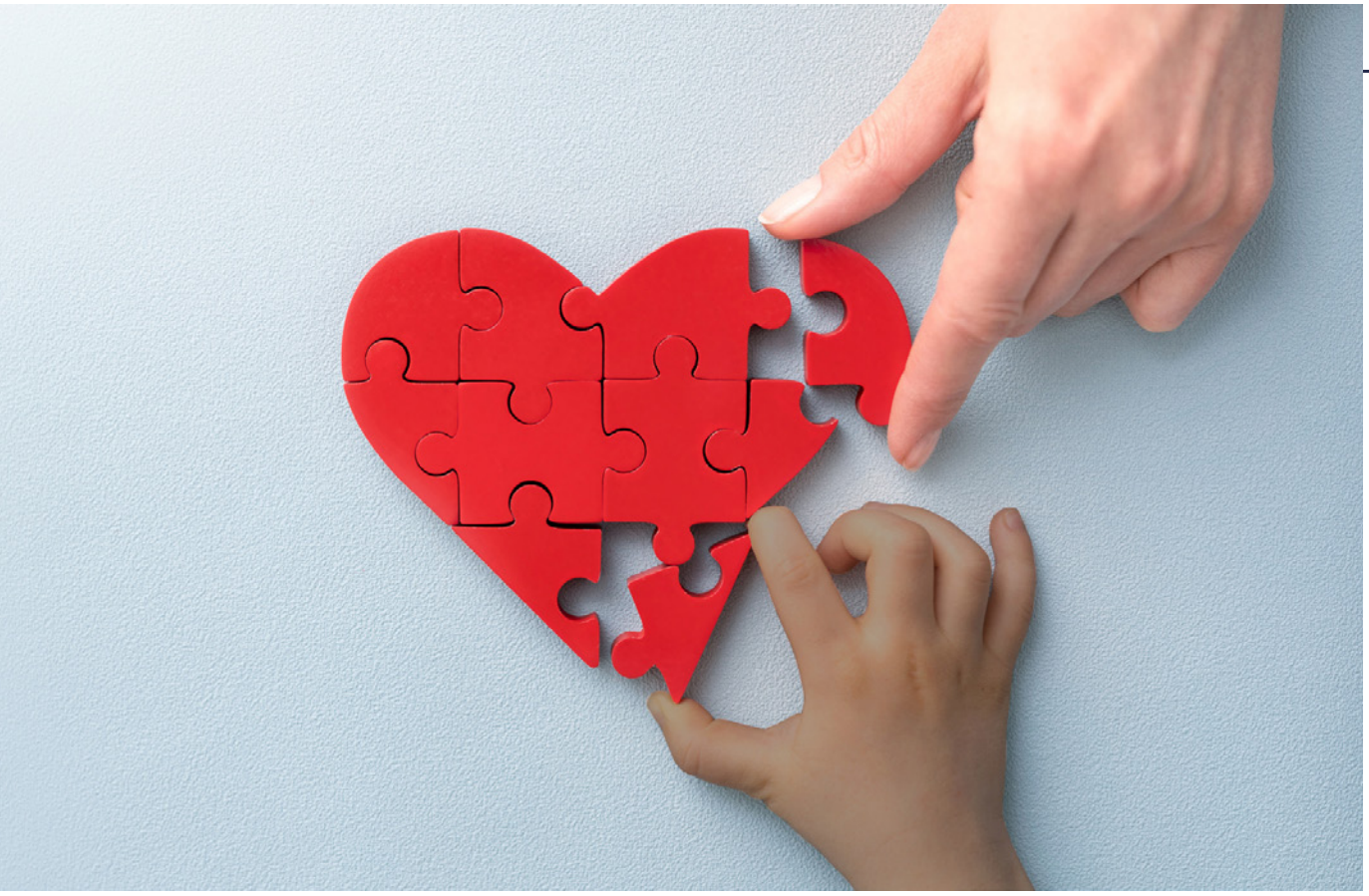
UNIVERSITY OF BIRMINGHAM

Transforming collaborative research

A national strategy to address the James Lind Alliance priorities for children and adults with congenital heart disease

July 2023



In partnership with:

James Lind Alliance
Priority Setting Partnerships

CHF

Somerville Heart Foundation

British Congenital Cardiac Association

SCTS
Society for Cardiothoracic Surgery in Great Britain and Ireland

supported by Birmingham Children's Hospital Charity
Doing more for sick kids

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FOREWORD

Congenital heart disease (CHD) is the most common type of birth defect, affecting 13 children born every day in the United Kingdom (UK).[1] It has a lifelong impact and with improved outcomes during childhood, there are now more adults than children living with CHD in developed countries.[2]

