


A referral pathway for potentially abnormal neurodevelopment in children with heart disease in the United Kingdom: a Delphi consensus

Aparna Hoskote ^{1,2} Jo Wray,^{1,2} Victoria Banks,^{1,2} Katherine Brown,^{1,2} Monica Lakhanpaul^{2,3,4}

To cite: Hoskote A, Wray J, Banks V, *et al.* A referral pathway for potentially abnormal neurodevelopment in children with heart disease in the United Kingdom: a Delphi consensus. *BMJ Paediatrics Open* 2020;**4**:e000587. doi:10.1136/bmjpo-2019-000587

► Additional material is published online only. To view please visit the journal online (<http://dx.doi.org/10.1136/bmjpo-2019-000587>).

Received 14 October 2019
Revised 22 January 2020
Accepted 30 January 2020



© Author(s) (or their employer(s)) 2020. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

¹Heart and Lung Directorate, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK

²NIHR Great Ormond Street Hospital Biomedical Research Centre, London, UK

³Population, Policy and Practice Department, UCL Great Ormond Street Institute of Child Health, London, UK

⁴Whittington Health NHS Trust, London, UK

Correspondence to

Prof Monica Lakhanpaul; m.lakhanpaul@ucl.ac.uk

ABSTRACT

Introduction Children with congenital heart disease have complex medical and neurodevelopmental needs. We aimed to develop a multi-professional consensus-based referral pathway applicable to action the results of the brief developmental assessment (BDA), a validated early recognition tool, that categorises the neurodevelopmental status as green (appropriate for age), amber (equivocal) or red (delayed) in children aged between 4 months and 5 years.

Methods A Delphi consensus survey detailing two scenarios—a child categorised as delayed (red) and another as equivocal (amber) on administration of the BDA at the time of discharge from the tertiary centre—was sent to 80 expert professionals from primary, secondary and tertiary care seeking agreement on next steps and referral pathways. An iterative process was proposed with a pre-defined rule of 75% for consensus.

Results The survey was completed by 77 Delphi panel experts in Round 1, 73 in Round 2 and 70 in Round 3. Consensus was achieved (1) for the child with amber or red BDA, the child should be under the care of a paediatrician with expertise in cardiology (PEC) (or general paediatrician if no PEC) based at their local hospital, (2) for the child with red BDA, the PEC should initiate referral to community services at first assessment, (3) for child with amber BDA, a re-assessment by the health visitor should occur within 1–2 months, with referral to community services and notification to the PEC if on-going concerns.

Conclusions The Delphi process enabled a consensus to be reached between health professionals on referral pathways for specialist neurodevelopmental assessment/treatment for children with heart disease, in response to amber or red BDA results. The agreed referral pathway, if implemented, could underpin a national guideline to address and intervene on the neurodevelopmental difficulties in children with heart disease.

INTRODUCTION

Children with congenital heart disease (CHD) have complex medical requirements (cardiac surgery, multiple hospitalisations, medications, associated syndromes), which may negatively impact their neurodevelopment—a key factor in their general well-being

What is known about the subject?

- Children with congenital heart disease (CHD) have complex medical and neurodevelopmental needs with neurodevelopmental abnormalities leading to potentially devastating long-term sequelae.
- Routine neurodevelopmental surveillance, screening and evaluation in children with CHD is well-established in cardiothoracic programmes in USA and parts of Europe unlike in UK.
- An early recognition tool leading to intervention is an important first step in identifying children with delayed development in a high-volume cardiology service.

What this study adds?

- A multi-professional Delphi consensus identified referral pathways for children (4 months–5 years) with congenital heart disease (CHD) using early recognition tool–Brief Developmental Assessment–BDA.
- Consensus that children with delayed development (red BDA) should be referred immediately and those suspected (amber) should be re-assessed before referral to community paediatricians.
- The paediatrician with expertise in cardiology is central to the management of children with CHD and follow-up of suspected and/or confirmed neurodevelopmental delay.

