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Neurodevelopmental and psychosocial interventions for individuals with congenital heart disease: A research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative

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Abstract

In 2018, the Neurodevelopmental and Psychosocial Interventions Working Group of the Cardiac Neurodevelopmental Outcome Collaborative convened through support from an R13 grant from the National Heart, Lung, and Blood Institute to survey the state of neurodevelopmental and psychosocial intervention research in congenital heart disease and to propose a slate of critical questions and investigations required to improve outcomes for this growing population of survivors and their families. Prior research, although limited, suggests that individualized developmental care interventions delivered early in life are beneficial for improving a range of outcomes including feeding, motor and cognitive development, and physiological regulation.

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Conflicts of Interest
None

Interventions to address self-regulatory, cognitive, and social-emotional challenges have shown promise in other medical populations yet their applicability and effectiveness for use in individuals with congenital heart disease have not been examined. To move this field of research forward, we must strive to better understand the impact of neurodevelopmental and psychosocial intervention within the congenital heart disease population including adapting existing interventions for individuals with congenital heart disease. We must examine the ways in which dedicated cardiac neurodevelopmental follow-up programs bolster resilience and support children and families through the myriad transitions inherent to the experience of living with congenital heart disease. And we must ensure that interventions are person-/family-centered, inclusive of individuals from diverse cultural backgrounds as well as those with genetic/medical comorbidities, and proactive in their efforts to include individuals who are at highest risk but who may be traditionally less likely to participate in intervention trials.

Keywords

congenital heart disease; developmental care; intervention; neurodevelopmental outcomes; psychosocial outcomes

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 (NHLBI) of the National Institutes of Health (NIH) 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]

Now that individuals with congenital heart disease (CHD) are living longer, it is clear that the neurodevelopmental and psychosocial challenges they face are among the strongest correlates and predictors of quality of life across the lifespan.[9] This risk is especially great among those diagnosed with critical CHD requiring surgery within the first year of life. Elevated risk of early feeding, motor, and self-regulatory difficulties during infancy give way to later-emerging deficits in attention, executive function, visual-spatial processing, and social cognitive capacities during childhood and adolescence, which in turn undermine the development of adaptive skills necessary to successfully manage the transition to adulthood and subsequent independence.[10] Despite increasing recognition of these challenges,[10,11] little attention has been given to the design and implementation of CHD-specific neurodevelopmental and psychosocial interventions.[12,13]

At this time, our knowledge of the risks for adverse neurodevelopmental and psychosocial outcomes in individuals with CHD dramatically outstrips our knowledge of how to mitigate those risks – an imbalance that has become untenable as patients, families, care providers,

and other stakeholders look increasingly for guidance regarding how best to optimize individual potential and maximize quality of life for each child and family affected by CHD.

Further, since both biological and social determinants of health are critical when optimizing wellness, interventions must be designed to have socio-ecologic validity and the capacity to reach all individuals, especially those facing greater psychosocial challenges and health disparities.[14]

The *Neurodevelopmental and Psychosocial Interventions Working Group* of the Cardiac Neurodevelopmental Outcome Collaborative included content area experts in psychology and neuropsychology, cardiology, feeding and speech/language pathology, health disparities, and family support including a patient and a parent stakeholder (Table 1). Working Group participants included members from the United States and Europe who convened in 2018 to address the following goals: 1) Describe the state of neurodevelopmental and psychosocial intervention research in CHD and 2) propose an interventions research agenda aimed at optimizing the neurodevelopmental and psychosocial potential of individuals affected by CHD. 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 two-day meeting of multidisciplinary, multinational experts and patient/caregiver stakeholders in Kansas City, MO.

To achieve its goals, the Working Group developed five critical questions to guide the development of an intervention research agenda for CHD for the next decade (Table 2). Each critical question focused on interventions that are inclusive for individuals of all backgrounds including those who traditionally face health disparities and those with genetic diagnoses and other medical comorbidities. The research agenda included interventions that have both randomized controlled trial and quality improvement designs, occur across settings (e.g., home, school, hospital, e-Health/telemedicine, camp), are tailored to the challenges associated with CHD, are preventative, include cost-effectiveness analysis, and are focused on optimization for both individual and group differences.

Critical Question 1: How do we adapt effective interventions in other medical populations that address known risk factors in CHD?