It therefore may be surprising that there is a lack of evidence to inform clinical decision-making in both children and adults with CHD. The Cochrane Library contains fewer than twenty reviews on CHD topics,[3] and there is a need for high-quality, multi-centre clinical trials that answer important questions to improve the daily lives and outcomes of those affected.[4] The British Heart Foundation (BHF) identified that 'we urgently need research breakthroughs to ensure survivors [of CHD] lead longer and healthier lives'.[5]

With national commissioning of specialised services and lifetime follow-up, access to National Institute for Health and Care Research (NIHR) infrastructure, well-developed clinical trials units, and the recently established All-Island CHD Network, the UK and Ireland should be an ideal environment in which to conduct world-leading CHD clinical research. However, the last two decades have been challenging times for CHD services in the UK, following the Bristol Royal Infirmary Inquiry,[6] coupled with a lack of clarity regarding the future configuration of national services, leading to an environment of uncertainty and limited collaboration between centres. In recent years, following publication of the British Congenital Cardiac Association (BCCA) statement on 'Multi-centre working',[7] this has begun to change, with several prospective multi-centre studies and there is enthusiasm amongst families, healthcare professionals and research funders to develop more collaborative research.

Working with the James Lind Alliance, we brought together patients with lived experience of CHD, their families, charities, and healthcare professionals in the UK to form the national Congenital Heart Disease Priority Setting Partnership.[8] Through a shared decision-making process, we determined two Top 10 lists of priorities for CHD research, one child/antenatal and one adult. Remarkably, six of the priorities were present on both lists, leading to 14 distinct clinical priorities: four child/antenatal, four adult and six throughout life.

This document sets out these research priorities and describes a national strategy to address them through collaborative research, endorsed by both professional bodies and national charity partners. First, to establish a UK and Ireland network for multi-centre research, focusing on clinical trials and other studies that have the potential to change clinical practice. Second, to set-up a national CHD Patient and Public Involvement (PPI) group, comprising engaged patient, parent, and charity members with lived experience or affected by CHD, to contribute through all stages of the research. Third, to develop specific working groups of clinicians, researchers, and PPI members, to address each of the priorities. And finally, to learn from the experiences of others in conducting multi-centre CHD research and translating research priorities into funded clinical studies.

The priorities provide a platform for conducting the research that matters most, whilst the strategy outlines a structure through which they can be translated into research questions and funded studies. I believe that together these present a unique opportunity to transform collaborative CHD research in the UK and Ireland for the benefit of the whole community.

Mr Nigel E Drury, PhD FRCS(CTh)

Academic Consultant in Paediatric Cardiac Surgery, Birmingham Children's Hospital
Associate Clinical Professor, Institute of Cardiovascular Sciences, University of Birmingham
Lead, Congenital Heart Disease Priority Setting Partnership
Congenital Cardiac Surgery Lead, SCTS Research committee

PROJECT TIMELINE

James Lind Alliance
Priority Setting Partnerships

JUNE 2020
SCTS Research committee identified need for congenital research priorities.

JANUARY 2021
George Davies Charitable Trust funds the CHD Priority Setting Partnership.

MARCH 2021
Initial James Lind Alliance PSP steering group meeting held via Zoom.

JUNE–OCTOBER 2021
Initial public survey open, promoted via partners and social media @congenitalPSP #CHDpriorities. 524 respondents submitted 1,373 questions.

OCTOBER 2021–MARCH 2022
Questions filtered, out-of-scope removed, summary questions formed and checked against literature. 56 child/antenatal and 47 adult uncertainties progressed.

MARCH–MAY 2022
Second surveys open, vote to prioritise topics. 250 child/antenatal and 252 adult survey responses.

MAY–JUNE 2022
Top priorities identified, 26 questions from each survey progressed.

JUNE 2022
Two final workshops in Birmingham, involving patients, parents, charities, and healthcare professionals, agreed Top 10 lists for child/antenatal and adult CHD.

NOVEMBER 2022
Priorities launched at BCCA Annual Conference in Birmingham.

CONGENITAL HEART DISEASE PRIORITY SETTING PARTNERSHIP

Priority Setting Partnerships (PSPs) provide an equitable mechanism for identifying and prioritising research that is important to patients, their families, and clinicians, through shared decision-making.[9] Over the last two years, we worked with charity and professional partners to identify national priorities for CHD research, as shown in the timeline.

A steering group of stakeholders with a wide range of lived experiences or professional interests in CHD, including patients, parents, charities, and healthcare professionals was assembled (see contributors) and agreed a protocol. The scope of the PSP was collectively defined as:

The management of CHD throughout life, including prior to birth, focusing on:

- Diagnosis, during pregnancy or after birth.
- Treatment: medical therapy, catheter intervention, surgery including mechanical support & transplantation, lifestyle, or psychosocial intervention.
- Outcomes of the conditions and/or treatments and the impact on patients and their families, including the physical, psychological, and social effects of living with CHD.

The PSP excluded from its scope questions about non-management related aspects of CHD, such as aetiology or non-clinical genetics; acquired heart disease, other than occurring in the context of CHD; and other co-morbidities, such as non-cardiac aspects of associated syndromes.

