

Optimising neurodevelopmental and psychosocial outcomes for survivors with CHD: a research agenda for the next decade

Original Article

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Abstract

Neurodevelopmental and psychosocial impairments negatively impact health-related quality of life for survivors with CHD and complicate the transition to independent adulthood. Risk for neurodevelopmental and psychosocial impairments is influenced by a complex interplay among genetic, foetal, surgical, perioperative, family, and social factors, requiring a multi-pronged approach to neuroprotection and intervention. To ensure future research can ultimately reduce the burden of CHD for individuals, families, and society, the most pressing issues in cardiac neurodevelopment requiring scientific investigation must be identified.

Through funding from an R13 Grant from the National Heart, Lung, and Blood Institute of the National Institutes of Health of the United States of America, the Cardiac Neurodevelopmental Outcome Collaborative convened a two-day meeting of international experts in cardiac neurodevelopmental and psychosocial research, clinical care, and health disparities, including patient and family stakeholders, to define the cardiac neurodevelopmental and psychosocial outcomes research agenda for the next decade. Seven multidisciplinary working groups were formed to address key domains crucial to the advancement of cardiac neurodevelopmental and psychosocial outcomes research: 1) Foetal Brain Development and Neuroprotection, 2) Surgical/Perioperative Neuroprotection and Neurodevelopment, 3) Characterization of Neurodevelopmental and Psychological Outcomes, 4) Neurodevelopmental and Psychosocial Intervention, 5) Parent Mental Health and Family Functioning, 6) Neurodevelopmental Education, Outreach and Advocacy, and 7) Health Disparities and Neurodevelopmental Outcomes. Working groups identified significant gaps in knowledge and critical questions that must be answered to further knowledge, policy, care, and outcomes. The development of a research agenda in cardiac neurodevelopmental and psychosocial outcomes is critical for informing collaborative initiatives and allocation of funding for research to scientific inquiries of highest value to key stakeholders.

The November 2020 issue of *Cardiology in the Young* contains the inaugural five manuscripts from the Cardiac Neurodevelopmental Outcome Collaborative¹⁻⁵, marking the beginning of the partnership between the Cardiac Neurodevelopmental Outcome Collaborative and *Cardiology in the Young*:

- Bradley Marino MD MPP MSCE MBA and colleagues described the development and organisation of the Cardiac Neurodevelopmental Outcome Collaborative and the infrastructure that facilitates collaboration across sites and with patients and caregivers, meaningful and innovative science, and the establishment and dissemination of best-practice guidelines.²
- Thomas A. Miller DO and colleagues characterised the variation in structure and personnel across cardiac neurodevelopmental follow-up programmes within member institutions of the Cardiac Neurodevelopmental Outcome Collaborative and highlighted the need for more resources dedicated to school-age, adolescent, and young adult patients.³
- Finally, Janice Ware, PhD and colleagues and Dawn Ilardi, PhD ABPP-CN and colleagues described strategies, considerations, and best practices for conducting the neurodevelopmental evaluation of individuals with CHD across the lifespan and provided specific recommendations for brief core assessment batteries and extended comprehensive assessment batteries, as defined by expert working groups comprised of multidisciplinary members of the Cardiac Neurodevelopmental Outcome Collaborative.^{4,5}

In this issue of *Cardiology in the Young*, we are very pleased to publish 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

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across seven domains of cardiac neurodevelopmental and psychosocial outcomes research:

- Jacqueline Sanz PhD ABPP-CN and colleagues present “Characterization of neurodevelopmental and psychological outcomes in CHD: a research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative”⁶
- Adam R. Cassidy PhD ABPP-CN and colleagues present “Neurodevelopmental and psychosocial interventions for individuals with CHD: a research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative”⁷
- Finally, Erica Sood PhD and colleagues present “Parent mental health and family functioning following diagnosis of CHD: a research agenda and recommendations from the Cardiac Neurodevelopmental Outcome Collaborative.”⁸

This current introductory article that you are now reading, as well as the eight articles listed above,¹⁻⁸ are all available at the *Cardiology in the Young* website on the Affiliate Collections page of The Cardiac Neurodevelopmental Outcome Collaborative [<https://www.cambridge.org/core/journals/cardiology-in-the-young/collections/affiliate-collections/cnoc-collection>].