Existing Knowledge

There is a sizeable body of evidence supporting the efficacy of interventions which address known neurodevelopmental and psychosocial risk factors in other high-risk populations such as children born preterm and those diagnosed with developmental disabilities. Moreover, theoretical frameworks exist for adapting interventions for use in individuals with various medical conditions.[15,16]

Individualized developmental care programs show particular promise for promoting positive neurodevelopmental outcomes among medically at-risk children when implemented in the newborn period. Individualized developmental care is a model of care that minimizes the mismatch between infant neurobiological needs and the often toxic hospital environment.

The Newborn Individualized Developmental Care and Assessment Program[17,18], is the only evidence based developmental care program and is well-validated in the preterm infant population where it has been shown to decrease length of hospital stay and improve physiological functioning, long-term neurodevelopment, parent confidence, and patient and family satisfaction among infants born preterm.[18–21] Other programs, such as Trauma Informed Care and Family Centered Care, incorporate an understanding of trauma and need to recognize the central importance of family into routine care and treatment of illness.[22–24] Specific aspects of these programs, such as skin-to-skin contact, interdisciplinary developmental care rounds, cue-based care, family support, and education for providers have been found to be developmentally supportive of children and families affected by CHD and contribute to improved neurodevelopmental outcomes.[17,25–31] The use of breast milk and breast feeding can also support infant growth and oral feeding, as well as promote bonding with family and improve cognitive development, [32] and social-emotional growth over time.[33,34]

In addition to inpatient supports, early intervention has demonstrated positive effects on the developmental achievements of children with or at-risk for developmental disability.[35–37] Families from high-risk populations who received prenatal and infancy home visits by nurses showed improved cognitive, academic, behavioral, and sociodemographic outcomes for their children.[38]

Interventions targeting areas of deficit commonly observed among individuals with CHD have been developed and well-established with non-CHD populations. As an example, practice guidelines for behavioral and psychotropic interventions for individuals diagnosed with attention-deficit/hyperactivity disorder are established for other populations yet there has been limited research involving individuals with CHD who have special considerations due to cardiovascular effects of common medications[39,40]. Similarly, the efficacy of behavioral and psychotropic interventions for individuals with mood[41] and anxiety disorders,[42,43] including procedural anxiety[44] and coping with medical illness,[45] have been established but these interventions have been understudied among individuals with CHD. A 2013 Cochrane review identified no randomized controlled trials demonstrating the efficacy of cognitive-behavioral interventions for depression in adolescents or adults with CHD,[46] and a more recent review continued to report limited efficacy among these populations.[13]

Among healthy school-age children and adolescents, as well as those with various medical conditions, there is a strong interest in addressing neurocognitive deficits (e.g., in executive function, attention), using, for example, computerized interventions such as Cogmed; however, data on the effectiveness of these programs have been mixed.[47,48] Therapeutic camp programs have been shown to improve mood, self-concept, empathy, quality of life, and emotional well-being for children with cancer and their families.[49] Adolescents with chronic illness also benefit, in terms of adjustment and well-being, from peer-based support programs, including programs that are school-based and disease-specific, as well as those that are community-based.[50] Emerging evidence exists for telemedicine and e-Health interventions in improving outcomes for adolescents with chronic medical and psychiatric conditions, including PTSD.[51]

Significant Gaps in Knowledge

Despite decades of research evaluating neurodevelopmental and psychosocial interventions for other medical populations, the safety, feasibility, acceptability, accessibility, efficacy, and effectiveness of these interventions for use in individuals with CHD are largely unknown. Adapting interventions from other populations will require an understanding of the unique characteristics and challenges inherent in CHD, and their relevance to the particular intervention considered. In addition, while continued surveillance and consultation is recommended for children with complex CHD, it is unclear how many are receiving early intervention following discharge and how this impacts long-term development.

Outside the hospital setting, and particularly among school-age children, adolescents, and young adults with CHD, we remain largely uninformed regarding the long-term effectiveness of neurocognitive interventions (e.g., Cogmed), as well as their potential impact on academic and social domains; the effectiveness of specific educational and peer mentorship interventions; the impact of health inequities and barriers that may prevent individuals and families from accessing interventions; how best to engage telemedicine and e-Health, social media, and other technology tools to broaden the reach of interventions beyond the clinic setting; and the short- and longer-range economic implications and cost-effectiveness of intervening to mitigate the host of risks associated with CHD.