and educational attainments.^{1–7} Currently, there is no formalised neurodevelopmental screening or surveillance pathway for children with CHD in the UK above that offered to healthy children. Research in the USA has identified neurodevelopmental abnormalities in as many as 25% of surgical survivors^{18–17} and the American Heart Association in their scientific statement¹⁸ has outlined an algorithm for a surveillance, evaluation and management for children with CHD. In the UK, a recent consultation exercise with



clinicians from primary, secondary and tertiary care and lay stakeholders as part of the National Institute for Health Research (NIHR)-funded paediatric cardiac morbidity surgery study¹⁹ ranked neurodevelopmental problems as the number one complication concern for those undergoing intervention for CHD.²⁰ Our research indicates that there are gaps in the follow-up of children with CHD with developmental difficulties.²¹ While there are many screening tools, all require specialised personnel, are time-consuming, expensive and undeliverable on a wide-scale in a resource-constrained nationalised health service (NHS). Hence, within the context of this NIHR study, an innovative, easy to administer 'early recognition tool'—brief developmental assessment (BDA)—was developed and validated in 971 children aged 4 months–5 years in three paediatric tertiary cardiac centres in London, UK.^{22 23} The BDA covers different age bands (17–34.9 weeks, 35–60 weeks, 15 months–2.9 years, 3.0–4.9 years) accounting for different stages of child development and covers domains of gross motor skills, fine motor skills, daily living skills, communication, socialisation and general understanding. The BDA allows categorisation of a child's current neurodevelopment as green (appropriate for age), red (delayed) or amber (equivocal) to help direct care through early recognition, enabling children with concerns to be referred for specialist assessment/treatment. The ultimate goal is for the BDA to be used in the NHS by practitioners who are not neurodevelopmental specialists, as resources do not exist for specialist assessments to be carried out on every child with CHD.

Aim and objectives of the Delphi survey

The aim was to develop a multi-professional consensus-based protocol for actions on the application of the BDA in age groups (from 4 months to 5 years) in children with CHD in the UK. The main objectives were to (1) recruit a multidisciplinary group of health professionals from primary, secondary and tertiary healthcare caring for children with CHD and (2) seek agreement on referral pathways for managing a child with amber or red BDA result to maximise effectiveness of the BDA tool.

METHODS

Recruitment and selection of the Delphi panel

We identified a multidisciplinary group of primary, secondary and tertiary healthcare professionals within the CHD networks in the UK, comprising of paediatric cardiologists, paediatric neurologists, paediatric neurodisability professionals, paediatricians with expertise in cardiology (PEC), general paediatricians, community paediatric nurses, community paediatricians, health visitors (HVs), cardiac nurse specialists, advanced nurse practitioner/nurse consultant, general practitioners (GP) and parent representatives. To ensure an adequate knowledge base, those who had been in their role for at least 2 years were invited by email giving information on the BDA and

requesting participation in the Delphi panel. Those who formally agreed were sent the Delphi survey. The regions identified were London, East of England, South East, North East, East Midlands, West Midlands, South West of England, Yorkshire and Humber, Wales, Scotland, Northern Ireland and North West of England. The Delphi Consensus Survey was registered as a service improvement project and ethical approval was not required.

Study design for Delphi consensus survey

The survey was designed using SurveyMonkey software containing two scenarios, each presenting a BDA result for a child with CHD aged between 4 months and 5 years at discharge from the tertiary centre. This age bracket has been chosen as the BDA was validated in this age group.²³ Alongside each scenario, respondents were sent a relevant example of the BDA along with standard information to be shared with all relevant health professionals. Scenario 1 described a child with amber BDA admitted to the tertiary centre for an intervention (surgery or catheter treatment), who is found to have *developmental concerns on the BDA* (score of amber=not fulfilling some of the milestones based on population norms, equivocal result). Scenario 2 described a child with red BDA admitted to the tertiary centre for an intervention (surgery or catheter treatment), who is found to have *developmental concerns on the BDA* (score of red=lagging behind the milestones based on population norms). The survey consisting of a mix of open questions with free-text response and specific closed questions on the two scenarios was electronically sent with a unique web link to participating professionals, who were asked to rate responses on a Likert scale: strongly disagree=1, disagree=2, moderately disagree=3, mildly disagree=4, undecided/don't know=5, mildly agree=6, moderately agree=7, agree=8, strongly agree=9. Non-responders would be sent reminder emails to complete the survey.

Development process of the Delphi survey

A 4-step process was followed in the development of the Delphi survey. A core team—AH, KB, JW—developed the Delphi survey based on findings from two previously published studies in which the BDA was developed²² and then validated²³ (funded by NIHR Health Services and Delivery Research). This draft survey was refined by the fourth author—ML. Following this, the Delphi survey and the proposed methodology were reviewed by an independent expert on Delphi methodology, which led to further refinements. The final step was pilot use of the survey with two paediatricians, leading to further minor modification based on their feedback.

