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Review

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Parent mental health and family functioning following diagnosis of CHD: a research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative

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Abstract

Diagnosis of CHD substantially affects parent mental health and family functioning, thereby influencing child neurodevelopmental and psychosocial outcomes. Recognition of the need to proactively support parent mental health and family functioning following cardiac diagnosis to promote psychosocial adaptation has increased substantially over recent years. However, significant gaps in knowledge remain and families continue to report critical unmet psychosocial needs. The *Parent Mental Health and Family Functioning Working Group* of the Cardiac Neurodevelopmental Outcome Collaborative was formed in 2018 through support from an R13 grant from the National Heart, Lung, and Blood Institute to identify significant knowledge gaps related to parent mental health and family functioning, as well as critical questions that must be answered to further knowledge, policy, care, and outcomes. Conceptually driven investigations are needed to identify parent mental health and family functioning factors with the strongest influence on child outcomes, to obtain a deeper understanding of the biomarkers associated with these factors, and to better understand how parent mental health and family functioning influence child outcomes over time. Investigations are also needed to develop, test, and implement sustainable models of mental health screening and assessment, as well as effective interventions to optimise parent mental health and family functioning to promote psychosocial adaptation. The critical questions and investigations outlined in this paper provide a roadmap for future research to close gaps in knowledge, improve care, and promote positive outcomes for families of children with CHD.

Introduction

The November, 2020 issue of Cardiology in the Young contains the inaugural five manuscripts from the Cardiac Neurodevelopmental Outcome Collaborative[1–5] marking the beginning of the partnership between the Cardiac Neurodevelopmental Outcome Collaborative and Cardiology in the Young. In this issue of Cardiology in the Young, this article is part of the first set of three papers from the Cardiac Neurodevelopmental Outcome Collaborative

R13 grant funded by the National Heart, Lung, and Blood Institute of the National Institutes of Health of the United States of America, which defines the research agenda for the next decade across seven domains of cardiac neurodevelopmental and psychosocial outcomes research[6–8]:

Neurodevelopmental and psychological difficulties affect more than half the population of individuals with complex forms of CHD.[9–12] Efforts over the past decade to identify modifiable factors that influence the neurodevelopmental and psychological sequelae of CHD have focused predominantly on surgical and perioperative variables;[12] however, individuals with CHD spend most of their developing years out of the hospital with their parents, caregivers, and other broader family and social networks. Parent mental health, parenting behaviours, and the family environment are known to exert a powerful influence on child neurodevelopment, behavior, and emotional well-being[13,14] and can exacerbate or mitigate the effects of medical experiences on child outcomes.[15]

Given the demands of parenting a child with CHD and repeated exposure to potentially traumatic medical events,[16–18] it is not surprising that parent mental health can be substantially affected. CHD diagnosis and treatment often occur (or at least begin) during the prenatal and early postnatal periods, a time when mothers and fathers are at heightened risk for mental health difficulties,[13,19–21] and when the bond between infant and parent is in the formative stages of development.[22] A systematic review of mental health symptoms in parents of children with CHD found that over 80% of parents report clinically significant symptoms of post-traumatic stress, 30–80% report severe psychological distress, and 25–50% report depression and/or anxiety.[23] These rates of mental health symptoms far exceed what is reported in the general population and are similar to other trauma-exposed populations, including the military.[24] Fewer studies have focused on fathers specifically, but those that have suggest fathers may experience the stress of CHD differently from mothers and exhibit different mental health symptoms.[18,25,26] A growing literature suggests that parent education level, household income, family structure, country of birth, and language may also influence parent mental health following diagnosis of CHD.[27,28] Elevated parent distress within and beyond the perinatal period is associated with an increased risk of emotional and behavioural difficulties and poorer health-related quality of life in children with CHD.[29–33] Moreover, parent mental health is often a stronger predictor of child developmental outcomes than medical variables, including CHD severity and surgical factors.[29,33] Psychosocial adaptation in the parents may reduce the risk for parent mental health difficulties and promote positive child neurodevelopmental and psychosocial outcomes.

Family functioning encompasses many facets of the family environment including relationships amongst family members, parent–child attachment, and levels of conflict, cohesion, adaptation, communication quality, and organisation.[34,35] CHD has been shown to influence family relationships, including parent–child, marital/partner, sibling, and extended family relationships, as well as parenting style/practices.[18,25,36,37] While many families report increased family conflict, decreased communication, feelings of isolation, and difficulties with parent–infant bonding following a diagnosis of CHD, others report greater family cohesion and support.[18,25,38] The financial burden of CHD and disruptions to employment and family routine associated with long hospital stays and frequent medical appointments likely influence family adaptation following CHD diagnosis as well.[39,40]

Recognition of the need to proactively support parent mental health and family functioning to reduce suffering, promote psychosocial adaptation, and improve child neurodevelopmental and psychosocial outcomes has increased substantially over recent years.[41–44] Published research does not, however, provide the necessary evidence to determine the type, timing, or delivery mode of interventions most likely to improve parent mental health and family functioning. Additionally, prior research has rarely focused on identifying which difficulties are of greatest concern to parent stakeholders or exert the strongest influence on child and family outcomes and are, therefore, of highest priority for intervention.[45,46] Significant gaps in knowledge remain, and parents and families continue to report critical unmet psychosocial needs.[46]

The *Parent Mental Health and Family Functioning Working Group* of the Cardiac Neurodevelopmental Outcome Collaborative is comprised of multidisciplinary topic area experts (in psychology, cardiology, nursing, social work) from three continents (North America, Europe, Australia), a health disparities expert, and parent stakeholders (Table 1). This working group is one of the seven formed by the Cardiac Neurodevelopmental Outcome Collaborative in 2018 to identify significant gaps in knowledge and critical questions that must be answered to advance neurodevelopmental care and outcomes. The effort was supported by a National Heart, Lung, and Blood Institute R13 grant awarded to the Cardiac Neurodevelopmental Outcome Collaborative in collaboration with the Ann & Robert H. Lurie Children's Hospital of Chicago, which funded a 2-day meeting of multidisciplinary, multinational experts, and patient/caregiver stakeholders in Kansas City, Missouri. The specific goals of the *Parent Mental Health and Family Functioning Working Group* were to identify: (1) significant knowledge gaps related to parent mental health and family functioning within the context of CHD; (2) critical questions that must be answered to further knowledge, policy, care, and outcomes; and (3) investigations needed to answer these critical questions. Although parents and families of children with complex forms of CHD requiring cardiac surgery during infancy were the primary focus of the working group, recommendations may also apply to parents/families of children with milder forms of CHD, as objective illness severity does not consistently predict mental health outcomes.[28,47] The term "parent" in this context broadly refers to all primary caregivers including biological and adoptive and long-term foster parents, and other adults serving in a primary caregiving role. The term "family" includes both nuclear and extended family members. This paper presents the top five critical questions identified by the working group (Table 2) and provides specific recommendations for science and health policy to inform the next decade of research on parent mental health and family functioning in CHD.

Critical Question 1: Which parent mental health and family functioning factors are associated with child neurodevelopmental and psychosocial outcomes? How are these associations mediated and/or moderated by additional factors?

Existing knowledge

A growing body of research indicates that parent mental health symptoms are associated with poorer outcomes for children with

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Table 1. Parent Mental Health and Family Functioning Working Group Participants

WG participants	Discipline/Role	Institution/Organisation	Country
Erica Sood*	Paediatric Psychologist	Nemours/Alfred I. duPont Hospital for Children; Thomas Jefferson University	USA
Cheryl L. Brosig*	Paediatric Psychologist	Children's Wisconsin; Medical College of Wisconsin	USA
Emily Jackson	Social Worker	Nemours/Alfred I. duPont Hospital for Children	USA
Mary R. Gregory	Nurse Scientist	Children's Mercy Hospital; Missouri Western State University	USA
Nadine Kasparian	Paediatric Psychologist	Sydney Children's Hospitals Network; Cincinnati Children's Hospital; University of Cincinnati College of Medicine	Australia
Amy Jo Lisanti	Nurse Scientist/Clinical Nurse Specialist	Children's Hospital of Philadelphia; University of Pennsylvania School of Nursing	USA
Keila N. Lopez**	Paediatric Cardiologist	Texas Children's Hospital; Baylor College of Medicine	USA
Trent Neely	Parent Stakeholder	Sisters by Heart/Brothers by Heart	USA
Amy Randall	Parent Stakeholder	Mended *Little* Hearts of Wisconsin	USA
Sarah E. Woolf-King	Clinical Health Psychologist/ Parent Stakeholder	Syracuse University	USA
Jo Wray	Health Psychologist	Great Ormond Street Hospital for Children NHS Foundation Trust	UK
Sinai C. Zyblewski	Paediatric Cardiologist	Medical University of South Carolina	USA

WG = Working Group. *Working Group Co-Lead. **Health Disparities Expert.

CHD. Parent post-traumatic stress, referring to specific psychological and physiological symptoms (e.g., flashbacks, avoidance, hyperarousal) following exposure to a traumatic event (e.g., witnessing their child go into cardiac arrest), is associated with lower psychosocial functioning[29] and quality of life[30] for children and adolescents with CHD. Parenting stress, which refers to the magnitude of stress in the parent–child system, is associated with greater emotional and behavioural problems in children[32,47,48] and lower psychosocial functioning in adolescents with CHD.[29] Parent mental health symptoms are stronger predictors of child psychosocial functioning than medical or surgical factors,[29,33] and parent mental health can influence parent report of child health-related quality of life.[31,49] There is also preliminary evidence that untreated mental health symptoms may influence mother–child interactions (e.g., lower maternal responsivity, lower child positive interactivity) and increase the risk for child developmental delay,[50] consistent with the broader literature on maternal mental health in non-CHD populations[51–53] and highlighting the need for interventions targeting parent mental health symptoms.

Significant gaps in knowledge

While there is growing evidence that parent mental health and aspects of the parent–child relationship influence the outcomes of children with CHD, available studies have examined a range of parent mental health and family functioning factors using a variety of measures, often in the absence of a theoretical model to guide the selection of constructs and measures. Although a few researchers have proposed theoretical models for families of children with CHD that could guide the selection of constructs and measures,[27,54–56] most studies do not select constructs or measures based on an underlying theoretical model. Post-traumatic stress symptoms, parenting stress, anxiety, and depression are distinct clinical problems that may influence child outcomes through different mechanisms (e.g., overprotective *versus* disengaged parenting; emotionally over-involved *versus* hostile family environment),[14,57,58] but more work is needed to understand these underlying mechanisms in families affected by CHD. Parent, child, and environmental characteristics, such as sex, race, ethnicity, socioeconomic status, language barriers, physical health, stigma, discrimination, social networks, and parental vulnerability, are likely to moderate associations between parent mental health and child outcomes,[27] but these associations have not been adequately studied in families affected by CHD. Without the use of a theoretical model to guide hypothesis generation, research design, and outcomes measurement, the literature on parent mental and family functioning are unnecessarily heterogeneous, study findings are difficult to synthesise, and it is unclear which parent mental health or family factors are best to target in intervention design and clinical care.[23,44,45]

Investigations needed

(1) Ensure that future research is guided by a theoretical model.