Investigations Needed

1. ***Investigate the safety and feasibility of individualized developmental care interventions delivered during a cardiac hospitalization.*** Safety and feasibility studies of individualized developmental care interventions in the cardiac intensive care unit are needed to adapt evidence-based programs to the unique needs of infants with CHD and their families. Smaller-scale quality improvement studies should lead to larger-scale, multi-center, randomized controlled trials to assess child neurodevelopmental and physiologic outcomes in the newborn period, as well as in early infancy and over the course of the lifespan. Such research could potentially highlight the implications of early-life intervention on later markers of health and well-being, along with improved family outcomes, decreased parent stress, and reduced healthcare utilization and economic burdens.

2. ***Examine best practices for promoting handling and moving of infants and young children during cardiac hospitalization.*** Concept and safety/feasibility studies are necessary to examine safe practices for handling/moving infants in the inpatient acute cardiac care setting, providing skin-to-skin holding, and increasing parent involvement in care and handling. This should be followed thereafter by quality improvement projects geared at increasing parent/caregiver comfort regarding the full range of developmental care interventions while hospitalized. These endeavors would be strengthened by a team-based, interdisciplinary approach that includes collaborative partnerships among nursing, cardiology/cardiac surgery, physical therapy, occupational therapy, child life and music therapy, nutrition, speech/language pathology, and psychology.

3. ***Study short and longer range outcomes associated with individualized developmental care interventions in acute cardiac inpatient setting.*** As mentioned, there are many positive outcomes associated with individualized developmental care intervention, including decreased length of hospitalization and improved feeding, among children born preterm.[19–21,35] In the case of infants with CHD, for which length of hospital stay is among the strongest risk factors for adverse outcomes,[52–54] reduced length of hospital stay would lower hospital costs and reduce exposures to potentially noxious elements in the acute inpatient environment (e.g., plasticizers,[55] loud sounds, bright lights, inadequate protection of sleep, inadequate attention to parent mental health, separation from family, stress reduction, and the use of non-pharmacologic comfort interventions[56]) that may contribute to worse outcomes for these children. Individualized developmental care interventions also advocate for staff support to reduce stress and burnout, which would positively affect the patient and family. Interventions to support growth and weight gain, use of human milk, early breast feeding, and decreasing time to full oral feeding in patients with CHD[57–61] is of utmost importance as oral feeding ranks the greatest stressor for caregivers following cardiac surgery and often lengthens hospital stay.[62,63] In addition there is no current gold standard program for infant feeding in cardiology, but this should be explored. Programs that monitor development over time and provide intervention beyond infancy such as early intervention and early supports in the school system would likely reduce concerns seen in adolescence and adulthood.

4. ***Conduct translational research studying empirically-supported psychosocial and neurocognitive interventions developed for other populations in individuals with CHD.*** Building on existing knowledge of interventions that work in other populations, translational studies are needed to effectively adapt interventions for use among children with CHD. Psychosocial interventions targeting anxiety, mood concerns, and the effectiveness of cognitive-behavioral therapy in addressing these issues will be particularly important given their high prevalence among individuals with CHD. Efficacious interventions developed for individuals with other chronic illnesses, such as cancer and diabetes, that address comorbidities such as pain, adherence to medical regimens, family functioning, transition from pediatric to adult healthcare, and traumatic stress could be adapted to benefit individuals with CHD.[64] Neurocognitive interventions that address attention, executive function, and visual-spatial deficits are also necessary, and should investigate a range of delivery modalities including computerized and in-person formats.

5. ***Investigate new modalities for delivering neurodevelopmental and psychosocial interventions within the CHD population.*** Concept, pilot, and quality improvement studies can explore new modalities of intervention delivery, followed thereafter by larger scale, multi-center implementation studies. For example, telemedicine for post-surgical developmental follow-up may promote earlier identification and treatment of neurodevelopmental and psychosocial

issues.[65,66] Randomized controlled trials of these interventions in hospital settings or via telemedicine[67] may establish effectiveness for patients with CHD, reach a larger population, and provide preventative intervention.

Critical Question 2: What is the impact of neurodevelopmental and psychosocial interventions in individuals with congenital heart disease (CHD)?