Data analysis

The results were grouped as: agree—if the level of agreement was 7, 8, 9; middle ground—if the level of agreement was 4, 5, or 6; and disagree—if the level of disagreement was 1, 2 or 3 to facilitate identification of areas of agreement or disagreement or middle ground with the main goal of reaching a consensus. This methodology has been

well described in several widely used paediatric guidelines in the UK.^{24–26} Free-text responses were categorised and analysed as follows: in line with the Delphi methodology,²⁷ three of the authors—AH, KB, JW—independently looked at all of the comments and identified the key themes. These themes were then discussed by the authors, and were then collated, summarised and synthesised to inform the next round of questions. Comments from the participants were also used to provide additional context for the findings.

Definition of consensus

We established an a priori criterion of 75% approval to define consensus—75% of the panellists selecting 7, 8 or 9 of the 9-point Likert scale. Any questions or statements with (1) clear disagreement or (2) middle ground (no clear agreement or disagreement) would be revised and re-sent on a subsequent Delphi round until consensus was reached. The results would be sent with each iterative round.

In accordance with current national healthcare provisions, we stated a presumption that there would be a PEC in most district general hospitals, and if not, the default would be a general paediatrician. We also presumed that children <5 years of age will have a HV in their local community.

Patient and public involvement

Lay stakeholders including parents were consulted along with clinicians from primary, secondary and tertiary care as part of the National Institute for Health Research-funded paediatric cardiac morbidity surgery study¹⁹ and they ranked neurodevelopmental problems as the

number one complication concern for those undergoing intervention for congenital heart disease²⁰. Parent representatives and other stakeholders have been involved with the design, conduct and dissemination at every stage of the project.

RESULTS

Of the 164 invited professionals across the UK, 87 (53%) agreed to participate (table 1). The geographical distribution was weighted towards South East of England (table 2). There were no differences in professional background or geographical location between participants and non-participants. All panellists were experienced with >5 years of experience in their specialty, at the time of completing the survey, 53 (66%) had been in the role within their current organisation for >5 years, while the rest were in the role for 2–5 years.

Round 1

(online Supplementary material) The results of Round 1 are outlined in table 3. The respondents supported their choice by giving open text comments. Selected pertinent comments and themes that emerged were as follows:

Amber BDA scenario: (1) the tertiary cardiac team who has administered and identified the amber BDA will be aware of the child's GP and PEC, but will not be familiar with the community paediatric services in the child's local area, (2) the GP needs to be the health professional initiating new referrals, because referrals may not be accepted if made by one consultant to another (ie, bypassing the GP), (3) the PEC or general paediatrician may not be

Table 1 Professionals who participated in the Delphi consensus rounds

Specialty	Invited	Accepted	Completed round 1 n=77	Completed round 2 n=73	Completed round 3 n=70
Tertiary care professionals n=32*					
Paediatric cardiologist	20	12	9 (75)	9 (75)	8 (66)
Clinical nurse specialist	28	12	12 (100)	9 (75)	9 (75)
Advanced nurse practitioner/nurse consultant	5	4	4 (100)	4 (100)	4 (100)
Paediatric neurologist	8	4	4 (100)	4 (100)	4 (100)
Secondary care professionals n=25*					
Paediatric neurodisability	14	8	8 (100)	8 (100)	8 (100)
Paediatrician with expertise in cardiology	15	11	11 (100)	11 (100)	11 (100)
General paediatrician	15	6	5 (83)	4 (66)	3 (50)
Primary care professionals (n=24)*					
Community paediatric nurse	5	3	3 (100)	3 (100)	3 (100)
Community paediatrician	22	12	11 (92)	11 (92)	11 (92)
Health visitor	9	4	3 (75)	3 (75)	2 (50)
General practitioner	12	5	3 (60)	3 (60)	3 (60)
Parent representatives n=6*					
Parent representatives	11	6	4 (67)	4 (67)	4 (67)

*Total number who accepted the invitation to join the Delphi panel.

