach, L.A. Rescorla, Manual for the ASEBA School-Age: Forms & Profiles: An Integrated System of Multi-Informant Assessment: Child Behavior Checklist for Ages 6–18, Teacher's Report For, Youth Self-report, Aseba, 2001.
[40] M. Mikulincer, V. Florian, R. Tolmacz, Attachment styles and fear of personal death: a case study of affect regulation, J. Pers. Soc. Psychol. 58 (1990) 273–280, https://doi.org/10.1037/0022-3514.58.2.273.
[41] W.H. Fitts, The Tennessee Self-Concept Scale Manual, Counselor Recording and Tests, Nashville, TN, 1965.
[42] L. Bellak, D.M. Abrams, The T.A.T., The C.A.T., and The S.A.T. in Clinical Use, 6th edition, Allyn & Bacon, 1996.
[43] M. Rutter, J. Tizard, K. Whitmore, Education, Health and Behavior: A Psychological Study of Child Development, Wiley & Sons, New York, 1970.
[44] W.B. Carey, S.C. McDevitt, Revision of the infant temperament questionnaire, Pediatrics 61 (1978) 735–739, https://doi.org/10.1542/peds.61.5.735.
[45] I.B. Pless, K. Roghmann, R.J. Haggerty, Chronic illness, family functioning, and psychological adjustment: a model for the allocation of preventive mental health services*, Int. J. Epidemiol. 1 (1972) 271–277, https://doi.org/10.1093/ije/1.3.271.
[46] J.H. Beitchman, S. Raman, The assessment of childhood psychopathology: the construction of a new self-report psychiatric rating scale for children, Multivar. Exp. Clin. Res. 4 (1979) 23–31.
[47] E.H. Schludermann, S.M. Schludermann, Children's Report on Parent Behavior (CRPBI-108, CRPBI-30) for Older Children and Adolescents (Tech. Rep.), University of Manitoba, Department of Psychology, Manitoba, Canada, 1988.
[48] P. Moons, K.V. Deyk, L.D. Bleser, K. Marquet, E. Raes, S.D. Geest, W. Budts, Quality of life and health status in adults with congenital heart disease: a direct comparison with healthy counterparts, Eur. J. Cardiovasc. Prev. Rehabil. 13 (2006) 407–413, https://doi.org/10.1097/01.hjr.0000221864.19415.a0.
[49] A. Antonovsky, Health, stress, and coping, new Perspect. Ment. Phys. Well-Being, 1979, pp. 12–37.
[50] J. Bouma, A. Ranchor, R. Sanderman, E. van Sonderen, The Measurement of Symptoms of Depression with the CESD: A Manual, Noordelijk Centrum voor Gezondheidsvraagstukken, Rijksuniversiteit Groningen, 1995.

[51] R.E. Roberts, P.M. Lewinsohn, J.R. Seeley, A brief measure of loneliness suitable for use with adolescents, Psychol. Rep. 72 (1993) 1379–1391, https://doi.org/10.2466/pr0.1993.72.3c.1379.
[52] K. Uzark, K. Jones, T.M. Burwinkle, J.W. Varni, The pediatric quality of life inventory™ in children with heart disease, Prog. Pediatr. Cardiol. 18 (2003) 141–149, https://doi.org/10.1016/S1058-9813(03)00087-0.
[53] D. Wechsler, Wechsler Preschool and Primary Scale of Intelligence—Revised, The Psychological Corporation, San Antonio, TX, 1989.
[54] L.M. Dunn, L.M. Dunn, Peabody Picture Vocabulary Test - Third Edition (PPVT-III), APA PsycTests, 1997.
[55] S.S. Sparrow, D.A. Balla, D.V. Cicchetti, Vineland Adaptive Behavior Scales: Interview Edition: Expanded Form Manual, American Guidance Service, Circle Pines, MN, 1984.
[56] R. Folio, R. Fewell, Peabody Developmental Motor Scales-2, PRO-Ed, Austin, TX, 2000.
[57] L.R. Derogatis, The Brief Symptom Inventory (BSI): Administration, Scoring and Procedures Manual, National Computer Systems, Minneapolis, 1993.
[58] K.A. DeVet, H.T. Ireys, Psychometric properties of the maternal worry scale for children with chronic illness, J. Pediatr. Psychol. 23 (1998) 257–266, https://doi.org/10.1093/jpepsy/23.4.257.
[59] R.H. Moos, B.S. Moos, Family Environment Scale, McGraw-Hill, NY, 1991.
[60] L.R. Derogatis, R.S. Lipman, K. Rickels, E.H. Uhlenhuth, L. Covi, The Hopkins symptom checklist (HSCL): a self-report symptom inventory, Behav. Sci. 19 (1974) 1–15, https://doi.org/10.1002/bs.3830190102.