Existing Knowledge

Preliminary interventions in infants with CHD show improvement in infant oral feeding,[68,69] physiological regulation,[70] early cognitive development,[70] family functioning,[70] and reduced length of hospital stay following surgery.[68,69] Moreover, findings from the Congenital Heart Disease Intervention Project, a series of controlled trials aimed at improving psychosocial and neurodevelopmental outcomes among young children with severe CHD, support the use of parent-oriented psychoeducation for improving infant mental, social, and emotional development at 6 months of age and gains in family functioning and fewer days of missed school among 4–6-year-old children.[71–75] However, a similarly designed randomized controlled trial utilizing both parent- and child-oriented psychoeducation reported only small, non-significant improvements in child psychosocial adjustment relative to standard care.[76] For children and adolescents with CHD, computerized interventions are being studied to examine their impact on executive function and social skills.[77] Aerobic exercise has been associated with self or proxy-reported improvements in cognitive functioning, social outcomes, and health-related quality of life. Recommendations for reducing child anxiety related to invasive cardiac procedures[78] have been documented but not clinically tested. Single center interventions including psychotropic medication,[10] access to a psychologist in clinic,[11] mindfulness training,[12] and increased physical activity[13–15] have demonstrated reduced symptoms of depression, anxiety, stress, and improved quality of life for adolescents and adults with CHD.

Significant Gaps in Knowledge

Most prior neurodevelopmental/psychosocial intervention studies are single-centered, cross-sectional, and have not made use of randomized controlled designs which remain the gold standard for clinical trials. Many of these investigations had limited statistical power to detect a meaningful effect, and outcome measurements varied greatly between studies. Further, efficacy of interventions in adolescence, to date, has been weak[13] and more trials are needed. Finally, many of these intervention studies exclude individuals with CHD with comorbidities such as genetic syndromes, which may substantially impact intervention design, administration, interpretation, and generalizability of findings.

Investigations Needed

1. ***Operationalize clinically meaningful intervention outcomes across development.*** It is critical to carefully consider outcome measures based on the age and functional status of the child, and any behavioral and emotional constructions of relevance to the intervention. Initially, studies focused on

global neurodevelopmental skills such as overall intelligence quotient scores, but as more has been learned about risk, outcomes are being tailored to aspects of neurodevelopment that are more often impaired among a CHD population such as executive function and visual-spatial processing. Standardized measurement protocols to assess neurodevelopmental outcomes, as well as key moderators of intervention efficacy and effectiveness (e.g., SES, language), must be identified, and may include formal assessment, structured observational measures, caregiver-/self-report questionnaires, and measures of neurobiological change (i.e., structural or functional variations on neuroimaging). It is important to have consistency across sites to reduce bias that can come from single-center reporting and promote generalizability of findings. The Cardiac Neurodevelopmental Outcome Collaborative has made recommendations[4,5] for a standardized assessment battery from infancy through teen years, which will help to guide future intervention research when selecting outcome measures to assess the impact of interventions on the neurodevelopment of individuals with CHD.[4,5] Large-scale, multi-center studies, which will be feasible within the context of the Cardiac Neurodevelopmental Outcome Collaborative data registry, are necessary to allow for adequate clinical stratification and inclusion of potential comorbidities as well as more diverse sociodemographic variables.

2. ***Conduct prospective randomized controlled trials with longer-term follow-up to investigate efficacy and effectiveness beyond the snapshot of a pre-post intervention.*** Studies with sequential post-intervention visits, at predetermined time-intervals, would provide evidence of cost-effectiveness and potential generalization of treatment effects in the long-term. The number and timing of follow-up should take developmental period into account with more immediate follow-up during early development and longer-term follow-up of more complex neurodevelopmental skills into adolescence and beyond. Further, efficacy trials (does an intervention work in an ideal setting) should be developed with a mind to effectiveness (does an intervention work in a real-world setting and are they feasible given limitations such as cost).

3. ***Partner with key stakeholders to define "clinically meaningful" outcomes.*** Determining what constitutes a clinically meaningful change post-intervention involves more than statistical significance. Indeed, the threshold for clinically meaningful changes pre- versus post-intervention should be interpreted in light of both individual and population-based changes in CHD.[79] In all interventions research, it will be important to enlist the input of patients, families, and other stakeholders to ensure accurate understanding of the real-world relevance of selected outcome measures and to consider such an understanding alongside quantitative indicators of change (e.g., effect sizes quantification and use of reliable change index estimates, along with statistical significance). For instance, a 2-point standard score drop on a measure of externalizing behavior may be a statistically significant change, but is unlikely to be a noticeable change in real-world behavior.

Critical Question 3: How are cardiac neurodevelopmental programs currently utilized, in what ways do these coordinated programs impact outcomes, and what are the best program practices?