Studies of parent mental health and family functioning should have a clear rationale for the selection of constructs based on a guiding theoretical model. Numerous existing models provide conceptual foundations for the influence of parent mental health, family functioning, and/or parenting on child outcomes (e.g., Double ABCX model, self-efficacy theory, parenting stress model, control theory, attachment theory).[59–64] Investigators have adapted and tested these models with high-risk families, including families of children with disabilities,[59,65–67] families of infants in the neonatal ICU,[68,69] families impacted by other paediatric illnesses or developmental disorders,[70,71] and families exposed to trauma.[72,73] Researchers have also developed or adapted theoretical models specifically for families of children with CHD including the Family Adaptation to Child Chronic Illness Model,[54] Parental Satisfaction and Wellness Model,[55] Paediatric Cardiac ICU

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Table 2. Parent mental health and family functioning: critical questions, significant gaps in knowledge, and investigations needed

Critical questions	Significant gaps in knowledge	Investigations needed
CQ1. Which parent mental health and family functioning factors are associated with child neurodevelopmental and psychosocial outcomes? How are these associations mediated and/or moderated by additional factors?	• Many prior studies have been conducted without a guiding theoretical model, resulting in unnecessary heterogeneity in constructs and measures, and difficulty in synthesising results across studies. • Underlying mechanisms via which parent MH and FF influence child outcomes are largely unknown. • Parent, child, and environmental characteristics that moderate associations between parent MH, FF, and child outcomes are unknown.	• Ensure that future research is guided by a theoretical model. • Investigate potential mediators and moderators. • Identify measures to assess constructs within a selected theoretical model
CQ2. What methodologies can be used to investigate associations between parent mental health and biological markers of stress?	• Extent to which maternal stress responses in the pre and perinatal period influence child outcomes in the context of CHD is not known. • Biological factors that provide the best indication of risk for stress/distress in children with CHD and their parents have not been identified. • Emotional and biobehavioural attunement (or misattunement) in the developing CHD parent–infant relationship has not been sufficiently studied.	• Establish an evidence base for the use of biomarkers of stress and distress in research with parents and their children with CHD. • Investigate associations between biological, psychological, and social responses to stress and distress in parents and their children with CHD.
CQ3. How do parent mental health and family functioning change and influence child neurodevelopmental and psychosocial outcomes *over time*? Which parent, child, and environmental factors mediate and moderate the trajectories of parent mental health and family functioning?	• There are limited data on how parent MH and FF fluctuate over time, when parents and families may be at greatest risk, and the extent to which associations between parent MH, FF, and child outcomes are causal. • Parent, child, and environmental characteristics that influence trajectories of parent MH and FF (including positive outcomes) are unknown.	• Conduct longitudinal research to evaluate relationships between parent MH, FF, and child neurodevelopmental and psychosocial outcomes. • Leverage multisite clinical registries to collect data on parent MH, FF, and child neurodevelopmental and psychosocial outcomes. • Utilise qualitative and mixed methods to understand the experiences of families over time.
CQ4. How and when should parent mental health and family functioning be assessed in clinical care settings?	• Not clear which aspects of parent MH and FF (including both risk and protective factors) are most important to assess at which time points. • Factors impacting the feasibility and acceptability of MH and FF screening within routine clinical practice are unknown. • Impact of screening on access to care and parent, family, and child outcomes is unknown.	• Identify optimal tools and timing for screening and assessment of parent MH and FF. • Determine feasibility and acceptability of screening and assessment processes for diverse stakeholders. • Evaluate whether screening of parent MH and FF increases access to care and improves outcomes.
CQ5. How and when should interventions be offered to bolster parent mental health and family adaptation, and optimise child neurodevelopmental and psychosocial outcomes?	• Limited rigorous RCTs to determine the efficacy of interventions for CHD families and no data on the potential cost-effectiveness of intervention. • Comparative effectiveness, mechanisms of action, and feasibility of heterogeneous intervention approaches are largely unknown. • Extent to which evidence-based interventions for other patient populations can be adapted for CHD is unknown. • Effects of incorporating shared decision-making, peer support, and technology into interventions are unknown.	• Evaluate interventions using rigorous randomised controlled efficacy trials, followed by effectiveness trials that guide real-world implementation. • Adapt interventions developed for other high-risk populations and determine optimal intervention content, format, timing, and dose. • Explore effects of incorporating shared decision-making and peer support into interventions. • Explore technology-based modes of intervention delivery.

CHD = congenital heart disease; CQ = Critical Question; FF = family functioning; MH = mental health; RCT = randomised controlled trial.

Parental Stress Model,[56] and Parental Stress and Resilience Model for CHD.[27] The latter is the only published model to provide a conceptual foundation for how parent mental health following CHD diagnosis influences child outcomes and includes environmental factors (e.g., neighbourhood, access to care) that may contribute to health disparities. The process of model selection for a particular study should be directly informed by study objectives and should include patient and parent involvement to ensure the model and included constructs are perceived as meaningful and relevant to those with lived experience.[74] As studies incorporate theory-driven methodology, theoretical models can be adapted based on empirical evidence and ultimately validated for families of children with CHD.

(2) Investigate potential mediators and moderators.

Future research should directly examine potential mechanisms by which parent mental health and family functioning influence outcomes for children with CHD, including attachment, co-regulation of the physiological stress response, parent–child interaction, parenting style, and family environment. Research should also include parent, child, and environmental characteristics that

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may moderate associations amongst parent mental health, family functioning, and child outcomes, such as sex, race, ethnicity, cultural factors, socioeconomic status, language, physical health, discrimination, and social networks. Identification of mediators and moderators will inform theory development and adaptation, help to identify targets for intervention, and enhance our understanding of intervention effects.[75]

(3) Identify measures to assess constructs within a selected theoretical model.

Measures to assess constructs within a selected theoretical model should be identified and utilised consistently in research. Investigators have conducted systematic reviews summarising the psychometric properties of a wide range of measures (e.g., adult mental health, parent–child attachment and interaction, child social–emotional and behavioural outcomes) with expectant parents, families of young children, and families impacted by other chronic illnesses, [76–80] which can guide the selection of measures for families affected by CHD. Some measures may require psychometric adaptation and validation for use with families affected by CHD, such as those designed to capture responses to a specific experience or environment. For example, the Parental Stressor Scale: Neonatal ICU was adapted to measure stress resulting specifically from infant hospitalisation,[81] and the resulting measure (Parental Stressor Scale: Infant Hospitalisation) was psychometrically evaluated with the parents of medically fragile, hospitalised infants.[82] Similar processes have been conducted with measures of post-traumatic stress symptoms resulting from specific types of trauma.[83] Parent and patient perspectives should be incorporated into the selection and/or adaptation of measures to ensure that they are clinically meaningful and acceptable to stakeholders.[84] Measures should also be linguistically and culturally appropriate and accessible for diverse families (e.g., available in multiple languages, literacy levels, and modalities).

Critical Question 2: What methodologies can be used to investigate associations between parent mental health and biological markers of stress?

Existing knowledge

Exposure to stress elicits a cascade of physiological responses. The hypothalamic–pituitary–adrenal axis, for example, is particularly responsive to psychosocial stress, with dynamic interaction between the brain, autonomic nervous system, endocrine and immune systems, and the gut microbiome.[85] Persistent high levels of psychosocial stress can result in a state of *allostatic load*; a physiological accumulation of the effects of stress that may contribute to a range of adverse health outcomes, including cardiovascular disease, neurocognitive decline, mental illness, and mortality.[85–90]

Up to 80% of parents of children with complex CHD report high levels of psychological stress and distress,[23] potentially increasing the risk of developing allostatic load and its consequences.[21,27] Biomarkers of HPA axis functioning (e.g., cortisol) can be measured by analysis of saliva, hair, skin, nail, stool, or blood samples, and have been used in studies investigating a range of psychological outcomes, including symptoms of stress, anxiety, and depression in children and adults.[91–95] In CHD, preliminary research has found that prenatal exposure to maternal stress and anxiety is associated with differences in foetal brain growth, including smaller hippocampal and cerebellar volumes.[96]

Significant gaps in knowledge

There is a dearth of data on biomarkers of parental psychological stress in CHD. The extent to which high levels of maternal prenatal and postnatal psychological stress may influence health outcomes for mothers and their children with CHD is an area of growing interest.[21] In other populations, maternal biological stress responses, such as HPA axis functioning in the perinatal period, have been shown to influence offspring health outcomes across the lifespan, including the risk of neurodevelopmental delay, and emotional and behavioural disorders, such as anxiety, depression, and attention deficit hyperactivity disorder.[97–101] There is, however, limited psychobiological research directly investigating these associations in the context of CHD and it is unclear which biomarkers, if any, may provide meaningful and reliable indications of future health risk.[21]

In addition to elucidating the effects of stress on individual health outcomes, this line of inquiry may also expand our understanding of the developing infant-parent relationship, particularly in terms of emotion regulation and biobehavioural attunement in the context of serious childhood illness.[102] Oxytocin, for example, has been widely studied as a biomarker of emotional co-regulation in parent–child dyads without CHD and has been associated with child developmental disorders such as autism spectrum disorder.[103,104] Higher maternal oxytocin levels have also been linked to more affectionate parenting behaviours, lower blood pressure, and lower stress; however, the role of oxytocin in the context of parent–child bonding and attachment, and the accuracy of measurements in mothers of infants with CHD, have yet to be determined.[105] Interest in the potential role of the microbiome is also growing, but to our knowledge, no published studies have examined the link between parental psychological stress and the developing infant gut microbiome in CHD, though studies are underway and may potentially provide strategies to augment existing therapies, such as the use of probiotics.[106,107]

Investigations needed

(1) Establish an evidence base for the use of stress biomarkers in research with parents and their children with CHD.

While the association between maternal biomarkers of psychological stress and child neurodevelopment and mental health has been investigated in other populations, an evidence base in CHD has yet to be established. Factors such as cost, participant burden, scepticism about clinical usefulness, and limited expertise in the field contribute to slow progress. Studying the interplay between psychological responses and physiological processes in CHD may provide a more comprehensive understanding of the mediators and moderators of parent and child mental health outcomes, and support the design and implementation of timely, tailored interventions.[21]

(2) Investigate associations between biological, psychological, and social responses to stress and distress in parents and their children with CHD.

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Aligned with the National Institutes of Health Symptom Science Model,[108] a comprehensive understanding of the underlying biological processes associated with psychological stress may yield improved diagnostic tools, prevention and intervention opportunities, and treatment approaches. To maximise impact, this work should also explore the potential differential effects of acute versus chronic stress (or allostatic load), and the existence of particularly "sensitive" periods when stress may have the strongest influence on health outcomes (e.g., during foetal development and early infancy).[89] It is well established that exposure of an individual to a stressor elicits a physiologic stress response, and studies in mental health are revealing associations of stress biomarkers with mental health symptoms and diagnoses. For example, recent pilot studies suggest salivary cortisol and heart rate variability may serve as non-invasive biomarkers of stress in infants with CHD and their mothers.[105,109–111] Larger investigations of the associations of stress biomarkers with mental health symptoms within the context of CHD is an important next step.

Critical Question 3: How do parent mental health and family functioning change and influence child neurodevelopmental and psychosocial outcomes *over time?* Which parent, child, and environmental factors mediate and moderate the trajectories of parent mental health and family functioning?