To protect potential priorities for the growing population of adults living with CHD, the steering group decided to split the process into parallel 'child/antenatal' and 'adult' tracks after the initial question gathering stage. The PSP comprised four stages:

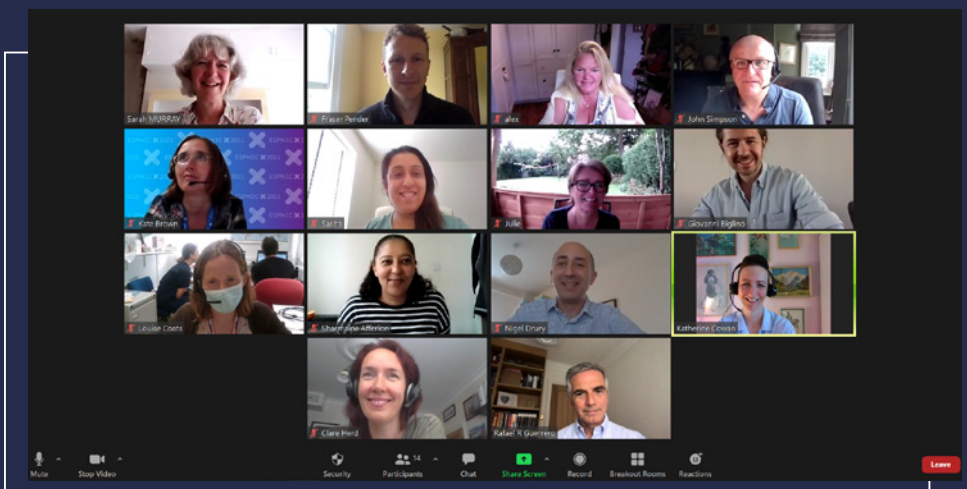
1. Initial survey: In a public survey, we asked: '*What questions would you like to see answered by future research, relating to the diagnosis, treatment, or outcomes of congenital heart disease?*' and invited respondents to pose up to three questions. The survey was publicised online, through partner organisations, and social and traditional media. A total of 524 patients, parents, charities, healthcare professionals, and others completed the survey.

2. Data processing and evidence checking: Responses were collated and tagged as relevant to children, adults, or both, and those identified as out of scope were removed. The remaining questions were divided into categories and the steering group developed indicative summary questions, using an iterative process to combine similar or overlapping questions, and reword into plain, consistent language. Of 1,373 submitted questions, 313 were deemed to be out of scope or duplicates and the remaining 1,060 questions were used to generate summary questions.

These were checked against the literature and those that were already answered were removed, with 56 child/antenatal and 47 adult uncertainties taken forward to the next stage.

3. Interim prioritisation surveys: Two second surveys, one child/antenatal and one adult, were conducted in which respondents were asked to choose up to ten of the most important uncertainties. 250 respondents completed the child/antenatal survey and 252 completed the adult survey. The questions ranked most highly by clinicians and/or non-clinicians were taken forward to the final workshops.

4. Final priority setting workshops: Two workshops were held in Birmingham in June 2022, one child/antenatal and one adult, bringing together patients, parents, charities, and healthcare professionals, with a range of conditions/expertise from across the UK and Ireland. Three experienced advisors from the James Lind Alliance facilitated the discussions to build consensus using an adapted nominal group technique, and the two workshops, comprising entirely separate cohorts of participants, independently agreed the final rankings. There was excellent engagement from all participant groups, with moving personal stories and passionate informed debate. Consensus was reached relatively quickly on both days and two final Top 10 lists of national research priorities were agreed, as shown over the following pages.




Welcome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Beyond the PSP

The Top 10s cover a wide range of clinical research areas including surgery, catheter interventions, intensive care, antenatal screening, psychology, cardio-obstetrics, electrophysiology, epidemiology, bioinformatics, pharmacology, technology, bioengineering, and transplantation. Many encompass holistic outcomes, looking beyond early mortality to improve the quality of survivorship and reduce the impact of living with CHD. Whilst diverse methodologies will be required to address these priorities, including qualitative studies, database analysis and translational research, many are well suited to clinical trials.

Randomised controlled trials are the gold standard for comparing healthcare interventions, using a predefined protocol, random allocation to treatments, rigorous testing, and minimisation of biases. When well designed and conducted, they are the most powerful tool that we have for determining whether one intervention is better than another for defined outcomes. Early phase II trials, often small, single centre studies, are useful to determine efficacy and safety but it is larger, usually multi-centre, phase III trials which determine clinical effectiveness, influence guidelines, and have the potential to change clinical practice worldwide. It is therefore through effective collaboration between centres that we will be able to best address these priorities.