Existing Knowledge

Early intervention programs for high-risk populations, such as preterm and/or very low birthweight infants, are well-established and associated with improved neurodevelopmental[80] and psychosocial functioning[81] and have demonstrated the positive impact of inpatient neurodevelopmental care[82,83] and outpatient neurodevelopmental follow-up.[84] Networks of newborn follow-up programs serve as data registries providing program benchmarks, initiating multi-site quality improvement projects to improve standard of care, and allow for the development of best practice guidelines.[85] Indeed, the importance of standardized follow-up programs for former medically-fragile neonates is so strongly recognized that it is a requirement for accreditation for graduate medical education in neonatal-perinatal medicine by the Accreditation Council for Graduate Medical Education.[86]

The National Pediatric Cardiology Quality Improvement Collaborative [87] and the Cardiac Neurodevelopmental Outcome Collaborative have created data registries to track neurodevelopmental outcomes for children with CHD. The creation of data registries and benchmarking, especially when approached through the lens of quality improvement science, will inform the development, implementation, and dissemination of best practice guidelines. For other complex pediatric conditions including cancer [88] and cystic fibrosis,[89] the best practices of care have been driven by data derived from patient registries.

Significant Gaps in Knowledge

While much is known about the neurodevelopmental and psychosocial benefits of developmental follow-up programs in neonatology, there are no published studies of the impact of participation in cardiac neurodevelopmental follow-up programs. These programs provide what are thought to be critical intervention supports and services, and yet empirical data are currently lacking.

Investigations Needed

1. **Conduct feasibility, acceptability, and accessibility studies to examine processes (e.g., screening, monitoring procedures) and components (e.g., types of services) that result in the most beneficial outcomes.** Outcome measurements, standardized across programs, should focus on assessing domains that are most clinically meaningful to individuals with CHD and their families (e.g., quality of life, successful transition to independence). Studies may also include measurement of program access, utilization, cost-effectiveness, and socio-demographic variation as well as patient experience and pathways to care. Determining methods to reduce barriers to accessing cardiac neurodevelopmental programs would boost attendance, a key aspect of universal protection/prevention screening and assessment programs. It will be particularly important to examine

availability of trained personnel, time to appointment date or waitlist, physical space, cost, and insurance coverage.

2. ***Examine whether centers that have coordinated cardiac neurodevelopmental programs actually have improved neurodevelopmental and psychosocial outcomes for individuals with CHD.*** Study designs should include pre-post program implementation data collection, and should compare outcomes across time points as well as between centers with and without cardiac neurodevelopmental programs on variables such as percentage of children entering school with appropriate educational supports, patient/family satisfaction, quality of life, and performance on formal measures of neurodevelopmental and psychosocial functioning. Establishing model programs as the standard-of-care across medical centers will require clear evidence of effectiveness for a variety of stakeholders, including patients and families, advocacy groups, hospital administration, and insurance carriers.

3. ***Develop efficient ways to screen individuals seen in cardiac neurodevelopmental programs and tailor to different levels of intervention.*** Insofar as timely and appropriate identification and stratification of risk facilitates efficient access to limited assessment and treatment resources, it will be important to design and test procedures for screening individuals with CHD to ensure that resources are allocated appropriately. In line with screening models proposed by Kazak and colleagues,[90] for pediatric psychology, and Hardy et al.,[91] for pediatric neuropsychology, large-scale, multi-site studies which evaluate the appropriateness of tiered screening procedures implemented within primary care/cardiology clinic settings would identify individuals most in need of neurodevelopmental and/or psychosocial support. As these models suggest, the majority of patients may succeed with only periodic surveillance and recommendations while the minority will require more intensive interventions. Developing a way to screen patients into these tiered interventions should result in more efficient care and could result in resource savings.

Critical Question 4: How do we foster the development of resilience in individuals with CHD?

Existing Knowledge

The concept of resilience, defined as “a dynamic process wherein individuals display positive adaptation despite experiences of significant adversity or trauma,”[92] is perhaps best understood as a capacity that develops over time, rather than as an inherent personality trait.[93] Resilience, and other wellness-promoting concepts such as posttraumatic growth and grit, are positively associated with better health outcomes[94] and decreased stress responses[95] within the general population, and improved psychosocial functioning and self-management in individuals with chronic illness.[96–98] For example, among young, highly stressed children in foster care, therapeutic interventions have been shown to promote resilience by mitigating the effects of early adversity on hypothalamic-pituitary-adrenal axis activity and promoting the development of adaptive caregiver attachment relationships.[99] In

addition, adolescents with greater knowledge of their own medical history and associated complications, higher resilience, and more positive family dynamics have been found to better adhere to health-promoting behaviors such as following exercise and nutrition recommendations and reporting more adaptive stress management strategies.[100] Resilience in individuals with CHD is also related to a lower level of a depressive symptoms[101] and is influenced by parenting factors such as emotional warmth, rejection, punishment, control, and overprotection in adolescence.[102]

Other than a feasibility study[103] and some evidence to support the use of a group-based intervention to improve aspects of resilience in adolescents with CHD, [42] studies examining resilience in individuals with CHD are limited. Numerous studies report that participation in disease-specific camp programs positively influences perceived health, interpersonal relationships, and self-esteem,[49,104–108] which are components of resilience, but resilience has not yet been specifically measured.