Existing knowledge

The small number of studies examining parent mental health across multiple time points after CHD diagnosis suggest psychological symptoms and physical manifestations change over time[112–115] and predict future child neurodevelopmental and psychosocial outcomes.[29,32,48] Studies have not longitudinally studied family functioning in CHD samples, but research with other patient populations suggests that changes in parental roles and responsibilities, in the quality and/or status of the relationship between parents, financial strain, and child medical care needs may influence family functioning.[116,117] Qualitative and survey-based research with parents of children with CHD has identified unique stressors specific to different phases of care (e.g., alterations in parental role within the ICU; difficulties with feeding and weight gain after hospital discharge; balancing protection with independence during the transition to school; navigating neurodevelopmental and learning challenges during school age; redefining the parental role during transition to adult care), and these may correlate with fluctuations in parent mental health, parenting behaviours, and family functioning.[16,25,118–123] However, not all parents and families exhibit long-term problems.[124] Preliminary research has begun to identify protective parent, child, and environmental resilience factors that moderate parent mental health effects and promote psychosocial adaptation and quality of life.[18,27,30,123,124] In general, research indicates that parent perceptions of child illness, coping strategies, perceived social support, and socioeconomic status are stronger predictors of parent mental health outcomes than objective illness characteristics.[32,125,126]

Significant gaps in knowledge

Investigators have predominantly assessed parent mental health and family functioning following diagnosis of CHD through cross-sectional studies and there remains much to be learned regarding how experiences fluctuate over time, when parents and families may be at greatest risk, and to what extent the associations between parent mental health and child outcomes are causal. Given that CHD is a lifelong condition, a snapshot of parent mental health and family functioning at one point in time (e.g., pregnancy) and in one setting (e.g., intensive care) may not accurately represent a family's holistic experience over time, nor the full range of outcomes for a child. While cross-sectional studies have begun to identify parent, child, and environmental factors that may moderate parent mental health and quality of life outcomes,[18,27] how they interact synergistically and their relative weights are still not entirely understood. Additionally, very few studies have examined protective factors or psychosocial adaptation over time in families affected by CHD.[127]

Investigations needed

(1) Conduct longitudinal research to evaluate relationships between parent mental health, family functioning, and child neurodevelopmental and psychosocial outcomes.

To better inform intervention, we recommend assessment of parent mental health, family functioning, and child neurodevelopmental and psychosocial outcomes *over time* within longitudinal cohort studies. Measures of parent mental health and family functioning should be included as early as the prenatal/postnatal periods and throughout childhood and adolescence. Inclusion of standardised measures assessing early effects will yield immediate clinical value and also provide longitudinal data for understanding lifespan trajectories. Cohort studies should include diverse and representative samples (including fathers, families with a lower socioeconomic status or non-traditional family structures, diverse racial and ethnic populations, rural populations) across multiple regions to ensure generalisability of results to diverse populations and identification of sociodemographic moderators that influence trajectories of parent mental health and family functioning.[128]

(2) Leverage multisite clinical registries to collect data on parent mental health, family functioning, and child neurodevelopmental and psychosocial outcomes.

Given the extensive resources required to conduct longitudinal cohort studies and the value of secondary data collected as part of routine care,[129–132] we recommend that multisite clinical registries be leveraged whenever possible to collect data on parent mental health, family functioning, and child neurodevelopmental and psychosocial outcomes. An increasing number of paediatric cardiac centres provide neurodevelopmental and psychosocial assessments as part of routine care for children with CHD and their families,[3,42] and clinical registries in North America, Europe, Australia, and New Zealand have recently been developed or expanded to include these data.[2,130,131,132–134] As an example, the Cardiac Neurodevelopmental Outcome Collaborative clinical registry launched in 2019 as a module of the Pediatric Cardiac Critical Care Consortium and Pediatric Acute Care Cardiology Collaborative registries and may serve as a valuable resource for researchers interested in studying parent mental health and family functioning in relation to child neurodevelopmental and psychosocial outcomes over time.[2] However, disparities in access to

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neurodevelopmental and psychosocial services must be addressed to ensure equitable healthcare and diverse samples.[135,136]

(3) Utilise qualitative and mixed methods to understand the experiences of families over time.

Qualitative and mixed methods (i.e., integrating quantitative and qualitative) are recommended to obtain a comprehensive understanding of parent and family lived experiences over time, as well as the range of responses elicited by stress, life events, transitions in care, or discrimination.[137] These approaches may be particularly helpful for better understanding the experiences of groups previously under-represented in research (e.g., fathers, families with a lower socioeconomic status, diverse racial and ethnic populations, LGBTQ communities, rural populations), and may also further our knowledge of understudied constructs and outcomes, such as which family functioning factors are likely to be protective, or how parents define and experience resilience, post-traumatic growth, and psychosocial adaptation.

Critical Question 4: How and when should parent mental health and family functioning be assessed in clinical care settings?

Existing knowledge

The need for routine assessment of parent mental health and family functioning to identify at-risk families who may benefit from increased support to promote psychosocial adaptation is well described in the literature,[23,31,42,140,141] and formal recommendations from the Association for European Pediatric and Congenital Cardiology Psychosocial Working Group,[41] Australian National Strategic Action Plan for Childhood Heart Disease,[142] and American Board of Pediatrics[43] include screening and assessment of parent mental health as crucial components of CHD care. There is an extensive literature on models of screening and assessment for family psychosocial risk and psychological distress within other paediatric illness populations,[143,144] and recent papers provide preliminary support for their applicability to parents of newborns with prenatally diagnosed birth defects in the early post-partum period,[145] parents of children with CHD following cardiac surgery,[146–147] and parents of children who underwent heart transplantation.[148] In addition, a trial evaluating a family-based mental health screening and stepped care model within paediatric cardiology is underway in Australia.[42,45]

Significant gaps in knowledge

Although the importance of routinely assessing parent mental health and family functioning within the context of CHD is well understood, existing research has not defined which specific aspects of mental health and family functioning should be assessed at which time points. Screening efforts to date have focused primarily on parent and family risk factors, with less emphasis on protective factors to promote psychosocial adaptation and positive child neurodevelopmental and psychosocial outcomes. Additionally, factors impacting the feasibility and acceptability of screening parents of children with CHD within routine clinical practice, and facilitating access to mental health services following a positive screen, have not been adequately explored. Finally, it is not known whether screening results in improved outcomes for families of children with CHD, and whether these outcomes are similar across diverse groups.

Investigations needed

(1) Identify the optimal tools and timing for routine screening and assessment of parent mental health and family functioning in clinical care settings.

Informed by theoretical models and psychometric studies (Critical Question 1) and by widespread efforts to promote mental health screening in health care settings,[141,143,149–151] research must identify the optimal tools for routine screening and assessment of important parent and family risk and resilience factors within the CHD population. Screening and assessment tools should screen for constructs that are clinically meaningful to parents and families,[138] gather information from multiple members in the family system,[152] emphasise building a family resilience framework,[153] consider potential financial burden and opportunity costs,[39,40] and be available and provide accommodations for use across languages, cultures, and ability levels. Building on knowledge regarding longitudinal trajectories of parent mental health and family functioning (Critical Question 3), research must also identify optimal time points for routine screening and assessment, such as times when families are likely to be at greatest risk or during windows of opportunity for promoting psychosocial adaptation.

(2) Determine the feasibility and acceptability of screening and assessment processes for diverse stakeholders.

Qualitative and mixed methods approaches may be helpful for obtaining the perspectives of diverse stakeholder groups (e.g., parents, physicians, social workers, clinic staff) regarding screening/assessment processes and strategies for connecting families with the appropriate supports following a positive screen.[84,137] Evidence-based online or mobile screening applications have demonstrated feasibility and acceptability in other high-risk populations such as military veterans,[154] children affected by war,[155] and patients with cancer,[156] and could be adapted and tested with families of children with CHD.

(3) Evaluate whether routine screening of parent mental health and family functioning increases access to care and improves outcomes.

Rigorous studies are needed to evaluate the clinical efficacy and cost-effectiveness of routine screening of parent mental health and
family functioning in both the short- and long term. Barriers to disclosing mental health need to medical providers[157] and to accessing mental health assessment and intervention services following a positive screen are common[158,159] and must be identified and addressed for diverse families impacted by CHD. In particular, studies should evaluate whether routine screening processes reduce existing ethnic, racial, and socioeconomic disparities in identifying risks and accessing services within the CHD population.[135,136] Research should also identify the resources necessary to implement family psychosocial screening within paediatric cardiology. Based on studies evaluating mental health screening in perinatal settings,[145,160] mental health providers embedded

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within the health care practice and staff trained to respond to urgent mental health needs seem to be critical components of an efficacious screening process.

Critical Question 5: How and when should interventions be offered to bolster parent mental health and family adaptation, and optimise child neurodevelopmental and psychosocial outcomes?

Existing knowledge

Two recent systematic reviews synthesised and critically appraised evidence on the efficacy of psychological interventions for parents and families of children with CHD.[44,45] Interventions vary widely in terms of therapeutic approach (e.g., education and parenting skills training, promoting parent–infant interaction and bonding, early paediatric palliative care). Tested interventions demonstrated reductions in maternal anxiety, with mixed evidence of efficacy for other parent (e.g., depression) and child (e.g., neurodevelopment, feeding) outcomes. Notably, very few studies included fathers or racial and ethnic minorities. Research examining parent preferences indicates that parents want interventions focused on direct management of parent mental health symptoms.[46] In addition, parents want peer support provided by other experienced parents as well as education on how to effectively communicate with medical teams, advocate for their child's needs, and partner in their child's medical and developmental care during hospitalisation.[46] Studies on parent preferences for communication and decision-making suggest that candid medical information delivered over time, sensitivity to cultural and linguistic diversity, provider validation of emotional responses and needs, and acknowledgement of the impact of medical care on the family can be helpful for mitigating parent stress.[46,161,162]

Significant gaps in knowledge

Literature on the development, adaptation, and implementation of interventions targeting parent mental health and family functioning in CHD is still in its infancy and many knowledge gaps remain. Existing interventions for this population are heterogeneous in terms of content, dose, timing, delivery format, and target outcomes, and the comparative effectiveness, feasibility, and cost-effectiveness of various intervention approaches and mechanisms of action are unknown. The needs of fathers, racial and ethnic minorities, and LGBTQ communities have not been specifically targeted, and strategies to increase the participation of under-represented groups in CHD intervention research have not been well-defined. Numerous family-based interventions have been developed and tested for other high-risk populations,[163] yet few studies have examined the extent to which these interventions can be successfully adapted and delivered to families affected by CHD. Despite the growing literature on parent preferences for intervention, support, and communication,[17,25,46,161,162,164] formalised peer support interventions have not been developed or evaluated for their impact on parent mental health, and studies exploring effective models of parent–provider communication and shared decision-making are very limited in CHD.[161] Cultural nuances regarding parent preferences for support and communication have also not been adequately explored.[165] Importantly, there is an absence of rigorous randomised controlled trials designed to determine the efficacy of family-based interventions for improving parent mental health, family functioning, and ultimately child neurodevelopmental and psychosocial outcomes in CHD.[44,45]

Investigations needed

(1) Evaluate interventions using rigorous randomised controlled efficacy trials, followed by effectiveness trials that guide real-world implementation.

A recent systematic review and best practice statement strongly endorses the provision of mental health interventions for parents of infants with CHD during ICU admission.[44] In order to implement evidence-based interventions in clinical care settings, this recommendation first requires the establishment of efficacy through use of tightly controlled randomised clinical efficacy trials using validated and theory-based outcome measures. Once efficacy is established, effectiveness trials that are designed to answer questions about real-world implementation and inclusive of economic evaluation are an essential next step. It is important to note here that while efficacy and effectiveness trials are distinct and equally important steps in the development of evidence-based behavioural treatments, efficacy trials can be designed with effectiveness in mind by considering the setting to which the intervention will be eventually delivered.[166–167] For example, evaluating a mode of delivery (e.g., telehealth) that is likely to be both cost-effective and implementable.[168] Multicentre studies are required to ensure adequate population diversity and adequate sample sizes. The "trial within a registry" method, which uses clinical registries as data platforms for randomised clinical trials, has been proposed as a way to leverage existing data, decrease research study costs, and enhance generalisability across settings.[169]

(2) Adapt interventions developed for other high-risk populations and determine optimal intervention content, format, timing, and dose.