**Table 2** Distribution of Delphi panel experts

Region	Number of Delphi panellists
London	29
East of England	10
South East	12
North East	2
East Midlands	3
West Midlands	6
South West	5
Yorkshire and Humber	1
Wales	2
Scotland	4
Northern Ireland	1
North West	2
Total	77

equipped to assess child development and hence undertake re-assessment, and (4) there were contradictory comments about the HV skills (positive from secondary/primary care) and comments that cardiac professionals may have limited understanding of the HV service.

Red BDA scenario: there was no consensus on which health professional should be undertaking the referral to the community paediatrician which necessitated a second Delphi round. Respondents cautioned against referral delays and suggested that 'it did not matter who made the referral as long as it happened' online supplementary material.

Round 2

The results of Round 2 are outlined in [table 4](#). For amber BDA scenario: the required level of consensus of 75% was not reached on (1) referral by HV to PEC and (2) whether the child should be re-assessed before referral to the community paediatrician, resulting in a third Delphi round.

For red BDA scenario: the panel agreed that referral with red BDA result should be made by the PEC to the community paediatrician (82%). The Delphi panellists commented that (1) there should be robust communication between primary, secondary and tertiary professionals, (2) delays in referral communication and multiple referral/appointments should be avoided and (3) referral to community services should be prompt online supplementary material.

Round 3

For amber BDA scenario, the panel agreed that if the HV had concerns in the 1–2 month assessment after discharge from the tertiary centre, the HV should refer to the community paediatrician (with notification to the PEC); however, only 73% of the panel agreed that the child should be re-assessed by the HV 1–2 months after

discharge ([table 4](#)). On analysing the free text comments, it appears that the response was influenced by the perception that (1) there is significant variation in HV service within the UK, (2) HV service is overburdened and (3) children who have started school (at 4 years) may no longer be under HV in some areas. Of note, all the HVs/GPs in the panel agreed that re-assessment should be undertaken by the HV online supplementary material.

The results from 3 Delphi rounds are summarised in [table 5](#).

DISCUSSION

Neurodevelopmental difficulties can potentially be the most devastating long-term sequelae for children with CHD^{28–30} and as the number of survivors with complex cardiac conditions increase, there is a growing demand for these children to have adequate support within the wider healthcare system. Routine formal neurodevelopmental testing can be difficult to enforce within the constraints of the NHS. The validated BDA would theoretically function as an early recognition tool to sign-post those children with CHD who have neurodevelopmental problems. However, implementation of such a tool serves no useful purpose unless intervention and referral pathways are established. Using the Delphi consensus process with iterative rounds and feedback loops, with a panel comprising experts from primary, secondary and tertiary care within different parts of the UK, our study showed that consensus on referral patterns can be established for children with CHD who have been identified to have neurodevelopmental concerns by the BDA.

Consensus

The Delphi expert panel reached consensus that: children with either amber or red BDA should be under the care of the PEC and referred at the time of discharge from the tertiary centre by the child's primary cardiologist with results shared with the GP, HV, cardiologist and other relevant health professionals ([figure 1](#)). Children with a red BDA should be referred to a community paediatrician and local child development team (if not already under one) at the time of the assessment to minimise any delay. Children with an amber BDA should have a re-assessment by the local HV 1–2 months after discharge and be referred to the community paediatrician if there are persisting concerns, with a notification to the PEC. The PEC or designated paediatrician at the local hospital and the HV (in the case of younger babies and toddlers) were identified as the key health professionals in an ideal position to link up a child with CHD with developmental problems and their local child development team. In addition, the PEC was identified as an important link with the tertiary cardiac centre. Of note, the Delphi responses did not support a new referral to the PEC/paediatrician coming from the specialist nursing team in the tertiary hospitals.