Significant Gaps in Knowledge

Literature on the experiences impacting resilience, interventions to bolster resilience, and measurement is limited in the CHD population. For both children and adolescents, there is evidence to suggest that exercise/physical activity interventions promote improved cardiovascular health and enhanced psychosocial functioning and quality of life individuals with CHD;[109–112] although findings are somewhat mixed[113] and the impact of physical activity interventions on resilience remains to be examined. Even with more than 35 years of physical activity promotion and exercise training in patients with CHD,[114,115] research into optimal training methods and resilience as an outcome of physical exercise programs is lacking.

Investigations Needed

1. ***Adopt a conceptual framework for designing and conducting resilience-promoting intervention studies that appropriately captures the nature and complexity of resilience.*** A model from the National Scientific Council on the Developing Child recommends that interventions designed to facilitate resilience should include 1) ways to improve the caregiver-child relationship, 2) methods for building self-efficacy and perceived control, 3) strengthening adaptive and self-regulatory functioning, and 4) incorporating faith, hope, and cultural traditions.[95] Moreover, resilience interventions should encompass the entire lifespan and should begin with monitoring, supporting, and promoting the development of adaptive coping strategies for the family, if possible, before the child with CHD is even born, as further discussed below.

2. ***Capitalize on early identification to begin bolstering caregiver/family resilience prior to delivery.*** At prenatal cardiac diagnosis, interventions designed to shape the communication provided to the family, with a particular focus on the developing parent-infant relationship, education regarding infant neurodevelopment, and maternal and paternal self-care would support family well-being. By focusing on optimization and resilience, the emphasis of the

prenatal visit may shift to infant neuroprotection and promotion of optimistic parent perceptions of their child, potentially reducing parental stress during the pregnancy. This focus on family well-being before the child is born could result in improved long-term outcomes for the child with CHD.[116]

3. ***Recognize individual and family-based differences in perception and experience in living with CHD.*** The development of valid, CHD-specific tools to measure aspects of resilience will be important for assessing each child/family's unique experiences with CHD, including systemic and cultural factors, and family stress and available support. Adding measurement of resilience to a standardized battery could help to better understand how different experiences bolster resilience and identify targets for future intervention.

4. ***Identify interventions for promoting resilience during childhood and adolescence.*** Studies are needed to directly evaluate the potential benefit of physical activity interventions on resilience among individuals with CHD. Additionally, specifically measuring resilience as part of a CHD camp program could help to better understand positive outcomes from this experience. Other potential interventions may include developing a mentoring program for individuals with CHD or qualitative research with focus groups of adults with CHD to examine individual factors associated with resilience. Studies similar to those in adults with cancer, which utilize stress management to improve resilience,[117] would likely benefit adults with CHD as well and should be investigated.

5. ***Include resilience as a primary outcome in CHD surgical trials.*** The effects of decreased stress[95] should be looked at with respect to surgical outcomes, in addition to ICU length of stay, post-operative complications, and other aspects of health and recovery. Furthermore, child, family, and cultural markers of resilience should be carefully examined in clinical and surgical trials as potential moderators of outcomes.

Critical Question 5: How do we develop systematic and effective approaches to optimize developmental and medical transitions for individuals with CHD and their families?

Existing Knowledge

The experience of living with CHD includes numerous transition points with possible vulnerability and potential for intervention. Some of the most salient transitions include: 1) acute inpatient care to stepdown unit care; 2) tube to oral feeding; 3) inpatient to outpatient settings; 4) surgical center to local medical care; 5) early intervention to the school system; 6) childhood to adolescence; 7) adolescence to young adulthood; 8) pediatric to adult CHD care; and, 9) for caregivers, from being primarily an observer of their child's medical care to being the primary provider of daily medical surveillance and care and eventually becoming less active participants as their child moves toward independence.