Using the literature with perinatal and neonatal ICU populations as a foundation may expedite the development of interventions in CHD.[170–172] Cognitive-behavioural approaches have demonstrated efficacy for these high-risk populations and warrant adaptation to and testing with parents of children with CHD.[171,173] Transdiagnostic interventions, referring to those treatments that target the core maladaptive processes underlying many mental health disorders (e.g., mindfulness-based interventions, acceptance and commitment therapy),[174] have also shown promise for reducing depression, anxiety, and trauma for mothers of infants in the neonatal ICU[175] and could be adapted and tested for parents of children with CHD during the prenatal period, during hospitalisation, and after hospital discharge.[176,177] Family integrated care, which enables parents to become primary caregivers in the neonatal ICU, has been found to decrease parent stress and anxiety and could be adapted for cardiac inpatient settings.[178] Interventions that have been shown to improve parent mental health, family functioning, and child outcomes by teaching parents skills to manage child emotional and behavioural problems may warrant adaptation for families of preschool and school-age children with CHD.[179–181] The process of adapting existing interventions and determining optimal content, format, timing, and frequency should be conducted in accordance with established guidelines[166,182,183] and cultural and linguistic competencies[184] and in partnership with parent and clinician stakeholders to ensure that the resulting intervention is acceptable and feasible and targets

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meaningful outcomes.[74] The adaptation process should conclude with full-scale efficacy trials followed by effectiveness trials as described above.

(3) Explore the effects of incorporating shared decision-making and peer support into interventions.

A clinical report from the American Academy of Pediatrics describes a systematic approach to the implementation of shared decision-making for children with disabilities, which includes involving children in decisions about their care and developing decision-support tools and technologies.[185] This may be a useful framework from which to develop, adapt, and evaluate communication and shared decision-making resources for children with CHD and their parents. Importantly, preferences for shared decision-making are likely to differ across cultural, racial, and ethnic groups,[186] and the concept may be incongruent with some cultural values. The process of shared decision-making is also likely to change throughout phases of care and with increased child age.[187] Studies should evaluate the impact of shared decision-making on parent, family, and child outcomes at key transition points, from the time of initial diagnosis through the transition to adulthood. Additionally, those designing or adapting interventions should consider incorporating formalised peer support, with a focus on diversity and representation, to enhance parent outcomes including perceived social support, confidence, and empowerment.[188] Hospital–community partnership activities, such as patient/family advocacy groups, summer camps, and family education days, are promising methods for the delivery of peer support,[188,189] but require further evaluation with diverse peer mentors/mentees, including long-term follow-up[189] and examination of sex and cultural differences regarding preferences for peer support.[25]

(4) Explore technology-based modes of intervention delivery.

Behavioural intervention technologies, such as mental health and wellness interventions using web-based programmes and videoconferencing techniques, should be considered given the potential of new technology to increase access and reduce barriers to behavioural healthcare for families.[190] Interventions demonstrated to be efficacious and effective with families of children with CHD could be translated into technology-based interventions.[191–195] Alternatively, behavioural intervention technologies could be incorporated in the early stages of intervention development.[196,197] The extent to which technology-based interventions can reduce disparities in access to behavioural healthcare for under-represented groups should be specifically evaluated.

Cross-cutting themes

Several themes cut across the five critical questions outlined above. Representation of patients, parents, and families from diverse backgrounds is critical to understanding how race, ethnicity, culture, sex, socioeconomic status, LGBTQ identity, implicit bias, discrimination, and other factors influence and potentially contribute to health disparities in parent mental health, family functioning, parenting and child outcomes in CHD. The feasibility, acceptability, efficacy, and effectiveness of mental health screening, assessment, and intervention strategies are likely to differ across diverse populations, and appropriate tailoring and adaptation are essential. Strategies that have been effective in increasing sample diversity (including non-English-speaking participants) for other populations should be utilised or adapted for studies of parent mental health and family functioning in CHD.[198–201] Health system diversity with regard to structure, financing, and policy (both within and across nations) and disparities in access to health insurance also influence how physical and mental health services are delivered to children and families, and which barriers to care are likely to be most salient.[202–204] These issues must be considered when designing and implementing efficacy and effectiveness trials and quality improvement initiatives related to parent mental health and family functioning. Despite the critical need for increased knowledge of parent mental health and family functioning following CHD diagnosis, much of the prior research on this topic represents cross-sectional, survey-based studies, single-centre qualitative investigations, secondary analysis of existing data, and/or controlled trials with high risk of bias, and primarily with English-speaking White mothers.[23,44,45] Innovative methodological approaches and technological advances in electronic data capture, biomarker-based approaches, screening and assessment processes, tightly controlled efficacy trials, and implementable intervention strategies must be applied to advance knowledge, clinical care, and outcomes for families of children with CHD.

Conclusions

Neurodevelopmental and psychological difficulties are amongst the most common comorbidities of individuals with CHD. While a complex constellation of factors contributes to individual outcomes, parent psychosocial factors are foundational elements contributing to child health and well-being. Caring for a child with complex forms of CHD places significant demands on parents with strong evidence for parent vulnerability to high levels of acute and persistent psychological stress. The limited number of published mental health interventions for parents of children with CHD provide preliminary evidence of efficacy in reducing maternal distress and improving coping and psychosocial adaptation, but there remain substantial knowledge gaps, particularly in terms of the efficacy and effectiveness of interventions in improving paternal and child mental health, or outcomes for families from culturally- or linguistically diverse backgrounds, LGBTQ communities, or low-resource settings.

After a comprehensive review of the literature and extensive stakeholder engagement, the *Parent Mental Health and Family Functioning Working Group* identified five priorities to guide the advancement of research, clinical practice, and health policy:

(1) Conceptually driven investigations using reliable and validated measures to identify parent mental health and family functioning factors with the strongest influence on child neurodevelopmental and psychosocial outcomes and health-related quality of life;

(2) Interdisciplinary collaborations to elucidate the psychobiological underpinnings of neurodevelopmental and psychosocial deficits in children with CHD, including a deeper understanding of the biomarkers associated with these outcomes;

(3) Trajectories of child, parent, and family mental health, and the individual and environmental factors that may influence pathways to both risk and resilience, as well as identification of "sensitive" or "critical" periods when intervention may be of greatest benefit;

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(4) Development, trial, and implementation of sustainable models of mental health screening and assessment with capacity for tailoring to a range of settings and cultures; and

(5) Establishment of a suite of efficacious and effective interventions to optimise parent mental health and family functioning in order to promote psychosocial adaptation to achieve the best possible neurodevelopmental and psychosocial outcomes for children with CHD.

These ambitious priorities highlight the diverse and often unmet mental health needs of families of children with CHD and provide a roadmap for the future of our field. With patients, families, researchers, clinicians, policymakers, and industry leaders working in strong partnership, we are set to change the landscape of mental healthcare – and outcomes – for people of all ages with CHD and their families.

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References

1. Sood E, Jacobs JP, Marino BS. The Cardiac Neurodevelopmental Outcome Collaborative: a new community improving outcomes for individuals with congenital heart disease. Cardiology in the Young. 2020;30(11):1595–1596.
2. Marino BS, Sood E, Cassidy AR, Miller TA, Sanz JH, Bellinger D, Newburger J, Goldberg CS. The origins and development of the Cardiac Neurodevelopmental Outcome Collaborative: creating innovative clinical, quality improvement, and research opportunities. Cardiology in the Young. 2020;30(11):1597–1602.
3. Miller TA, Sadhwani A, Sanz J, Sood E, Ilardi D, Newburger JW, Goldberg CS, Wypij D, Gaynor JW, Marino BS. Variations in practice in cardiac neurodevelopmental follow-up programs. Cardiology in the Young. 2020;30(11):1603–1608.
4. Ware J, Butcher JL, Latal B, Sadhwani A, Rollins CK, Brosig Soto CL, Butler SC, Eiler-Sims PB, Ullman Shade CV, Wernovsky G. Neurodevelopmental evaluation strategies for children with congenital heart disease aged birth through 5 years: recommendations from the cardiac neurodevelopmental outcome collaborative. Cardiology in the Young. 2020;30(11):1609–1622.
5. Ilardi D, Sanz JH, Cassidy AR, Sananes R, Rollins CK, Ullman Shade C, Carroll G, Bellinger DC. Neurodevelopmental evaluation for school-age children with congenital heart disease: recommendations from the cardiac neurodevelopmental outcome collaborative. Cardiology in the Young. 2020;30(11):1623–1636.
6. Sood E, Jacobs JP, Marino BS. Optimizing neurodevelopmental and psychosocial outcomes for survivors with congenital heart disease: a research agenda for the next decade. Cardiology in the Young; 2021;31(6):909–914. This Issue.
7. Sanz JH, Anixt J, Bear L, Basken A, Beca J, Marino BS, Mussatto KA, Nembhard WN, Sadhwani A, Sananes R, Shekerdemian LS, Sood E, Uzark K, Willen E, Ilardi D. Characterization of Neurodevelopmental and Psychological Outcomes in Congenital Heart Disease: A research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative. Cardiology in the Young; 2021;31(6):909–914. This Issue.
8. Cassidy AR, Butler SC, Briend J, Calderon J, Casey F, Crosby LE, Fogel J, Gauthier N, Raimondi C, Marino BS, Sood E, Butcher JL. Neurodevelopmental and psychosocial interventions for individuals with congenital heart disease: A research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative. Cardiology in the Young. 2021;31(6):909–914. This Issue.
9. Mussatto K, Hoffmann R, Hoffman G et al. Risk and Prevalence of Developmental Delay in Young Children With Congenital Heart Disease. Pediatrics. 2014;133(3):e570–e577. doi: 10.1542/peds.2013-2309
10. Gaynor J, Stopp C, Wypij D et al. Neurodevelopmental Outcomes After Cardiac Surgery in Infancy. Pediatrics. 2015;135(5):816–825. doi: 10.1542/peds.2014-3825
11. Verrall CE, Yang JYM, Chen J et al. Neurocognitive Dysfunction and Smaller Brain Volumes in Adolescents and Adults with a Fontan Circulation. Circulation. 2021;143:878–891. https://doi.org/10.1161/CIRCULATIONAHA.120.048202
12. Wernovsky G, Licht D. Neurodevelopmental Outcomes in Children With Congenital Heart Disease—What Can We Impact?. Pediatric Critical Care Medicine. 2016;17:S232–S242. doi: 10.1097/pcc.0000000000000800
13. Ramchandani P, Stein A, O'Connor T, Heron J, Murray L, Evans J. Depression in Men in the Postnatal Period and Later Child Psychopathology: A Population Cohort Study. Journal of the American Academy of Child & Adolescent Psychiatry. 2008;47(4):390–398. doi: 10.1097/chi.0b013e31816429c2
14. National Research Council (US) and Institute of Medicine (US) Committee on Depression, Parenting Practices, and the Healthy Development of Children; England MJ, Sim LJ, editors. Washington (DC): National Academies Press (US); 2009.
15. Bakula D, Sharkey C, Perez M et al. The Relationship Between Parent and Child Distress in Pediatric Cancer: A Meta-Analysis. J Pediatr Psychol. 2019;44(10):1121–1136. doi: 10.1093/jpepsy/jsz051
16. Wray J, Brown K, Tregay J et al. Parents' Experiences of Caring for Their Child at the Time of Discharge After Cardiac Surgery and During the Postdischarge Period: Qualitative Study Using an Online Forum. J Med Internet Res. 2018;20(5):e155. doi: 10.2196/jmir.9104
17. Woolf-King S, Arnold E, Weiss S, Teitel D. "There's no acknowledgement of what this does to people": A qualitative exploration of mental health among parents of children with critical congenital heart defects. J Clin Nurs. 2018;27(13-14):2785–2794. doi: 10.1111/jocn.14275
18. Gregory M, Prouhet P, Russell C, Pfannenstiel B. Quality of Life for Parents of Children With Congenital Heart Defect. J Cardiovasc Nurs. 2018:1. doi: 10.1097/jcn.0000000000000466
19. McDonald HM, Sherman KA, Kasparian NA. Factors associated with psychological distress among Australian women during pregnancy. Pers Individ Differ. 2021;172:110577. https://doi.org/10.1016/j.paid.2020.110577
20. Dennis C, Falah-Hassani K, Shiri R. Prevalence of antenatal and postnatal anxiety: Systematic review and meta-analysis. British Journal of Psychiatry. 2017;210(5):315–323. doi: 10.1192/bjp.bp.116.187179
21. Kasparian N. Heart care before birth: A psychobiological perspective on fetal cardiac diagnosis. Prog Pediatr Cardiol. 2019:101142. doi: 10.1016/j.ppedcard.2019.101142
22. Perry R, Blair C, Sullivan R. Neurobiology of infant attachment: attachment despite adversity and parental programming of emotionality. Curr Opin Psychol. 2017;17:1–6. doi: 10.1016/j.copsyc.2017.04.022
23. Woolf-King S, Anger A, Arnold E, Weiss S, Teitel D. Mental Health Among Parents of Children With Critical Congenital Heart Defects: A Systematic Review. J Am Heart Assoc. 2017;6(2). doi: 10.1161/jaha.116.004862
24. Richardson L, Frueh B, Acierno R. Prevalence Estimates of Combat-Related Post-Traumatic Stress Disorder: Critical Review. Australian & New Zealand Journal of Psychiatry. 2010;44(1):4–19. doi: 10.3109/00048670903393597
25. Sood E, Karpyn A, Demianczyk A et al. Mothers and Fathers Experience Stress of Congenital Heart Disease Differently. Pediatric Critical Care Medicine. 2018:1. doi: 10.1097/pcc.0000000000001528
26. Hoffman MF, Karpyn A, Christofferson J, Neely T, McWhorter LG, Demianczyk AC, James R, Hafer J, Kazak AE, Sood E. Fathers of Children With Congenital Heart Disease: Sources of Stress and Opportunities for Intervention. Pediatr Crit Care Med. 2020;21:e1002–e1009. doi: 10.1097/PCC.0000000000002388.