TOP 10 PRIORITIES FOR RESEARCH

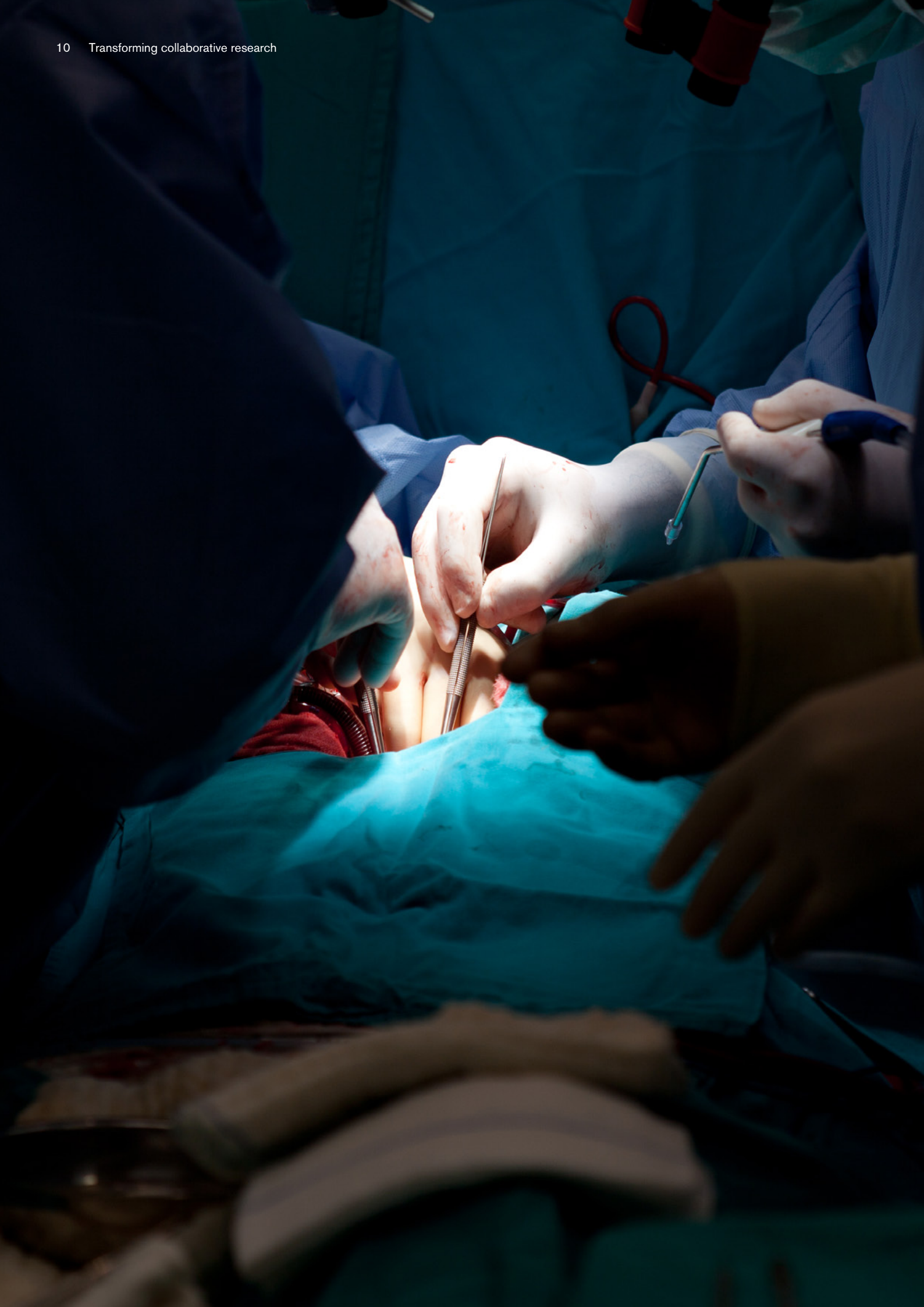
Top 10 priorities for child/antenatal congenital heart disease research

1. How can damage to organs (e.g. heart, brain, lung, kidney, bowel) during heart surgery in children with CHD be minimised to reduce complications, especially in those who require multiple operations?
2. How can pre- and post-natal screening strategies (e.g. scans, pulse oximetry, novel techniques) be improved to achieve greater accuracy, avoid late diagnosis, and reduce complications from CHD?
3. What are the effects of CHD, low oxygen saturations, and interventions on brain development and behavioural outcomes, and how can these be improved?
4. How can the frequency or need for reoperations be reduced for people with CHD (e.g. improved valve/conduit longevity or that grow with the patient)? *
5. How can technology be used to deliver personalised care and improve outcomes in CHD (e.g. artificial intelligence, 3D printing, genomics, stem cells, organ regeneration)? *
6. What is the impact of living with CHD on mental health in children and how can this be improved through access to psychological support and other therapies? *
7. What is the impact of living with CHD on quality of life in children and how can this be improved? *
8. How can less invasive interventions be performed for CHD with the same outcomes as open-heart surgery? *
9. How can the longevity of the Fontan circulation be prolonged and the impact of complications (e.g. liver, protein-losing enteropathy (PLE), renal, endocrine, fertility) be reduced? *
10. What are the long-term outcomes and life expectancy of children born with CHD?