Table 3 Delphi consensus survey—results from round 1

Round 1	% agree	% middle ground	% disagree
Q5. All children with CHD and amber BDA should be under the care of a (general paediatrician if no PEC) based at their local hospital.	75	16	9
Q6. If a child with CHD and amber BDA is not under the care of a PEC it is the responsibility of the child's paediatric cardiologist to refer the child to a PEC*(local general paediatrician if no PEC).	79	12	9
Q7. If a child with CHD and amber BDA is not under the care of a (local general paediatrician if no PEC), then a referral from the tertiary hospital under a specialist nursing team to a PEC/general paediatrician is acceptable.	60	25	16
Q8. The request for referral should have clinical details and the BDA assessment.	87	6	6
Q9. The complete results of the amber BDA should be shared with the child's PEC*(local general paediatrician).	91	6	3
Q10. The complete results of the amber BDA should be shared with the child's GP.	91	5	4
Q11. The complete results of the amber BDA should be shared with the child's HV.	84	10	5
Q12. All children with CHD and an amber BDA at the point of discharge following cardiac intervention should be re-assessed (in terms of development and general health) after a period of time by the PEC*(local general paediatrician).	65	21	14
Q13. All children with congenital heart disease and an amber BDA at the point of discharge following cardiac intervention should be re-assessed after a defined period of time by the child's HV.	71	18	10
Q14. All children with congenital heart disease and an amber BDA at the point of discharge following cardiac intervention should be re-assessed after a defined period of time by the child's GP.	35	43	22
Q15. Referral of children with CHD and amber BDA (not already under local health services) to community paediatrician should be undertaken at the point of first assessment when an amber BDA is detected at discharge following cardiac intervention.	64	22	14
Q16. Referral of children with CHD and amber BDA (not under local health services) to PEC*(local general paediatrician) should be undertaken at the point of first assessment when an amber BDA is detected at discharge following cardiac intervention.	70	18	12
Q17. Children with CHD and amber BDA should be re-assessed after a defined period and then referred to a community paediatrician if there is on-going concern.	70	18	12
Q18. Referral of children with CHD and amber BDA to a community paediatrician should be undertaken by the PEC*(local general paediatrician if no PEC).	69	21	10
Q19. Referral of children with CHD and amber BDA to a community paediatrician should be undertaken by the child's HV.	40	35	25
Q20. Referral of children with CHD and amber BDA to a community paediatrician should be undertaken by the tertiary paediatric cardiac team.	48	29	23
Q21. All children with CHD and red BDA should be under the care of a PEC* (local general paediatrician if no PEC) based at their local hospital.	77	17	6
Q22. If a child with CHD and red BDA is not under the care of a PEC* (local general paediatrician if no PEC), it is the responsibility of the child's paediatric cardiologist to refer the child to a PEC* (local general paediatrician if no PEC).	79	17	4
Q23. If a child with CHD and red BDA is not under the care of a PEC* (local general paediatrician), then a referral from the tertiary hospital specialist nursing team to a PEC*(local general paediatrician) is acceptable.	55	29	17
Q24. The complete results of the red BDA should be shared with the child's PEC* (local general paediatrician if no PEC).	94	5	1
Q25. The complete results of the red BDA should be shared with the child's GP.	91	8	1
Q26. The complete results of the red BDA should be shared with the child's HV.	92	6	1
Q27. The complete results of the red BDA should be shared with other relevant health professionals involved with the child such as neurologist, child development clinic, and geneticist.	95	4	1
Q28. All children with CHD and red BDA should be under the care of a community paediatrician and local child development team.	91	6	3

Continued

**Table 3** Continued

Round 1	% agree	% middle ground	% disagree
Q29. Referral of children with CHD and red BDA to a community paediatrician should be undertaken at the point of first assessment where an abnormal BDA is recorded at discharge following cardiac intervention (if child is not already under one).	81	16	4
Q30. Referral of children with CHD and red BDA to a community paediatrician should be undertaken if there is on-going concern after a period of re-assessment by the child's PEC*(local general paediatrician if no PEC).	64	21	16
Q31. Referral of children with CHD and red BDA to a community paediatrician should be undertaken by the child's *(local general paediatrician if no PEC).	73	21	6
Q32. Referral of children with CHD and red BDA to a community paediatrician should be undertaken by the child's HV.	43	31	26
Q33. Referral of children with CHD and red BDA to a community paediatrician should be undertaken by the child's GP	39	32	29
Q34. Referral of children with CHD and red BDA to a community paediatrician should be undertaken by the child's paediatric cardiac team	69	19	12

The results from responses were coded as: agree—if the level of agreement was 7, 8 or 9; middle ground—if the level of agreement was 4, 5 or 6; and disagree—if the level of disagreement was 1, 2 or 3.

BDA, brief developmental assessment; CHD, congenital heart disease; GP, general practitioner; HV, health visitor; PEC, paediatricians with expertise in cardiology.

Healthcare across sectors

As a background to this consensus process, it is important to acknowledge the current care provision relevant to developmental needs for children with CHD, although this can be highly variable.

Tertiary care: Children with CHD are always under a cardiologist and a cardiac specialist nursing team, who may not necessarily have 'general paediatric' and 'child

development' expertise but assess the child at critical time points particularly in early infancy. The BDA is intended for use as an early recognition tool for child neurodevelopment in this setting.