27. Lisanti A. Parental stress and resilience in CHD: a new frontier for health disparities research. Cardiol Young. 2018;28(9):1142–1150. doi: 10.1017/s1047951118000963
28. Franck L, Mcquillan A, Wray J, Grocott M, Goldman A. Parent Stress Levels During Children's Hospital Recovery After Congenital Heart Surgery. Pediatr Cardiol. 2010;31(7):961–968. doi: 10.1007/s00246-010-9726-5
29. DeMaso D, Labella M, Taylor G et al. Psychiatric Disorders and Function in Adolescents with d-Transposition of the Great Arteries. J Pediatr. 2014;165(4):760–766. doi: 10.1016/j.jpeds.2014.06.029
30. Ernst M, Marino B, Cassedy A et al. Biopsychosocial Predictors of Quality of Life Outcomes in Pediatric Congenital Heart Disease. Pediatr Cardiol. 2017;39(1):79–88. doi: 10.1007/s00246-017-1730-6
31. Denniss D, Sholler G, Costa D, Winlaw D, Kasparian N. Need for Routine Screening of Health-Related Quality of Life in Families of Young Children with Complex Congenital Heart Disease. J Pediatr. 2019;205:21-28.e2. doi: 10.1016/j.jpeds.2018.09.037
32. Visconti K, Saudino K, Rappaport L, Newburger J, Bellinger D. Influence of Parental Stress and Social Support on the Behavioral Adjustment of Children with Transposition of the Great Arteries. Journal of Developmental & Behavioral Pediatrics. 2002;23(5):314–321. doi: 10.1097/00004703-200210000-00003
33. McCusker CG, Armstrong MP, Mullen M, Doherty NN, Casey FA. A sibling-controlled, prospective study of outcomes at home and school in children with severe congenital heart disease. Cardiol Young. 2013 Aug;23(4):507–16. doi: 10.1017/S1047951112001667.
34. Alderfer M, Fiese B, Gold J et al. Evidence-based Assessment in Pediatric Psychology: Family Measures. J Pediatr Psychol. 2007;33(9):1046–1061. doi: 10.1093/jpepsy/jsm083
35. Lewandowski A, Palermo T, Stinson J, Handley S, Chambers C. Systematic Review of Family Functioning in Families of Children and Adolescents With Chronic Pain. The Journal of Pain. 2010;11(11):1027–1038. doi: 10.1016/j.jpain.2010.04.005
36. Berant E, Mikulincer M, Florian V. Marital satisfaction among mothers of infants with congenital heart disease: The contribution of illness severity, attachment style, and the coping process. Anxiety Stress Coping. *2003*;*16*(4):397-415.
37. Dale MT, Solberg Ø, Holmstrøm H, Landolt MA, Eskedal LT, Vollrath ME. Relationship satisfaction among mothers of children with congenital heart defects: A prospective case-cohort study. J Pediatr Psychol. 2013;38(8):915–926.
38. Jordan B, Franich-Ray C, Albert N et al. Early mother-infant relationships after cardiac surgery in infancy. Arch Dis Child. 2014;99(7):641–645. doi: 10.1136/archdischild-2012-303488
39. Elhoff J, McHugh K, Buckley J, Morris S, Simpson K, Scheurer M. Out-of-pocket medical expenses in severe CHD. Cardiol Young. 2018;28(8):1014–1018. doi: 10.1017/s1047951118000768
40. Connor J, Kline N, Mott S, Harris S, Jenkins K. The Meaning of Cost for Families of Children With Congenital Heart Disease. Journal of Pediatric Health Care. 2010;24(5):318–325. doi: 10.1016/j.pedhc.2009.09.002
41. Utens E, Callus E, Levert E, Groote K, Casey F. Multidisciplinary family-centred psychosocial care for patients with CHD: consensus recommendations from the AEPC Psychosocial Working Group. Cardiol Young. 2017;28(2):192–198. doi: 10.1017/s1047951117001378
42. Kasparian N, Winlaw D, Sholler G. "Congenital heart health": how psychological care can make a difference. Medical Journal of Australia. 2016;205(3):104–107. doi: 10.5694/mja16.00392
43. American Board of Pediatrics Roadmap Project. Supporting Resilience, Emotional, and Mental Health of Pediatric Patients with Chronic Conditions and Their Families. July 26, 2018. Accessed March 9, 2021. https://www.abp.org/sites/abp/files/pdf/bmh-change-package.pdf
44. Kasparian N, Kan J, Sood E, Wray J, Pincus H, Newburger J. Mental health care for parents of babies with congenital heart disease during intensive care unit admission: Systematic review and statement of best practice. Early Hum Dev. 2019;139:104837. doi: 10.1016/j.earlhumdev.2019.104837
45. Tesson S, Butow P, Sholler G, Sharpe L, Kovacs A, Kasparian N. Psychological interventions for people affected by childhood-onset heart disease: A systematic review. Health Psychology. 2019;38(2):151–161. doi: 10.1037/hea0000704
46. Gramszlo C, Karpyn A, Demianczyk A et al. Parent Perspectives on Family-Based Psychosocial Interventions for Congenital Heart Disease. J Pediatr. 2019. doi: 10.1016/j.jpeds.2019.09.059
47. DeMaso D, Campis L, Wypij D, Bertram S, Lipshitz M, Freed M. The Impact of Maternal Perceptions and Medical Severity on the Adjustment of Children with Congenital Heart Disease. J Pediatr Psychol. 1991;16(2):137–149. doi: 10.1093/jpepsy/16.2.137
48. Goldberg S, Janus M, Washington J, Simmons R, MacLusky I, Fowler R. Prediction of Preschool Behavioral Problems in Healthy and Pediatric Samples. Journal of Developmental & Behavioral Pediatrics. 1997;18(5):304–313. doi: 10.1097/00004703-199710000-00004
49. Marshall KH, d'Udekem Y, Sholler GF, et al. Health-related quality of life in children, adolescents and adults with a Fontan circulation: A meta-analysis. J Am Heart Assoc. 2020;9(6):e014172.
50. Laing S, McMahon C, Ungerer J, Taylor A, Badawi N, Spence K. Mother-child interaction and child developmental capacities in toddlers with major birth defects requiring newborn surgery. *Early Hum Dev*. 2010 Dec;86(12):793–800. doi: 10.1016/j.earlhumdev.2010.08.025. PMID: 20888152.
51. Kingston D, Tough S. Prenatal and postnatal maternal mental health and school-age child development: a systematic review. Matern Child Health J. 2014; 18(7):1728–1741.
52. Kingston D, Tough S, Whitfield H. Prenatal and postpartum maternal psychological distress and infant development: a systematic review. Child Psychiatry Hum Dev. 2012; 43(5):683–714.
53. Cook N, Ayers S, Horsch A. Maternal posttraumatic stress disorder during the perinatal period and child outcomes: A systematic review. J Affect Disord. 2018; 225:18–31.
54. Mussatto K. Adaptation of the child and family to life with a chronic illness. Cardiol Young. 2006;16(S3):110–116. doi: 10.1017/s104795110600103x
55. Lawoko S. Factors influencing satisfaction and well-being among parents of congenital heart disease children: development of a conceptual model based on the literature review. Scand J Caring Sci. 2007;21(1):106–117. doi: 10.1111/j.1471-6712.2007.00444.x
56. Lisanti A, Golfenshtein N, Medoff-Cooper B. The Pediatric Cardiac Intensive Care Unit Parental Stress Model. Advances in Nursing Science. 2017;40(4):319–336. doi: 10.1097/ans.0000000000000184
57. Clarke K, Cooper P, Creswell C. The Parental Overprotection Scale: Associations with child and parental anxiety. J Affect Disord. 2013;151(2):618–624. doi: 10.1016/j.jad.2013.07.007
58. Christie H, Hamilton-Giachritsis C, Alves-Costa F, Tomlinson M, Halligan S. The impact of parental posttraumatic stress disorder on parenting: a systematic review. Eur J Psychotraumatol. 2019;10(1):1550345. doi: 10.1080/20008198.2018.1550345
59. McCubbin H, Patterson J. The Family Stress Process. Marriage Fam Rev. 1983;6(1-2):7–37. doi: 10.1300/j002v06n01_02
60. Bandura A. Self-efficacy mechanism in human agency. American Psychologist. 1982;37(2):122–147. doi: 10.1037/0003-066x.37.2.122
61. Bretherton I. The origins of attachment theory: John Bowlby and Mary Ainsworth. Dev Psychol. 1992;28(5):759–775. doi: 10.1037/0012-1649.28.5.759
62. Abidin R. The Determinants of Parenting Behavior. J Clin Child Psychol. 1992;21(4):407–412. doi: 10.1207/s15374424jccp2104_12
63. Carver C, Scheier M. Control theory: A useful conceptual framework for personality-social, clinical, and health psychology. Psychol Bull. 1982;92(1):111–135. doi: 10.1037//0033-2909.92.1.111
64. Goodman S, Gotlib I. Risk for psychopathology in the children of depressed mothers: A developmental model for understanding mechanisms of transmission. Psychol Rev. 1999;106(3):458–490. doi: 10.1037//0033-295x.106.3.458
65. Saloviita T, Italinna M, Leinonen E. Explaining the parental stress of fathers and mothers caring for a child with intellectual disability: a Double ABCX Model. Journal of Intellectual Disability Research. 2003;47(4-5):300–312. doi: 10.1046/j.1365-2788.2003.00492.x
66. Hill C, Rose J. Parenting Stress Models and Their Application to Parents of Adults with Intellectual Disabilities. The British Journal