** indicates priorities appearing on both lists that are derived from the same summary questions*

Top 10 priorities for adult congenital heart disease research

1. **How can less invasive interventions be performed for CHD with the same outcomes as open-heart surgery? ***
2. **How can the longevity of the Fontan circulation be prolonged and the impact of complications (e.g. liver, protein-losing enteropathy (PLE), renal, endocrine, fertility) be reduced? ***
3. **What is the impact of living with CHD on mental health in adults and how can this be improved through access to psychological support and other therapies? ***
4. **How can technology be used to deliver personalised care and improve outcomes of those with CHD (e.g. artificial intelligence, 3D printing, genomics, stem cells, organ regeneration)? ***
5. **What are the risks and limitations associated with pregnancy, childbirth, and motherhood for women with CHD, and what information and support is available?**
6. **What are the best treatment strategies for heart failure in adults with CHD, in particular those with a systemic right ventricle?**
7. **How can the management of arrhythmias, including sudden cardiac death, in adults with CHD be improved?**
8. **How can the indications, timing of referral, and outcomes of transplantation and long-term mechanical support in adults with CHD be improved?**
9. **What is the impact of living with CHD on quality of life in adults and how can this be improved? ***
10. **How can the frequency or need for reoperations be reduced for people with CHD (e.g. improved valve/conduit longevity or that grow with the patient)? ***



A NATIONAL STRATEGY TO ADDRESS THE PRIORITIES

This strategy has been developed to address the clinical priorities identified by the Congenital Heart Disease Priority Setting Partnership, with endorsement from professional bodies, British Congenital Cardiac Association (BCCA) and Society for Cardiothoracic Surgery in Great Britain and Ireland (SCTS), and national charity partners, the Children's Heart Federation and Somerville Heart Foundation. It aims to provide a structure through which the priorities can be translated into funded studies, to improve clinical care and the lives of those affected by CHD. The plan aligns with national policy documents, including The NHS Long Term Plan,[10] The UK Rare Diseases Framework,[11] and Saving and Improving Lives: The Future of UK Clinical Research Delivery.[12] It is supported by the NHS England Congenital Heart Disease Clinical Reference Group and the Women and Children's Programme of Care, and reflects the Standards and Specifications requirement for all CHD networks to work in partnership with other centres in research activity to improve patient care.[13]

1. Congenital Heart Research Network

To establish a UK and Ireland collaborative network for multi-centre studies, focusing on clinical trials and other studies that address the priorities and have the potential to change clinical practice.

The network will bring together all CHD centres in the UK and Ireland, in an open, inclusive, equitable and transparent collaboration. It will provide a framework for investigators to develop and lead studies, through working groups and with integrated PPI throughout. It will be affiliated with BCCA and SCTS and led by an executive committee with an elected chair and representation from both professional bodies, the PPI group, and others. The network structures, governance and operational model will be defined in detail in a policy manual, which will be widely consulted on and agreed, to encourage all interested parties to participate on an equal footing. There will be clear and agreed terms of reference, with no obligation for individual centres to take part in any specific study but appropriate recognition for those who are involved, including a principal investigator at each site.

The remit of the network will be to develop collaborative research across the UK and Ireland, to improve clinical care and outcomes, primarily through studies to answer the questions that matter most to patients, their families, and clinicians, as established through the PSP; however, it will also support studies that fall outside the scope of the PSP and would benefit from multi-centre collaboration, such as in children with acquired heart disease. Multi-centre clinical trials should be streamlined, efficient and innovative; they will be designed, conducted, and reported to the highest standards, generating high quality evidence to improve patient care in the UK and Ireland, and internationally. Prospective observational studies, analyses of routinely collected data, systematic reviews, and surveys of practice will also be conducted to address priorities, identify knowledge gaps, or support the need for or design of clinical trials. The network will oversee working groups on specific priorities, link with the national PPI group, and support individual investigators to deliver studies, as described below.

The network will utilise existing research infrastructure including:

- BHF Clinical Research Collaborative (BHF-CRC), established in 2019 to support the planning and delivery of high-quality clinical cardiovascular research across the UK, in particular late-phase clinical trials. It has encouraged the development of disease-specific cardiovascular research networks and provides infrastructure to facilitate, enhance, and coordinate research, both physical and information technology.
- Clinical trials units with expertise in the design and conduct of multi-centre trials, including paediatric, interventional, and surgical trials.
- NIHR Research Support Service and Clinical Research Networks, to support the development and delivery of portfolio-adopted studies through additional staff, facilities, equipment, support services and training.
- Regional NHS Operational Delivery Networks for CHD and the All-Island CHD Network, to improve outcomes through the integration of research into the patient pathway.