The parents of medically-complex infants with CHD can experience high levels of stress, posttraumatic stress, anxiety, and depression,[118] which may negatively impact their ability to parent in ways most supportive of the high-risk child. The need to access services from multiple hospital and community systems makes communication and coordination among providers and caregivers critical. However, clear and understandable communication is often lacking.[119] Discharge instructions, for example, often include difficult medical terminology and can be confusing for families. School systems that are unfamiliar with the needs of children with CHD are unlikely to provide appropriate supports and services, increasing the risk for academic underachievement and discouragement. School-liaison programs, which serve as a bridge between clinic and school, and are considered standard-of-care in pediatric cancer,[120] are effective in promoting access to services and associated with increased parent satisfaction and parent beliefs that their child is meeting his/her academic potential.[121,122]

Specific to CHD, deficits in executive function, which are highly prevalent,[123,124] are likely to become more problematic during the transition to adolescence, undermining the development of independence and adaptive skills that may further compromise the transition to adulthood. Indeed, 40–60% of CHD patients experience a lapse in their care, particularly during the transition to adult medical care, and those who experience a lapse are three times more likely to require urgent cardiac intervention.[125,126] Lack of knowledge, self-management, and self-advocacy skills has also been documented among heart transplant patients.[127] However, participation during adolescence in a nursing-led educational intervention designed to prepare transplant patients for transition to adult care resulted in better maintenance of medical follow-up and increased CHD knowledge and self-management skills,[128] so an initial transition intervention has shown some promise.

Significant Gaps in Knowledge

Intervention research aimed at promoting optimal adaptation across the full range of transitions inherent in CHD is limited; and, existing research focuses almost exclusively on the transition from pediatric to adult care.[128] Interventions are still needed to aid parents in the transition from inpatient to outpatient care; to increase the effectiveness of patient/family CHD educational tools; and, to support successful transitions from hospital to community-based CHD providers and schools. Moreover, among adolescents with CHD, it remains unclear how best to promote functional independence, adaptive skills, and self-awareness/knowledge of one’s medical condition.

Addressing these critical questions should result in a more accurate understanding of the range of transitions inherent to CHD, which would in turn facilitate the generation of smoother and more standardized procedures and practice guidelines for promoting optimal development across times of transition. With enhanced communication and coordination across providers and settings, fewer patients would be missed or lost to follow-up, child and family support needs would be more readily identified and would trigger appropriate referrals and access to therapeutic services, and barriers to accessing services would be recognized and addressed to reduce healthcare inequities.

Investigations Needed

1. ***Comprehensively characterize the full range of transitions inherent to living with CHD.*** Large-scale, population-based parent and/or self-report surveys with both qualitative and quantitative analytic techniques are needed to characterize the full range of transitions experienced by individuals with CHD, as well as key challenges to adaptive/functional independence across the lifespan.

2. ***Utilize quality improvement science to improve strategies for assisting families in navigating CHD-specific challenges and transitions in the medical system.*** Quality improvement-oriented intervention studies are also indicated to evaluate and improve the effectiveness of strategies for preparing parents following prenatal CHD diagnosis. Additional interventions may include providing developmental summaries, modifying discharge information, and increasing the frequency of post-discharge follow-up – and should deliberately consider how technology such as phone-based apps and teleconferencing may be used to support families following discharge.

Conclusions

Advancements in neurodevelopmental and psychosocial interventions for individuals with CHD have the potential to radically reshape prevailing paradigms related to patient-care and expectations regarding short- and long-range outcomes from infancy to adulthood. Establishment of a coordinated cardiac neurodevelopmental program data registry,[2] the use standardized measurement of key neurodevelopmental and psychosocial outcomes across programs, and administrative support for follow-up and data collection is critical. From the perspective of research and quality improvement science, results of well-designed intervention trials, including trials within a well-designed data registry[129,130] would directly inform practice guidelines and improve long term outcomes for children and families managing CHD. Finally, to promote resilience and optimization for all individuals with CHD, it is crucial for outcomes to be individualized, to avoid exclusion based on genetics and other medical comorbidities, to address cultural differences and values that may impact the development of resilience, and to include outreach efforts to study interventions for those less likely to participate. Improvements in neurodevelopment and parent support will, in turn, result in a healthier, happier, more independent, more productive, and generally more resilient population, requiring fewer federal and state governmental services and well-positioned to contribute to society to the fullest extent possible.

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Table 1.