of Development Disabilities. 2010;56(110):19–37. doi: 10.1179/096979510799103023
67. Raina P. The Health and Well-Being of Caregivers of Children With Cerebral Palsy. Pediatrics. 2005;115(6):e626–e636. doi: 10.1542/peds.2004-1689
68. Melnyk B, Crean H, Feinstein N, Fairbanks E. Maternal Anxiety and Depression After a Premature Infant's Discharge From the Neonatal Intensive Care Unit. Nurs Res. 2008;57(6):383–394. doi: 10.1097/nnr.0b013e3181906f59
69. Lee Y, Garfield C, Kim H. Self-Efficacy Theory as a Framework For Interventions That Support Parents of NICU Infants. *Proceedings of the 6th International Conference on Pervasive Computing Technologies for Healthcare*. 2012. doi: 10.4108/icst.pervasivehealth.2012.248710
70. Pakenham K, Samios C, Sofronoff K. Adjustment in mothers of children with Asperger syndrome. Autism. 2005;9(2):191–212. doi: 10.1177/1362361305049033
71. Hocking M, Lochman J. Applying the Transactional Stress and Coping Model to Sickle Cell Disorder and Insulin-Dependent Diabetes Mellitus: Identifying Psychosocial Variables Related to Adjustment and Intervention. Clin Child Fam Psychol Rev. 2005;8(3):221–246. doi: 10.1007/s10567-005-6667-2
72. Scheeringa M, Zeanah C. A relational perspective on PTSD in early childhood. J Trauma Stress. 2001;14(4):799–815. doi: 10.1023/a:1013002507972
73. Creech S, Misca G. Parenting with PTSD: A Review of Research on the Influence of PTSD on Parent-Child Functioning in Military and Veteran Families. Front Psychol. 2017;8. doi: 10.3389/fpsyg.2017.01101
74. Goodman MS, Sanders Thompson VL. The science of stakeholder engagement in research: classification, implementation, and evaluation. Transl Behav Med. 2017;7(3):486–491. doi: 10.1007/s13142-017-0495-z
75. Gómez L, Schalock R, Verdugo M. The Role of Moderators and Mediators in Implementing and Evaluating Intellectual and Developmental Disabilities-Related Policies and Practices. J Dev Phys Disabil. 2019. doi: 10.1007/s10882-019-09702-3
76. Gridley N, Blower S, Dunn A, Bywater T, Bryant M. Psychometric Properties of Child (0–5 Years) Outcome Measures as used in Randomized Controlled Trials of Parent Programs: A Systematic Review. Clin Child Fam Psychol Rev. 2019;22(3):388–405. doi: 10.1007/s10567-019-00277-1
77. Blower S, Gridley N, Dunn A, Bywater T, Hindson Z, Bryant M. Psychometric Properties of Parent Outcome Measures Used in RCTs of Antenatal and Early Years Parent Programs: A Systematic Review. Clin Child Fam Psychol Rev. 2019;22(3):367–387. doi: 10.1007/s10567-019-00276-2
78. Mesman J, Emmen R. Mary Ainsworth's legacy: a systematic review of observational instruments measuring parental sensitivity. Attach Hum Dev. 2013;15(5-6):485–506. doi: 10.1080/14616734.2013.820900
79. Pinheiro L, McFatrich M, Lucas N et al. Child and adolescent self-report symptom measurement in pediatric oncology research: a systematic literature review. Quality of Life Research. 2017;27(2):291–319. doi: 10.1007/s11136-017-1692-4
80. Hodes R, Insel T, Landis S. The NIH Toolbox: Setting a standard for biomedical research. Neurology. 2013;80(Issue 11, Supplement 3):S1-S1. doi: 10.1212/wnl.0b013e3182872e90
81. Miles M, Funk S, Carlson J. Parental Stressor Scale. Nurs Res. 1993;42(3):148–152. doi: 10.1097/00006199-199305000-00005
82. Miles M, Brunssen S. Psychometric properties of the parental stressor scale: infant hospitalization. Advances in Neonatal Care. 2003;3(4):189–196. doi: 10.1016/s1536-0903(03)00138-3
83. F. W. Weathers, B. T. Litz, J. A. Huska, and T. M. Keane, The PTSD Checklist—Civilian Version (PCL-C), National Center for PTSD, Boston, Mass, USA, 1994.
84. Reader S, Ruppe N, Deatrick J et al. Caregiver perspectives on family psychosocial risks and resiliencies in pediatric sickle cell disease: Informing the adaptation of the Psychosocial Assessment Tool. Clin Pract Pediatr Psychol. 2017;5(4):330–341. doi: 10.1037/cpp0000208
85. Corwin E, Ferranti E. Integration of biomarkers to advance precision nursing interventions for family research across the life span. Nurs Outlook. 2016;64(4):292–298. doi: 10.1016/j.outlook.2016.04.007
86. McEwen B. Protective and Damaging Effects of Stress Mediators. New England Journal of Medicine. 1998;338(3):171–179. doi: 10.1056/nejm199801153380307
87. McEwen B. Stress, Adaptation, and Disease: Allostasis and Allostatic Load. Ann N Y Acad Sci. 1998;840(1):33–44. doi: 10.1111/j.1749-6632.1998.tb09546.x
88. Seeman T, McEwen B, Rowe J, Singer B. Allostatic load as a marker of cumulative biological risk: MacArthur studies of successful aging. Proceedings of the National Academy of Sciences. 2001;98(8):4770–4775. doi: 10.1073/pnas.081072698
89. Juster R, McEwen B, Lupien S. Allostatic load biomarkers of chronic stress and impact on health and cognition. Neuroscience & Biobehavioral Reviews. 2010;35(1):2–16. doi: 10.1016/j.neubiorev.2009.10.002
90. Carazo M, Kolodziej MS, DeWitt E, et al. Prevalence and prognostic association of a clinical diagnosis of depression in adults with congenital heart disease: Results of the Boston Adult Congenital Heart Disease Biobank. J Am Heart Assoc. 2020;28:e014820.
91. El-Farhan N, Rees D, Evans C. Measuring cortisol in serum, urine and saliva – are our assays good enough?. Ann Clin Biochem. 2017;54(3):308–322. doi: 10.1177/0004563216687335
92. Izawa S, Miki K, Tsuchiya M et al. Cortisol level measurements in fingernails as a retrospective index of hormone production. Psychoneuroendocrinology. 2015;54:24–30. doi: 10.1016/j.psyneuen.2015.01.015
93. Liu R. The microbiome as a novel paradigm in studying stress and mental health. American Psychologist. 2017;72(7):655–667. doi: 10.1037/amp0000058
94. Stalder T, Steudte-Schmiedgen S, Alexander N et al. Stress-related and basic determinants of hair cortisol in humans: A meta-analysis. Psychoneuroendocrinology. 2017;77:261–274. doi: 10.1016/j.psyneuen.2016.12.017
95. Iliadis SI, Comasco E, Sylvén S, et al. Prenatal and Postpartum Evening Salivary Cortisol Levels in Association with Peripartum Depressive Symptoms. PLoS One. 2015; 10(8):e0135471. doi: 10.1371/journal.pone.0135471.
96. Wu Y, Kapse K, Jacobs M, et al. Association of Maternal Psychological Distress With In Utero Brain Development in Fetuses With Congenital Heart Disease. JAMA Pediatr. 2020;174(3):e195316. doi: 10.1001/jamapediatrics.2019.5316.
97. Butler E, Randall A. Emotional Coregulation in Close Relationships. Emotion Review. 2012;5(2):202–210. doi: 10.1177/1754073912451630
98. Matthews S. Early programming of the hypothalamo–pituitary–adrenal axis. Trends in Endocrinology and Metabolism. 2002;13(9):373–380. doi: 10.1016/s1043-2760(02)00690-2
99. Cook N, Ayers S, Horsch A. Maternal posttraumatic stress disorder during the perinatal period and child outcomes: A systematic review. J Affect Disord. 2018;225:18–31. doi: 10.1016/j.jad.2017.07.045
100. Polanska K, Krol A, Merecz-Kot D et al. Maternal stress during pregnancy and neurodevelopmental outcomes of children during the first 2 years of life. J Paediatr Child Health. 2017;53(3):263–270. doi: 10.1111/jpc.13422
101. Gilles M, Otto H, Wolf I et al. Maternal hypothalamus-pituitary-adrenal (HPA) system activity and stress during pregnancy: Effects on gestational age and infant's anthropometric measures at birth. Psychoneuroendocrinology. 2018;94:152–161. doi: 10.1016/j.psyneuen.2018.04.022
102. Butler E, Randall A. Emotional Coregulation in Close Relationships. Emotion Review. 2012;5(2):202–210. doi: 10.1177/1754073912451630
103. Feldman R, Golan O, Hirschler-Guttenberg Y, Ostfeld-Etzion S, Zagoory-Sharon O. Parent–child interaction and oxytocin production in preschoolers with autism spectrum disorder. British Journal of Psychiatry. 2014;205(2):107–112. doi: 10.1192/bjp.bp.113.137513
104. Feldman R, Gordon I, Zagoory-Sharon O. Maternal and paternal plasma, salivary, and urinary oxytocin and parent-infant synchrony: considering stress and affiliation components of human bonding. Dev Sci. 2010;14(4):752–761. doi: 10.1111/j.1467-7687.2010.01021.x
105. Lisanti AJ, Demianczyk AC, Costarino A, et al. Skin-to-Skin Care Reduces Stress, Anxiety, and Salivary Cortisol while Supporting Attachment in

https://doi.org/10.1017/S1047951121002134 Published online by Cambridge University Press