The network will work with other organisations to encourage better use of routinely collected data for CHD research. Working with the National Institute for Cardiovascular Outcomes Research (NICOR), the Paediatric Intensive Care Audit Network (PICANet), the National Congenital Anomaly and Rare Disease Registration Service (NCARDRS), the BHF Data Science Centre and others, patient-level data can be used to define the extent of need in the population, assess feasibility, test eligibility criteria, inform study design, benchmark outcomes, and model treatment effects in virtual trials. Embedding follow-up with the Office for National Statistics (ONS) through NHS Digital into prospective studies will enable tracking of long-term health-related outcomes.

The network will also work with NIHR to promote research training, to enhance the knowledge and skills of the CHD workforce, including Principal Investigator and PPI training. It will encourage and facilitate the next generation of cardiologists, surgeons, nurses, and allied health professionals, to become the research leaders of the future. Specifically, trainees should gain an understanding of the research lifecycle and clinical trial methodology through tailored training and have the opportunity to participate in studies at their centre, including through the NIHR Associate Principal Investigator Scheme.

With funding, the network will hold biannual investigator meetings in May and November, the latter linked with the BCCA Annual Conference, to discuss progress, plan future developments, and learn from each other.

2. National Patient and Public Involvement group

To set-up a national CHD PPI group, comprising engaged patient, parent, and charity members with lived experience or affected by CHD, to actively contribute through all stages of the design, conduct, and reporting of research.

The national PPI group will provide a platform for patients, parents, and charities to continue involvement in driving the research agenda. It will inform the development of the network and provide a valuable resource for shaping study proposals. Members should actively contribute to the development, conduct and reporting of research, bringing the benefit of their lived experience.

Representation should include people living with CHD across age groups and conditions, and their families, with a paediatric and/or adult focus; specific attention should be paid to involving fathers and those from ethnic minorities, both of whom were underrepresented in the PSP process despite extensive efforts. The group should come together, appoint a chair and vice-chair, agree terms of reference and facilitation, and meet regularly via minuted video conferencing to feedback on progress and exchange ideas and learning. It should continue to work with a broad range of external providers including BHF patient support groups, particularly Teen Heart (13-18 years) and One Beat (18-30 years), and engage with other established PPI groups, including Young Persons Advisory Groups (YPAGs). Members should receive training in clinical research, such as via the NIHR Learn platform and NIHR Centre for Engagement and Dissemination, and reimbursement for their time commitment, according to NIHR standards for PPI.

3. Clinical study groups

To develop working groups to address each of the priorities, comprising clinicians, researchers, and PPI members, and overseen by the network.

Of the 20 priorities identified in the CHD PSP, six on each list are derived from the same summary questions, leading to 14 distinct priorities: four child/antenatal-specific, four adult-specific, and six throughout life. Using a phased approach to build on interest and expertise, clinical study groups will be established to focus on addressing one or more of these 14 domains, with open engagement of all interested parties. Where an established national group already exists, they will be invited to form the working group, including adopting additional PPI or other members, if required.

Clinical study groups will bring together interested individuals with the relevant interests and expertise, including doctors, nurses, allied health professionals, researchers, patients, parents, and charities. Each group will be co-led by a clinician/researcher and a PPI member, as agreed by the group, and include representation from interested parties, with at least 2 PPI group members. The role of the group will be to translate the priority of interest into specific research questions. Individuals within the group will lead on the development of studies, ideally with clinician/researcher and PPI co-leads, and submit initial proposals to the network for assessment of feasibility and support. The investigators will develop applications for funding, which may require preliminary work such as systematic reviews to identify the evidence gap or pilot studies using routinely collected data, and the involvement of a clinical trials unit or other group with relevant expertise. Once funded, the investigators will lead the study, with input from the clinical study group and support from the network to develop a protocol, obtain any regulatory approvals and if prospective, start recruitment at sites.

Funding opportunities

- NIHR recognise the importance of the James Lind Alliance approach and have established rolling calls across their funding schemes dedicated to studies that address PSP priorities; the programmes currently participating are Efficacy and Mechanism Evaluation (EME), Health and Social Care Delivery Research (HSDR), Health Technology Assessment (HTA), and Public Health Research (PHR).
- The BHF Clinical Studies Committee was established in 2017 to fund interventional clinical trials and observational studies. Of the 22 studies funded to date across cardiovascular disease, only one is in CHD, the multi-centre del Nido versus St Thomas' blood cardioplegia in the young (DESTINY) trial.
- The BHF-CRC Research Development Fund offers up to £10K funding to support idea development or gather pilot data in preparation for a larger clinical study application. Applications must be supported by a member society, such as BCCA or SCTS.