Neurodevelopmental and Psychosocial Interventions Working Group Participants

WG Participants	Discipline/Role	Institution/Organization	Country
Adam R. Cassidy *	Pediatric Neuropsychologist	Boston Children's Hospital; Harvard Medical School	USA
Jennifer Butcher *	Pediatric Psychologist	C.S. Mott Children's Hospital; Michigan Medicine	USA
Samantha Butler	Developmental and Clinical Psychologist	Boston Children's Hospital; Harvard Medical School	USA
Jennie Briend	Parent Stakeholder	Sisters by Heart	USA
Johanna Calderon	Pediatric Neuropsychologist	Boston Children's Hospital; Harvard Medical School	USA
Frank Casey	Pediatric Cardiologist	Paediatric Cardiology Belfast Trust; Royal Belfast Hospital For Sick Children	Ireland
Lori E. Crosby **	Pediatric Psychologist	Cincinnati Children's Hospital Medical Center; University of Cincinnati College of Medicine	USA
Jennifer Fogel	Speech-Language Pathologist	Advocate Children's Hospital	USA
Naomi Gauthier	Pediatric Cardiologist	Boston Children's Hospital; Harvard Medical School	USA
Carol Raimondi	Patient Stakeholder	Conquering CHD	USA

Note. WG = working group.

*Working Group Co-Lead

**Health Disparities Expert

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Table 2.

Interventions: Critical Questions, Significant Gaps in Knowledge, and Investigations Needed

Critical Questions	Significant Gaps in Knowledge	Investigations Needed
CQ1. How do we adapt effective interventions that address known risk factors in CHD?	• Despite considerable research evaluating the effectiveness of a range of neurodevelopmental and psychosocial interventions in other medical populations, the safety, feasibility, acceptability, and effectiveness of these interventions is limited among individuals with CHD • Very little is known about the effectiveness of neurocognitive interventions, particularly among school-age children, adolescents, and young adults with CHD	• Investigate the safety and feasibility of individualized developmental care interventions in the CICU • Examine best practices for promoting handling and moving of infants while inpatient. • Study short and longer range outcomes associated with individualized developmental care interventions in the CICU • Conduct translational research studying empirically-supported psychosocial and neurocognitive interventions developed for other populations in individuals with CHD • Investigate new modalities for delivering neurodevelopmental and psychosocial interventions within the CHD population
CQ2. What is the impact of neurodevelopmental and psychosocial interventions in individuals with CHD?	• Most prior neurodevelopmental and psychosocial intervention studies in CHD are limited to single-center, concurrent, observational studies • Many CHD neurodevelopmental intervention studies exclude individuals with genetic conditions	• Operationalize clinically meaningful intervention outcomes for each developmental stage • Conduct prospective randomized controlled trials with longer-term follow-up to investigate efficacy and effectiveness beyond the typical snapshot of a pre-post intervention • Partner with key stakeholders to define “clinically meaningful” outcomes
CQ3. How are CND programs currently utilized, in what ways do coordinated CND programs impact outcomes, and what are the best program practices?	•Limited number of published studies examining the benefits of developmental follow-up programs among individuals with CHD	• Conduct feasibility, acceptability, and accessibility studies to examine processes (e.g., screening, monitoring procedures) and components (e.g., types of services) that result in the most beneficial CND programs • Examine whether centers that have coordinated CND programs show improved neurodevelopmental and psychosocial outcomes for individuals with CHD • Develop efficient ways of triaging patients seen in CND programs to different levels of intervention based on individual needs
CQ4. How do we foster the development of resilience in individuals with CHD?	•Experiences driving the development of resilience among individuals with CHD and interventions to bolster the development of resilience in the CHD population have not been adequately examined	•Adopt a conceptual framework for designing and conducting resilience-promoting intervention studies that appropriately captures the nature and complexity of resilience •Capitalize on early identification to support caregiver/family resilience prior to delivery •Recognize individual and family-based differences in perception regarding the experience of living with CHD •Identify interventions for promoting resilience during childhood and adolescence •Include resilience as a primary outcome in CHD surgical trials
CQ5. How do we develop systematic and effective approaches to optimize developmental transitions and transitions in care for individuals with CHD and their families?	•The full range of transitions inherent to the experience of living with CHD has neither been adequately characterized nor have interventions to support effective transitions been examined •It remains unclear how best to promote functional independence, adaptive skills, and self-awareness among individuals with CHD	•Comprehensively characterize the full range of transitions inherent to living with CHD •Utilize quality improvement science to improve strategies to assist families in navigating CHDspecific challenges and transitions

Note. CHD = congenital heart disease, CQ = critical question, CND = cardiac neurodevelopmental.