Mothers of Infants with Critical Congenital Heart Disease. J Obstet Gynecol Neonatal Nurs. *2021;50,* 40–54. doi: 10.1016/j.jogn.2020.09.154
106. Rogers G, Keating D, Young R, Wong M, Licinio J, Wesselingh S. From gut dysbiosis to altered brain function and mental illness: mechanisms and pathways. Mol Psychiatry. 2016;21(6):738–748. doi: 10.1038/mp.2016.50
107. Kan J, Cowan C, Ooi C, Kasparian N. What can the gut microbiome teach us about the connections between child physical and mental health? A systematic review. Dev Psychobiol. 2019;61(5):700–713. doi: 10.1002/dev.21819
108. Cashion AK, Grady PA. The National Institutes of Health/National Institutes of Nursing Research intramural research program and the development of the National Institutes of Health Symptom Science Model. Nurs Outlook. 2015;63(4):484–7. doi: 10.1016/j.outlook.2015.03.001.
109. Lisanti AJ, Demianczyk AC, Costarino A, et al. Skin to Skin Care is a Safe and Effective Comfort Measure for Infants before and after Neonatal Cardiac Surgery. Pediatr Crit Care Med. 2020 Sep;21(9):e834–e841. doi: 10.1097/PCC.0000000000002493.
110. Harrison TM, Brown R. Autonomic nervous system function after a skin-to-skin contact intervention in infants with congenital heart disease. J Cardiovasc Nurs. 2017;32:E1–E13
111. Harrison TM, Chen CY, Stein P, et al. Neonatal skin-to-skin contact: Implications for learning and autonomic nervous system function in infants with congenital heart disease. Biol Res Nurs 2019; 21:296–306
112. Solberg Ø, Dale M, Holmstrøm H, Eskedal L, Landolt M, Vollrath M. Emotional reactivity in infants with congenital heart defects and maternal symptoms of postnatal depression. Arch Womens Ment Health. 2011;14(6):487–492. doi: 10.1007/s00737-011-0243-1
113. Solberg Ø, Gronning Dale M, Holmstrom H, Eskedal L, Landolt M, Vollrath M. Trajectories of Maternal Mental Health: A Prospective Study of Mothers of Infants With Congenital Heart Defects From Pregnancy to 36 Months Postpartum. J Pediatr Psychol. 2012;37(6):687–696. doi: 10.1093/jpepsy/jss044
114. Helfricht S, Latal B, Fischer J, Tomaske M, Landolt M. Surgery-related posttraumatic stress disorder in parents of children undergoing cardiopulmonary bypass surgery: A prospective cohort study. *Pediatric Critical Care Medicine.* 2008;9(2):217–223. doi: 10.1097/pcc.0b013e318166eec3
115. Brosig C, Whitstone B, Frommelt M, Frisbee S, Leuthner S. Psychological distress in parents of children with severe congenital heart disease: the impact of prenatal versus postnatal diagnosis. Journal of Perinatology. 2007;27(11):687–692. doi: 10.1038/sj.jp.7211807
116. Streisand R, Kazak A, Tercyak K. Pediatric-Specific Parenting Stress and Family Functioning in Parents of Children Treated for Cancer. Children's Health Care. 2003;32(4):245–256. doi: 10.1207/s15326888chc3204_1
117. Cousino M, Hazen R. Parenting Stress Among Caregivers of Children With Chronic Illness: A Systematic Review. J Pediatr Psychol. 2013;38(8):809–828. doi: 10.1093/jpepsy/jst049
118. Lisanti A, Allen L, Kelly L, Medoff-Cooper B. Maternal Stress and Anxiety in the Pediatric Cardiac Intensive Care Unit. American Journal of Critical Care. 2017;26(2):118–125. doi: 10.4037/ajcc2017266
119. March S. Parents' perceptions during the transition to home for their child with a congenital heart defect: How can we support families of children with hypoplastic left heart syndrome?. Journal for Specialists in Pediatric Nursing. 2017;22(3):e12185. doi: 10.1111/jspn.12185
120. McCusker C, Doherty N, Molloy B et al. A Randomized Controlled Trial of Interventions to Promote Adjustment in Children With Congenital Heart Disease Entering School and Their Families. J Pediatr Psychol. 2012;37(10):1089–1103. doi: 10.1093/jpepsy/jss092
121. Hartman D, Medoff-Cooper B. Transition to Home After Neonatal Surgery for Congenital Heart Disease. MCN, The American Journal of Maternal/Child Nursing. 2012;37(2):95–100. doi: 10.1097/nmc.0b013e318241dac1
122. McKean E, Kasparian N, Batra S, Sholler G, Winlaw D, Dalby-Payne J. Feeding difficulties in neonates following cardiac surgery: determinants of prolonged feeding-tube use. Cardiol Young. 2017;27(6):1203–1211. doi: 10.1017/s1047951116002845
123. Kaugars A, Shields C, Brosig C. Stress and quality of life among parents of children with congenital heart disease referred for psychological services. Congenit Heart Dis. 2017;13(1):72–78. doi: 10.1111/chd.12547
124. Wray J, Cassedy A, Ernst M, Franklin R, Brown K, Marino B. Psychosocial functioning of parents of children with heart disease—describing the landscape. Eur J Pediatr. 2018;177(12):1811–1821. doi: 10.1007/s00431-018-3250-7
125. Doherty N, McCusker C, Molloy B et al. Predictors of psychological functioning in mothers and fathers of infants born with severe congenital heart disease. J Reprod Infant Psychol. 2009;27(4):390–400. doi: 10.1080/02646830903190920
126. Davis C, Brown R, Bakeman R, Campbell R. Psychological Adaptation and Adjustment of Mothers of Children With Congenital Heart Disease: Stress, Coping, and Family Functioning. J Pediatr Psychol. 1998;23(4):219–228. doi: 10.1093/jpepsy/23.4.219
127. Li Y, Cao F, Cao D, Wang Q, Cui N. Predictors of posttraumatic growth among parents of children undergoing inpatient corrective surgery for congenital disease. J Pediatr Surg. 2012;47(11):2011–2021. doi: 10.1016/j.jpedsurg.2012.07.005
128. Sorlie P, Wei G. Population-Based Cohort Studies: Still Relevant?. J Am Coll Cardiol. 2011;58(19):2010–2013. doi: 10.1016/j.jacc.2011.08.020
129. Forrest C, Margolis P, Bailey L et al. PEDSnet: a National Pediatric Learning Health System. Journal of the American Medical Informatics Association. 2014;21(4):602–606. doi: 10.1136/amiajnl-2014-002743
130. Strange G, Stewart S, Farthing M et al. Living With, and Caring for, Congenital Heart Disease in Australia: Insights From the Congenital Heart Alliance of Australia and New Zealand Online Survey. Heart, Lung and Circulation. 2019. doi: 10.1016/j.hlc.2018.12.009
131. Celermajer D, Strange G, Cordina R et al. Congenital Heart Disease Requires a Lifetime Continuum of Care: A Call for a Regional Registry. Heart, Lung and Circulation. 2016;25(8):750–754. doi: 10.1016/j.hlc.2016.03.018
132. Schilling C, Dalziel K, Nunn R et al. The Fontan epidemic: Population projections from the Australia and New Zealand Fontan Registry. Int J Cardiol. 2016;219:14–19. doi: 10.1016/j.ijcard.2016.05.035
133. Alsaied T, Allen KY, Anderson JB et al. The Fontan outcomes network: first steps towards building a lifespan registry for individuals with Fontan circulation in the United States. Cardiol Young. 2020 Aug;30(8):1070–1075. doi: 10.1017/S1047951120001869.
134. Anderson JB, Brown DW, Lihn S et al. Power of a Learning Network in Congenital Heart Disease. World J Pediatr Congenit Heart Surg. 2019;10(1):66–71. doi: 10.1177/2150135118815023.
135. Loccoh EC, Yu S, Donohue J, Lowery R, et al. Prevalence and risk factors associated with non-attendance in neurodevelopmental follow-up clinic among infants with CHD. Cardiol Young. 2018;28(4):554–560. doi: 10.1017/S1047951117002748.
136. Gonzalez VJ, Kimbro RT, Cutitta KE, et al. Mental Health Disorders in Children With Congenital Heart Disease. Pediatrics. 2021;147(2):e20201693. doi: 10.1542/peds.2020-1693.
137. Alderfer M, Sood E. Using qualitative research methods to improve clinical care in pediatric psychology. Clin Pract Pediatr Psychol. 2016;4(4):358–361. doi: 10.1037/cpp0000164
138. Batalden M, Batalden P, Margolis P, Seid M, Armstrong G, Opipari-Arrigan L, Hartung H. Coproduction of healthcare service. BMJ Qual Saf. 2016 Jul;25(7):509–17. doi: 10.1136/bmjqs-2015-004315.
139. Sood E, Wysocki T, Alderfer M, et al. Topical Review: Crowdsourcing as a Novel Approach to Qualitative Research. J Pediatr Psychol. 2021;46:189–196.
140. Demianczyk A, Behere S, Thacker D et al. Social Risk Factors Impact Hospital Readmission and Outpatient Appointment Adherence for Children with Congenital Heart Disease. J Pediatr. 2019;205:35-40.e1. doi: 10.1016/j.jpeds.2018.09.038
141. Earls MF, Yogman MW, Mattson G, Rafferty J; Committee on Psychosocial Aspects of Child and Family Health. Incorporating Recognition and Management of Perinatal Depression Into Pediatric Practice. Pediatrics. 2019 Jan;143(1):e20183260. doi: 10.1542/peds.2018-3260.
142. National Strategic Action Plan for Childhood Heart Disease. February 2019. Accessed March 9, 2021. https://www.heartkids.org.au/congenital-heart-disease/national-strategic-action-plan-for-chd/chd-action-plan.
143. Kazak A, Schneider S, Didonato S, Pai A. Family psychosocial risk screening guided by the Pediatric Psychosocial Preventative Health Model