Adding value

In addition to primary studies, proposals should look to add value through adjunctive studies, such as translational sub-studies (e.g. blood/biopsies for genomic/molecular phenotyping), qualitative studies to explore participants lived experience, and studies within a trial (SWAT) to evaluate clinical trial methods, working with established groups in the UK and Ireland. They should also seek to develop and validate outcome measures, including patient reported outcomes, and contribute to the COMET Initiative, to develop and agree standardised core outcome sets to be measured and reported in CHD clinical trials.

LEARNING FROM OTHERS

Defining national clinical priorities for research represents a unique opportunity for our community and has the potential to be transformative for collaborative CHD research in the UK and Ireland. It is the first time that the James Lind Alliance process has been applied to CHD, to give patients and their families an equal voice to clinicians in shaping the direction of research and enable all stakeholders to focus on the questions which matter most.

Yet collaborative research is not a new idea; our colleagues in adult and paediatric oncology have been conducting practice-defining multi-centre and often international clinical trials for decades, making the opportunity to participate and benefit from late-phase trials part of the routine care pathway.[14] In CHD, the Pediatric Heart Network (PHN) has been at the forefront of paediatric cardiology research for 20 years, and in the UK, the Adult Cardiac Surgery PSP has been the stimulus for a step change in multi-centre clinical trials in surgery for acquired disease.

Pediatric Heart Network

The PHN was set-up in 2001, funded by the US National Heart Lung and Blood Institute (NHLBI), to improve the outcomes and quality of life in children with heart disease and in recent years, expanded its remit to include adults with CHD.[15]

It supports doctors and nurses to design and conduct clinical research so that patients can receive high-quality, evidence-based care, and has three main components: clinical research-active hospitals, including eight core centres and 31 auxiliary sites; a data coordinating centre (NERI/HealthCore); and a central office located on the National Institutes of Health (NIH) campus in Bethesda, MD.

The PHN is a collaboration, with all core centres involved in project development and expected to participate in trials, with additional sites brought on board depending on enrolment and expertise. It is core funded by NHLBI but project teams apply for specific study funding, usually via NIH grant programmes. Its processes are guided by a network policy manual containing operational procedures and guidelines for the design and conduct of studies, including oversight committees, protocol development, quality assurance, publication, and research training. The network holds biannual steering group meetings to discuss projects at all stages of the research lifecycle and provide training on clinical trials, with dedicated sessions for trainees.

The PHN has transformed the landscape of paediatric and adult CHD clinical research in North America. To date, the PHN have conducted 23 studies, including 12 multi-centre clinical trials, with samples sizes ranging up to 1,250 participants, and over 150 peer-reviewed publications including the Single Ventricle Reconstruction trial, pulsed corticosteroids in Kawasaki disease, Z-scores on echocardiogram, and atenolol v losartan in children and young adults with Marfan syndrome.

James Lind Alliance Priority Setting Partnership in Adult Cardiac Surgery

In July 2019, the Adult Cardiac Surgery PSP, led by Professor Gavin Murphy at the University of Leicester, identified the Top 10 research priorities for patients, carers, and clinicians.[16]

Whilst many PSPs publish their findings and leave it for others to take forward,[17] the cardiac surgical community came together to form the National Cardiac Surgery Clinical Trials Initiative, to translate these priorities into a programme of clinical trials that address the most important questions. They set up Clinical Study Groups, led by interdisciplinary teams composed of members of the public, health researchers, and clinicians, to develop research questions and trial proposals within each of the priorities, and a national PPI group with representation as co-leads for each group. This approach is already bearing fruit, with several successful funding applications for clinical trials and programme development grants and has the potential to revolutionise adult cardiac surgery research in the UK through the integration of multi-centre clinical trials into routine care.

MOVING FORWARD

The UK and Ireland is a great environment in which to conduct collaborative CHD research, with recent infrastructure developments, such as the NIHR, BHF-CRC, and All-Island CHD Network, and a wealth of long-term patient-level data through NICOR and NHS Digital.

As a CHD community, we have a responsibility to provide scientific leadership and work together to conduct well-designed, rigorously conducted, multi-centre clinical trials and other studies that address the most important questions, to improve clinical care and outcomes for our patients and their families.

Through the CHD PSP, we brought together patients, their families, charities, and healthcare professionals to determine national priorities for research using an established shared decision-making process. This national strategy provides a roadmap to build on the success of the PSP through a network for multi-centre clinical studies, a national PPI group, and clinical study groups to translate the priorities into research questions and funded studies. Whilst many of the challenges of CHD research are intrinsic to the field, a cultural change towards making involvement in research part of the standard of care will be vital to delivering better outcomes. The true value of the PSP will be determined by its legacy in driving forward collaborative CHD research, how the structures described in this strategy are developed, and whether they flourish as intended. Specific markers of success will be:

- **Community:** PPI and clinician/researcher engagement, active involvement of trainees in clinical research, future appointments to University Chairs in CHD research.
- **Collaboration:** Number of trials and other studies funded, centres actively involved, and patients recruited, and the potential development of international collaborations.
- **Impact:** Studies leading to a tangible change in clinical practice, incorporation into national and international guidelines, and ultimately improve the outcomes and daily lives of those born with CHD.