Secondary care: Within the setting of a non-specialist hospital, children with CHD are under the care of a PEC. In a small number of hospitals where a PEC is not in post, a named paediatrician will have designated responsibility

Table 4 Delphi consensus survey—results from rounds 2 and 3

	% agree	% middle ground	% disagree
Round 2			
Q1. At first assessment when identified to have an amber BDA, the child with CHD (if not already under local services) should be referred by the tertiary cardiologist to PEC (general paediatrician if no PEC).	77	8	14
Q2. Any on-going developmental concerns after discharge from tertiary cardiac centre if noted by HV should be referred to PEC (general paediatrician if no PEC at local hospital).	72	10	18
Q3. The child with amber BDA should be re-assessed before referral to the community paediatrician.	46	30	24
Q4. If there are any on-going developmental concerns, the PEC (general paediatrician where there is no nominated PEC) should refer to the community paediatrician	86	11	3
Q5. The referral to community paediatrician containing the results of the red BDA should be made by the PEC (general paediatrician if no nominated PEC).	82	11	7
Round 3			
Q1. The child with amber BDA should be re-assessed by the HV 1–2 months after discharge home.	73	15	12
Q2. If concerns are noted at the HV assessment 1–2 month after discharge from tertiary centre, the HV should refer to the community paediatrician with a notification to the PEC (general paediatrician if no PEC).	90	6	4

The results from responses were coded as: agree—if the level of agreement was 7, 8 or 9; middle ground—if the level of agreement was 4, 5 or 6; and disagree—if the level of disagreement was 1, 2 or 3.

BDA, brief developmental assessment; CHD, congenital heart disease; HV, health visitor; PEC, paediatricians with expertise in cardiology.

Table 5 Delphi consensus survey—summary of results from rounds 1, 2 and 3

	Amber BDA	Red BDA
Round 1		
75% consensus achieved	<ol style="list-style-type: none"> 1. The amber BDA result should be shared with the GP, HV, PEC and other relevant health professionals (91%). 2. Child should be under PEC at local hospital (75%). 3. It is the responsibility of the child's paediatric cardiologist in the tertiary centre to refer the child to a PEC (79%). 	<ol style="list-style-type: none"> 1. The red BDA result should be shared with the GP, HV, PEC and other relevant health professionals (90%). 2. Child should be under community paediatrician (91%). 3. Referral to a community paediatrician should be undertaken at the point of first assessment when abnormal BDA is recorded (81%).
75% consensus not achieved	<ol style="list-style-type: none"> 1. Timing of referral and to whom: at first assessment by tertiary cardiac centre to the PEC (70%) and to the community paediatrician (64%). 2. Whether re-assessment should be undertaken before referral to the community paediatrician (70%). 3. Which professional should undertake re-assessment: HV (71%), PEC (65%) and GP (35%). 4. Referral to community paediatrician by whom: PEC (69%), HV (40%) and tertiary cardiac centre (48%). 	<ol style="list-style-type: none"> 1. On who should make this referral to community paediatrician?—PEC (73%), tertiary cardiac team (69%), HV (43%) and GP (39%).
Round 2		
75% consensus achieved	<ol style="list-style-type: none"> 1. Child with CHD at first assessment when identified to have an amber BDA should be referred by the tertiary cardiologist to the PEC (77%). 	<ol style="list-style-type: none"> 1. The referral to the community paediatrician containing the results of the red BDA should be made by the PEC (82%). 2. If there are any on-going developmental concerns, the PEC should refer to the community paediatrician (86%).
75% consensus not achieved	<ol style="list-style-type: none"> 1. Any on-going developmental concerns after discharge from tertiary cardiac centre if noted by HV should be referred to PEC (72%). 2. The child with amber BDA should be re-assessed before referral to the community paediatrician (46%). 	—
Round 3		
75% consensus achieved	If the HV had concerns in the 1–2 month assessment after discharge from the tertiary centre, the HV should refer to the community paediatrician with a notification to the PEC—90%.	—
75% consensus not achieved	The child with amber BDA should be re-assessed by the HV 1–2 months after discharge home (73%).	—

BDA, brief developmental assessment; CHD, congenital heart disease; GP, general practitioner; HV, health visitor; PEC, paediatricians with expertise in cardiology.

for the paediatric needs of cardiac children. Not all cardiac children are formally under the care of a PEC or paediatrician (eg if they have never been to the local hospital); however a PEC or paediatrician with designated responsibility is available for a cardiac child should the need arise. The PEC (unlike the tertiary hospital team) will be aware of child development services available locally.