Rationale for this initiative

Over the past several decades, the population of survivors with CHD has increased dramatically^{9,10}, and survival after surgery to repair or palliate the congenitally malformed heart has also dramatically improved.¹¹ Meanwhile, important neurodevelopmental and psychosocial impairments in these surviving patients have become evident.¹²⁻¹⁵ These morbidities negatively impact functional outcomes and health-related quality of life for children with CHD and complicate or delay the transition to independent adulthood.¹⁶⁻¹⁸ Risk for neurodevelopmental and psychosocial impairments is influenced by a complex interplay among genetic, foetal, surgical, perioperative, family, and social factors,¹⁹ requiring a multi-pronged approach to neuroprotection and intervention to mitigate impairments and maximise outcomes. The National Heart, Lung, and Blood Institute; American Heart Association; American Academy of Pediatrics; and Adult Congenital Heart Association have all prioritised research to determine the prevalence of and risks for neurodevelopmental and psychosocial impairments, their associated impact across the lifespan, and the development of interventions to mitigate these morbidities for individuals with CHD.^{12,20,21} However, research to date has identified few neuroprotective strategies or interventions that have meaningfully improved neurodevelopmental and psychosocial outcomes and health-related quality of life for this large population of patients.²²

To advance scientific knowledge and ensure that future research has a high potential for reducing the burden of CHD for individuals, families, and society, the most significant and pressing issues in cardiac neurodevelopment requiring investigation from research teams must be identified. The development of a research agenda in cardiac neurodevelopmental and psychosocial outcomes is critical for informing collaborative initiatives and the allocation of funding for research to those scientific inquiries of highest value to key stakeholder groups, including patients and families. The Cardiac Neurodevelopmental Outcome Collaborative, a multicentre, multinational, multidisciplinary organisation that aims to determine and implement best practices of neurodevelopmental and psychosocial services for individuals with paediatric and congenital

cardiac disease and includes a large network of researchers, clinicians, and patient/family stakeholders² is well-positioned to define the agenda for cardiac neurodevelopmental and psychosocial outcomes research for the next decade.

In 2018, the Cardiac Neurodevelopmental Outcome Collaborative partnered with the Ann & Robert H. Lurie Children’s Hospital of Chicago to obtain an R13 Grant from the National Heart, Lung, and Blood Institute of the National Institutes of Health of the United States of America. This R13 Grant brought together multidisciplinary experts in cardiac neurodevelopmental and psychosocial outcomes research and clinical care for a two-day meeting in Kansas City, Missouri, United States of America, to define the agenda for cardiac neurodevelopmental and psychosocial outcomes research for the next decade. Experts in health disparities and patient and family stakeholders were included to ensure that the resulting research agenda focused on promoting health equity and reducing disparities and reflected the priorities of those with lived experience. Seven multidisciplinary working groups were formed, and each was tasked with exploring one key domain crucial to the advancement of cardiac neurodevelopmental and psychosocial outcomes research:

- 1) Foetal Brain Development and Neuroprotection,
- 2) Surgical/Perioperative Neuroprotection and Neurodevelopment,
- 3) Characterisation of Neurodevelopmental and Psychological Outcomes,
- 4) Neurodevelopmental and Psychosocial Intervention,
- 5) Parent Mental Health and Family Functioning,
- 6) Neurodevelopmental Education, Outreach, and Advocacy, and
- 7) Health Disparities and Neurodevelopmental Outcomes.

During the two-day meeting and throughout the following year, working groups identified:

- significant gaps in knowledge;
- critical questions that must be answered to further knowledge, policy, care, and outcomes; and
- investigations needed to answer these critical questions.

The results of this collaborative initiative are being published as a series of papers in *Cardiology in the Young* to provide a roadmap for cardiac neurodevelopmental research teams.

This issue contains the first three papers from the third, fourth, and fifth domains listed above:

- *Characterisation of Neurodevelopmental and Psychological Outcomes*,⁶
- *Neurodevelopmental and Psychosocial Intervention*⁷, and
- *Parent Mental Health and Family Functioning*⁸.

Sanz and colleagues emphasise the need for additional research to provide an accurate characterisation of neurodevelopmental and psychological outcomes in individuals with CHD in order to optimise patient outcomes through targeted interventions, and they present critical research questions and innovative methodologies to address significant gaps in knowledge⁶. Cassidy and colleagues describe the limited research on neurodevelopmental and psychosocial interventions for patients with CHD and propose critical research questions that need to be answered in order to effectively intervene and improve outcomes⁷. Sood and colleagues emphasise the need to support parental mental health and family functioning following diagnosis of CHD in their children in order to promote psychosocial adaptation, and they outline critical research questions

that must be answered to further the care and outcomes of families impacted by CHD⁸. Future papers from this series will be published in *Cardiology in the Young* later in 2021 and will propose critical questions that need to be answered through rigorous research to optimise foetal brain development and surgical/perioperative neuroprotection; improve cardiac neurodevelopmental education, outreach, and advocacy; and eliminate health disparities in neurodevelopmental and psychosocial outcomes for individuals with congenital cardiac disease and their families. This series of papers is intended to guide future funding proposals and stakeholder-informed research in cardiac neurodevelopment, thereby advancing scientific knowledge, clinical care, and meaningful outcomes for this large and growing population of survivors with CHD.

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Conflicts of interest. None.

Ethical standards. The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guidelines on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008.

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