We anticipate that the priorities identified in the CHD PSP will provide a platform for collaborative CHD research in the UK and Ireland for the next decade but as questions are addressed, there will be a need to reassess the priorities. The network will therefore monitor progress against the priorities and determine when there is a need to repeat the PSP process.

CONTRIBUTORS

In memory of Michael J Cumper, a long-time advocate for improving the lives of those affected by congenital heart disease, whose knowledge, empathy, and common sense approach were of great benefit in shaping the priority setting partnership, and sadly passed away in July 2022.

This report was written and developed by Mr Nigel Drury, on behalf of the Congenital Heart Disease Priority Setting Partnership steering group:

Clinicians

- Professor Katherine L Brown, Consultant in Cardiac Intensive Care, Great Ormond Street Hospital for Children, London and Chair, Institute of Cardiovascular Science, University College London
- Dr Louise Coats, Consultant Adult Congenital Cardiologist, Freeman Hospital, Newcastle upon Tyne and Clinical Intermediate Fellow, Population Health Sciences Institute, Newcastle University
- Mr Rafael R Guerrero, Consultant Congenital Cardiac Surgeon, Alder Hey Children's Hospital, Associate Professor, Faculty of Health and Life Sciences, University of Liverpool, and former Chair, SCTS Congenital Cardiac Surgery sub-committee (2019-22)
- Professor John M Simpson, Consultant Paediatric and Fetal Cardiologist, Evelina London Children's Hospital, Chair, School of Biomedical Engineering & Imaging Sciences, Kings College London, and former President, British Congenital Cardiac Association (2019-21)
- Professor John DR Thomson, Consultant Interventional Cardiologist, Leeds General Infirmary, and Professor of Pediatrics, Johns Hopkins Children's Center, Baltimore, MD

Patients and parents

- Alex Miskin, parent
- Sarah Murray, parent and Chair of the NICOR Patient Representative Group
- Fraser Pender, patient
- Sasha Rooprai, parent
- Jara Weinkauf, patient
- Julie Wootton, parent and Chair of Trustees, Children's Heart Federation

Other members

- Katherine Cowan, Senior Advisor, James Lind Alliance, National Institute for Health and Care Research
- Dr Clare P Herd, Systematic Reviewer, Institute of Applied Health Research, University of Birmingham
- Dr Giovanni Biglino, Associate Professor, Bristol Medical School, University of Bristol, and National Heart and Lung Institute, Imperial College London
- Sharmaine Afferion, Administrator, Institute of Cardiovascular Sciences, University of Birmingham

Correspondence to:
Mr Nigel Drury, Department of Paediatric Cardiac Surgery, Birmingham Children's Hospital, Steelhouse Lane, Birmingham, B4 6NH, UK
Email: **n.e.drury@bham.ac.uk.**

Further information about the priority setting partnership is available from the project website:
https://www.birmingham.ac.uk/congenital-psp
and the James Lind Alliance: **https://www.jla.nihr.ac.uk/priority-setting-partnerships/congenital-heart-disease**

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Finally, we thank Dr Gail D Pearson and colleagues at the Pediatric Heart Network, National Heart Lung and Blood Institute, Bethesda, MD for welcoming Nigel Drury on two visits in 2017-18, sharing their experiences of developing the network, giving access to their protocols and other documents, and providing the opportunity to join their steering committee meeting as a special guest. The visits were funded by the British Heart Foundation, as part of an Intermediate Clinical Research Fellowship, and the University of Birmingham, through the Birmingham International Engagement Fund.

DISCLAIMER

The views expressed are those of the author and not necessarily those of the National Health Service, National Institute for Health and Care Research, British Congenital Cardiac Association, Society for Cardiothoracic Surgery in Great Britain and Ireland, Children's Heart Federation, Somerville Heart Foundation, British Heart Foundation, or University of Birmingham.

The Priority Setting Partnership and thereby this strategy for addressing the priorities was explicitly focused on clinical priorities for research. The steering group agreed on this approach to maximise the potential for translation into clinical trials with potential patient benefit in the short- to medium-term. However it is acknowledged that by excluding non-management related research domains, such as understanding the underlying causes of congenital heart disease, we dismissed the major translational impact which these more fundamental questions may have for future generations. This strategy therefore is not meant to represent a comprehensive approach for all congenital heart disease research, rather a focused plan to harness the collective power of the community to address the clinical priorities we have established.

The plans outlined in this strategy, including the establishment of the Congenital Heart Research Network, are not designed to replace existing collaborations but to provide a mechanism for building further multi-centre collaborations with patients at their centre. There is no obligation for clinicians or researchers to use the network for conducting multi-centre studies, but we hope that the benefits will be clear.

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