[61] J. Squires, D. Bricker, L. Potter, Revision of a parent-completed developmental screening tool: ages and stages Questionnaires1, J. Pediatr. Psychol. 22 (1997) 313–328, https://doi.org/10.1093/jpepsy/22.3.313.
[62] R. Goodman, The strengths and difficulties questionnaire: a research note, J. Child Psychol. Psychiatry 38 (1997) 581–586, https://doi.org/10.1111/j.1469-7610.1997.tb01545.x.
[63] J.E. Bates, C.A.B. Freeland, M.L. Lounsbury, Measurement of infant difficultness, Child Dev. 50 (1979) 794–803, https://doi.org/10.2307/1128946.
[64] J.A. Flaherty, F.M. Gaviria, D.S. Pathak, The measurement of social support: The social support network inventory, Compr. Psychiatry 24 (1983) 521–529.
[65] S. Bölte, Brief report: The social responsiveness scale for adults (SRS-A): initial results in a German cohort, J. Autism Dev. Disord. 42 (2012) 1998–1999, https://doi.org/10.1007/s10803-011-1424-5.
[66] H. Christiansen, O. Hirsch, R. Drechsler, S. Wanderer, E.-L. Knospe, T. Günther, K. Lidzba, German validation of the Conners 3U+00AE rating scales for parents, teachers, and children, Z. Kinder Jugendpsychiatr. Psychother. (2016), https://doi.org/10.1024/1422-4917/a000408 (accessed January 14, 2026).
[67] K.K. Shelton, P.J. Frick, J. Wootton, Assessment of parenting practices in families of elementary school-age children, J. Clin. Child Psychol. 25 (1996) 317–329, https://doi.org/10.1207/s15374424jccp2503_8.
[68] D. Wechsler, Wechsler Intelligence Scale for Children–Fifth Edition, 2024, https://doi.org/10.1037/t79359-000.
[69] M. Döpfner, A. Görtz-Dorten, DISYPS-III - Diagnostik-System für Psychische Störungen nach ICD-10 und DSM-5 für Kinder und Jugendliche - III. https://psy-lab.librarika.com/search/detail/7319647, 2017 (accessed July 15, 2026).
[70] U. Ravens-Sieberer, U. Ellert, M. Erhart, Gesundheitsbezogene Lebensqualität von Kindern und Jugendlichen in Deutschland, Bundesgesundheitsbl. Gesundheitsforsch. Gesundheitsschutz 50 (2007) 810–818, https://doi.org/10.1007/s00103-007-0244-4.
[71] J.E. Ware, C.D.S.R. work(s): The MOS 36-item short-form health survey (SF-36): I. Conceptual framework and item selection, Med. Care 30 (1992) 473–483.
[72] G.M. Wagnild, The Resilience Scale User's Guide: For the US English Version of the Resilience Scale and the 14-Item Resilience Scale (RS-14), Resilience center, 2011.
[73] J. Cohen, Statistical Power Analysis for the Behavioral Sciences, 2nd ed., Lawrence Erlbaum Associates, Hillsdale, NJ, 1988.
[74] C. Erdei, C.H. Liu, M. Machie, P.T. Church, R. Heyne, Parent mental health and neurodevelopmental outcomes of children hospitalized in the neonatal intensive care unit, Early Hum. Dev. 154 (2021) 105278, https://doi.org/10.1016/j.earlhumdev.2020.105278.
[75] U. Orth, How large are actor and partner effects of personality on relationship satisfaction? The importance of controlling for shared method variance, Personal. Soc. Psychol. Bull. 39 (2013) 1359–1372, https://doi.org/10.1177/0146167213492429.
[76] E. Sood, J.W. Newburger, J.S. Anixt, A.R. Cassidy, J.L. Jackson, R.A. Jonas, A. J. Lisanti, K.N. Lopez, S. Peyvandi, B.S. Marino, On behalf of the American Heart Association Council on lifelong congenital heart disease and heart health in the young and the council on cardiovascular and stroke nursing, Neurodevelopmental outcomes for individuals with congenital heart disease: updates in neuroprotection, risk-stratification, evaluation, and management: a scientific statement from the American Heart Association, Circulation 149 (2024) e997–e1022, https://doi.org/10.1161/CIR.0000000000001211.
[77] C.G. McCusker, N.N. Doherty, B. Molloy, N. Rooney, C. Mulholland, A. Sands, B. Craig, M. Stewart, F. Casey, A controlled trial of early interventions to promote maternal adjustment and development in infants born with severe congenital heart disease, Child Care Health Dev. 36 (2010) 110–117, https://doi.org/10.1111/j.1365-2214.2009.01026.x.
[78] C.G. McCusker, N.N. Doherty, B. Molloy, N. Rooney, C. Mulholland, A. Sands, B. Craig, M. Stewart, F. Casey, A randomized controlled trial of interventions to promote adjustment in children with congenital heart disease entering school and their families, J. Pediatr. Psychol. 37 (2012) 1089–1103.