(PPPHM) using the Psychosocial Assessment Tool (PAT). Acta Oncol (Madr). 2015;54(5):574–580. doi: 10.3109/0284186x.2014.995774
144. Haverman L, van Oers H, Limperg P et al. Development and Validation of the Distress Thermometer for Parents of a Chronically Ill Child. J Pediatr. 2013;163(4):1140-1146.e2. doi: 10.1016/j.jpeds.2013.06.011
145. Cole J, Olkkola M, Zarrin H, Berger K, Moldenhauer J. Universal Postpartum Mental Health Screening for Parents of Newborns With Prenatally Diagnosed Birth Defects. Journal of Obstetric, Gynecologic & Neonatal Nursing. 2018;47(1):84–93. doi: 10.1016/j.jogn.2017.04.131
146. Hearps S, McCarthy M, Muscara F et al. Psychosocial risk in families of infants undergoing surgery for a serious congenital heart disease. Cardiol Young. 2013;24(4):632–639. doi: 10.1017/s1047951113000760
147. McCarthy M, Hearps S, Muscara F et al. Family Psychosocial Risk Screening in Infants and Older Children in the Acute Pediatric Hospital Setting Using the Psychosocial Assessment Tool. J Pediatr Psychol. 2016;41(7):820–829. doi: 10.1093/jpepsy/jsw055
148. Cousino M, Schumacher K, Rea K et al. Psychosocial functioning in pediatric heart transplant recipients and their families. Pediatr Transplant. 2018;22(2):e13110. doi: 10.1111/petr.13110
149. Austin M-P, Highet N & Expert Working Group. Mental Health Care in the Perinatal Period: Australian Clinical Practice Guideline. Melbourne: Centre of Perinatal Excellence; 2017.
150. Babor TF, Del Boca F, Bray JW. Screening, Brief Intervention and Referral to Treatment: implications of SAMHSA's SBIRT initiative for substance abuse policy and practice. Addiction. 2017 Feb;112 Suppl 2:110–117. doi: 10.1111/add.13675. PMID: 28074569.
151. Cella D, Riley W, Stone AA et al. The Patient Reported Outcomes Measurement Information System (PROMIS) developed and tested its first wave of adult self reported health outcome item banks: 2005-2008. J Clin Epidemiol. 2010;63:1179–1194.
152. Van Schoors M, Caes L, Verhofstadt L, Goubert L, Alderfer M. Systematic Review: Family Resilience After Pediatric Cancer Diagnosis: Figure 1. J Pediatr Psychol. 2015;40(9):856–868. doi: 10.1093/jpepsy/jsv055
153. Faccio F, Renzi C, Giudice A, Pravettoni G. Family Resilience in the Oncology Setting: Development of an Integrative Framework. Front Psychol. 2018;9. doi: 10.3389/fpsyg.2018.00666
154. Pittman J, Floto E, Lindamer L, Baker D, Lohr J, Afari N. VA eScreening program: Technology to improve care for post-9/11 veterans. Psychol Serv. 2017;14(1):23–33. doi: 10.1037/ser0000125
155. Hashemi B, Ali S, Awaad R, Soudi L, Housel L, Sosebee S. Facilitating mental health screening of war-torn populations using mobile applications. Soc Psychiatry Psychiatr Epidemiol. 2017;52(1):27–33. doi: 10.1007/s00127-016-1303-7
156. Schepers S, Sint Nicolaas S, Haverman L et al. Real-world implementation of electronic patient-reported outcomes in outpatient pediatric cancer care. Psychooncology. 2016;26(7):951–959. doi: 10.1002/pon.4242
157. Wissow L, Anthony B, Brown J et al. A Common Factors Approach to Improving the Mental Health Capacity of Pediatric Primary Care. Administration and Policy in Mental Health and Mental Health Services Research. 2008;35(4):305–318. doi: 10.1007/s10488-008-0178-7
158. Shemesh E, Lewis B, Rubes M et al. Mental Health Screening Outcomes in a Pediatric Specialty Care Setting. J Pediatr. 2016;168:193-197.e3. doi: 10.1016/j.jpeds.2015.09.046
159. Struemph K, Barhight L, Thacker D, Sood E. Systematic psychosocial screening in a paediatric cardiology clinic: clinical utility of the Pediatric Symptom Checklist 17. Cardiol Young. 2015;26(6):1130–1136. doi: 10.1017/s1047951115001900
160. Gordon T, Cardone I, Kim J, Gordon S, Silver R. Universal Perinatal Depression Screening in an Academic Medical Center. Obstetrics & Gynecology. 2006;107(2, Part 1):342-347. doi: 10.1097/01.aog.0000194080.18261.92
161. Neubauer K, Williams E, Donohue P, Boss R. Communication and decision-making regarding children with critical cardiac disease: a systematic review of family preferences. Cardiol Young. 2018;28(10):1088–1092. doi: 10.1017/s1047951118001233
162. Pagel C, Bull C, Utley M et al. Exploring communication between parents and clinical teams following children's heart surgery: a survey in the UK. BMJ Paediatr Open. 2019;3(1):e000391. doi: 10.1136/bmjpo-2018-000391
163. Law E, Fisher E, Fales J, Noel M, Eccleston C. Systematic Review and Meta-Analysis of Parent and Family-Based Interventions for Children and Adolescents With Chronic Medical Conditions. J Pediatr Psychol. 2014;39(8):866–886. doi: 10.1093/jpepsy/jsu032
164. Blankenship A, Harrison S, Brandt S, Joy B, Simsic J. Increasing Parental Participation During Rounds in a Pediatric Cardiac Intensive Care Unit. American Journal of Critical Care. 2015;24(6):532–538. doi: 10.4037/ajcc2015153
165. Zurca A, Wang J, Cheng Y, Dizon Z, October T. Racial Minority Families' Preferences for Communication in Pediatric Intensive Care Often Overlooked. J Natl Med Assoc. 2019. doi: 10.1016/j.jnma.2019.09.005
166. Czajkowski S, Powell L, Adler N et al. From ideas to efficacy: The ORBIT model for developing behavioral treatments for chronic diseases. Health Psychology. 2015;34(10):971–982. doi: 10.1037/hea0000161
167. Carroll KM, Rounsaville BJ. Bridging the gap: A hybrid model to link efficacy and effectiveness research in substance abuse treatment. Psychiatr Serv. 2003;54(3):333–339.
168. Tesson S, Swinsburg D, Kasparian NA. Maintaining Momentum in Infant Mental Health Research During COVID-19: Adapting Observational Assessments. J Pediatr Psychol. 2021; In Press.
169. Lauer M, D'Agostino R. The Randomized Registry Trial — The Next Disruptive Technology in Clinical Research?. New England Journal of Medicine. 2013;369(17):1579–1581. doi: 10.1056/nejmp1310102
170. Jotzo M, Poets C. Helping Parents Cope With the Trauma of Premature Birth: An Evaluation of a Trauma-Preventive Psychological Intervention. Pediatrics. 2005;115(4):915–919. doi: 10.1542/peds.2004-0370
171. Mendelson T, Cluxton-Keller F, Vullo G, Tandon S, Noazin S. NICU-based Interventions To Reduce Maternal Depressive and Anxiety Symptoms: A Meta-analysis. Pediatrics. 2017;139(3):e20161870. doi: 10.1542/peds.2016-1870
172. Shaw R, St John N, Lilo E et al. Prevention of Traumatic Stress in Mothers With Preterm Infants: A Randomized Controlled Trial. Pediatrics. 2013;132(4):e886–e894. doi: 10.1542/peds.2013-1331
173. Sockol L. A systematic review of the efficacy of cognitive behavioral therapy for treating and preventing perinatal depression. J Affect Disord. 2015;177:7–21. doi: 10.1016/j.jad.2015.01.052
174. Newby J, McKinnon A, Kuyken W, Gilbody S, Dalgleish T. Systematic review and meta-analysis of transdiagnostic psychological treatments for anxiety and depressive disorders in adulthood. Clin Psychol Rev. 2015;40:91–110. doi: 10.1016/j.cpr.2015.06.002
175. Mendelson T, McAfee C, Damian A, Brar A, Donohue P, Sibinga E. A mindfulness intervention to reduce maternal distress in neonatal intensive care: a mixed methods pilot study. Arch Womens Ment Health. 2018;21(6):791–799. doi: 10.1007/s00737-018-0862-x
176. Golfenshtein N, Deatrick J, Lisanti A, Medoff-Cooper B. Coping with the Stress in the Cardiac Intensive Care Unit: Can Mindfulness Be the Answer?. J Pediatr Nurs. 2017;37:117–126. doi: 10.1016/j.pedn.2017.08.021
177. Cassidy A. Cognitive flexibility in critical CHD: a target for intervention. Cardiol Young. 2020; 30(8):1061–1069.
178. O'Brien K, Robson K, Bracht M, Cruz M, Lui K, Alvaro R, da Silva O, Monterrosa L, Narvey M, Ng E, Soraisham A, Ye XY, Mirea L, Tarnow-Mordi W, Lee SK; FICare Study Group and FICare Parent Advisory Board. Effectiveness of Family Integrated Care in neonatal intensive care units on infant and parent outcomes: a multicentre, multinational, cluster-randomised controlled trial. Lancet Child Adolesc Health. 2018 Apr;2(4):245–254. doi: 10.1016/S2352-4642(18)30039-7.
179. Lebowitz E, Marin C, Martino A, Shimshoni Y, Silverman W. Parent-Based Treatment as Efficacious as Cognitive-Behavioral Therapy for Childhood Anxiety: A Randomized Noninferiority Study of Supportive Parenting for Anxious Childhood Emotions. Journal of the American Academy of Child & Adolescent Psychiatry. 2019. doi: 10.1016/j.jaac.2019.02.014

180. Nowak C, Heinrichs N. A Comprehensive Meta-Analysis of Triple P-Positive Parenting Program Using Hierarchical Linear Modeling: Effectiveness and Moderating Variables. Clin Child Fam Psychol Rev. 2008;11(3):114–144. doi: 10.1007/s10567-008-0033-0
181. McEachern A, Fosco G, Dishion T, Shaw D, Wilson M, Gardner F. Collateral benefits of the family check-up in early childhood: Primary caregivers' social support and relationship satisfaction. Journal of Family Psychology. 2013;27(2):271–281. doi: 10.1037/a0031485
182. Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. Developing and evaluating complex interventions: the new Medical Research Council guidance. BMJ. 2008:a1655. doi: 10.1136/bmj.a1655
183. Developing and evaluating complex interventions. Mrc.ukri.org. https://mrc.ukri.org/documents/pdf/complex-interventions-guidance/. Published 2019. Accessed December 9, 2019.
184. Marsiglia F, Booth J. Cultural Adaptation of Interventions in Real Practice Settings. Res Soc Work Pract. 2014;25(4):423–432. doi: 10.1177/1049731514535989
185. Adams R, Levy S. Shared Decision-Making and Children With Disabilities: Pathways to Consensus. Pediatrics. 2017;139(6):e20170956. doi: 10.1542/peds.2017-0956
186. Perez Jolles M, Richmond J, Thomas K. Minority patient preferences, barriers, and facilitators for shared decision-making with health care providers in the USA: A systematic review. Patient Educ Couns. 2019;102(7):1251–1262. doi: 10.1016/j.pec.2019.02.003
187. Jordan A, Wood F, Edwards A, Shepherd V, Joseph-Williams N. What adolescents living with long-term conditions say about being involved in decision-making about their healthcare: A systematic review and narrative synthesis of preferences and experiences. Patient Educ Couns. 2018;101(10):1725–1735. doi: 10.1016/j.pec.2018.06.006
188. Hall S, Ryan D, Beatty J, Grubbs L. Recommendations for peer-to-peer support for NICU parents. Journal of Perinatology. 2015;35(S1):S9–S13. doi: 10.1038/jp.2015.143
189. Moola F, Faulkner G, White L, Kirsh J. The psychological and social impact of camp for children with chronic illnesses: a systematic review update. Child Care Health Dev. 2013;40(5):615–631. doi: 10.1111/cch.12114
190. Mohr D, Burns M, Schueller S, Clarke G, Klinkman M. Behavioral Intervention Technologies: Evidence review and recommendations for future research in mental health. Gen Hosp Psychiatry. 2013;35(4):332–338. doi: 10.1016/j.genhosppsych.2013.03.008
191. Canter K, Deatrick J, Hilgart M et al. eSCCIP: A psychosocial ehealth intervention for parents of children with cancer. Clin Pract Pediatr Psychol. 2019;7(1):44–56. doi: 10.1037/cpp0000264
192. Andersson G, Cuijpers P. Internet-Based and Other Computerized Psychological Treatments for Adult Depression: A Meta-Analysis. Cogn Behav Ther. 2009;38(4):196–205. doi: 10.1080/16506070903318960
193. Cuijpers P, Marks I, van Straten A, Cavanagh K, Gega L, Andersson G. Computer-Aided Psychotherapy for Anxiety Disorders: A Meta-Analytic Review. Cogn Behav Ther. 2009;38(2):66–82. doi: 10.1080/16506070802694776
194. Mohr D, Likosky W, Bertagnolli A et al. Telephone-administered cognitive–behavioral therapy for the treatment of depressive symptoms in multiple sclerosis. J Consult Clin Psychol. 2000;68(2):356–361. doi: 10.1037/0022-006x.68.2.356
195. Mohr D, Hart S, Julian L et al. Telephone-Administered Psychotherapy for Depression. Arch Gen Psychiatry. 2005;62(9):1007. doi: 10.1001/archpsyc.62.9.1007
196. Swallow V, Carolan I, Smith T et al. A novel Interactive Health Communication Application (IHCA) for parents of children with long-term conditions: Development, implementation and feasibility assessment. Informatics for Health and Social Care. 2014;41(1):20–46. doi: 10.3109/17538157.2014.948174
197. Wysocki T, Pierce J, Caldwell C et al. A Web-Based Coping Intervention by and for Parents of Very Young Children With Type 1 Diabetes: User-Centered Design. JMIR Diabetes. 2018;3(4):e16. doi: 10.2196/diabetes.9926
198. Otado J, Kwagyan J, Edwards D, Ukaegbu A, Rockcliffe F, Osafo N. Culturally Competent Strategies for Recruitment and Retention of African American Populations into Clinical Trials. Clin Transl Sci. 2015;8(5):460–466. doi: 10.1111/cts.12285
199. Horowitz C, Brenner B, Lachapelle S, Amara D, Arniella G. Effective Recruitment of Minority Populations Through Community-Led Strategies. Am J Prev Med. 2009;37(6):S195–S200. doi: 10.1016/j.amepre.2009.08.006
200. Yancey A, Ortega A, Kumanyika S. Effective Recruitment and Retention of Minority Research Participants. Annu Rev Public Health. 2006;27(1):1–28. doi: 10.1146/annurev.publhealth.27.021405.102113
201. Ejiogu N, Norbeck J, Mason M, Cromwell B, Zonderman A, Evans M. Recruitment and Retention Strategies for Minority or Poor Clinical Research Participants: Lessons From the Healthy Aging in Neighborhoods of Diversity Across the Life Span Study. Gerontologist. 2011;51(Supplement 1):S33–S45. doi: 10.1093/geront/gnr027
202. Macinko J, Starfield B, Shi L. The Contribution of Primary Care Systems to Health Outcomes within Organization for Economic Cooperation and Development (OECD) Countries, 1970-1998. Health Serv Res. 2003;38(3):831–865. doi: 10.1111/1475-6773.00149
203. Starfield B, Shi L. Policy relevant determinants of health: an international perspective. Health Policy. 2002;60(3):201–218. doi: 10.1016/s0168-8510(01)00208-1
204. Levesque J, Harris M, Russell G. Patient-centred access to health care: conceptualising access at the interface of health systems and populations. Int J Equity Health. 2013;12(1):18. doi: 10.1186/1475-9276-12-18