Primary and community care: all children, including those with CHD, are under a GP and at pre-school age, a HV. Both GP and HV are experienced in the referral of children with possible developmental problems to child development teams and will be cognisant of the local services. It is well known that HV services are under

pressure, with recent reports indicating that many children miss out on these visits.^{31–33} Child development teams are based within individual areas that they serve and are often linked to specific non-specialist hospitals. A child with CHD will be under the care of a child development team only if specifically referred.

Areas where agreement was more challenging

The main area where there was lack of agreement related to the child with an amber BDA. Of note, a child with suspected developmental delays may benefit from close follow-up and re-assessment given that these are crucial to identify the need for early intervention. Early identification and timely intervention contribute significantly

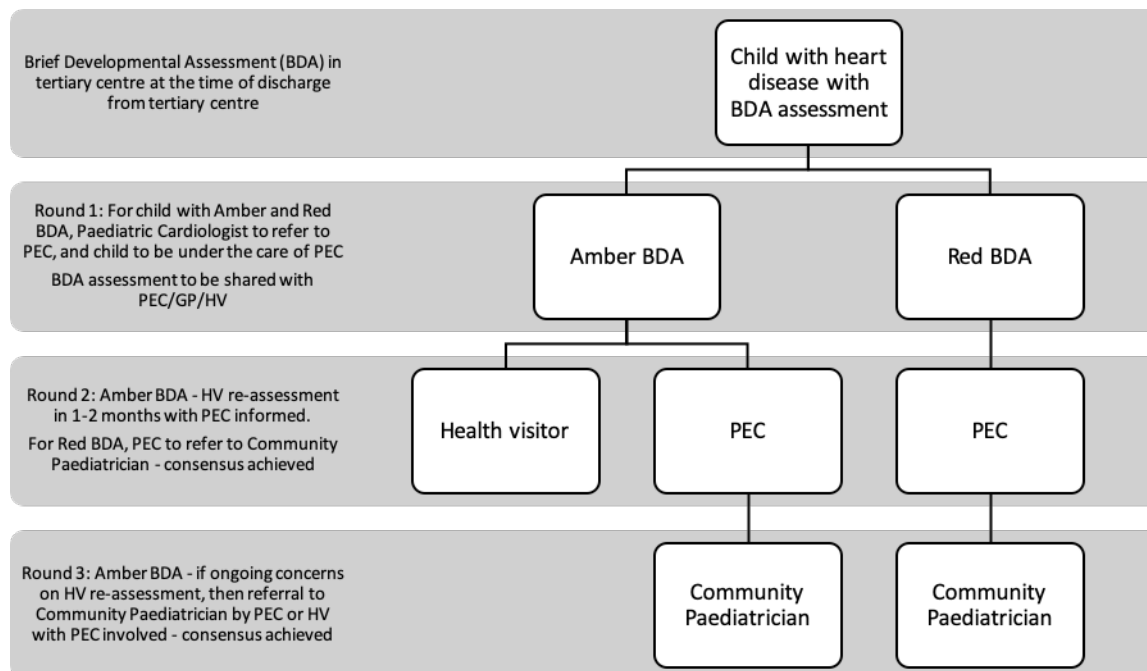


Figure 1 Delphi consensus for referral pathway for child with heart disease who has neurodevelopmental concerns—amber or red BDA—at discharge from tertiary centre. The consensus for referral pathway for amber or red BDA agreed from the iterative rounds of the Delphi consensus process is shown in the figure. For the child with amber BDA, this may need to be locally/regionally defined and adapted to local resource availability. BDA, brief developmental assessment; GP, general practitioner; HV, health visitor; PEC, paediatricians with expertise in cardiology.

towards a child's overall adjustment and quality of life.³⁴ There was moderate support for HV re-assessment, PEC re-assessment and referral to community paediatrics if on-going concerns with the child's development, but the consensus for re-assessment by the HV fell slightly short (73%) of the required 75% level for agreement. However, despite being in the minority in the panel, all the primary care representatives (HVs included) agreed that the child with amber BDA should have a re-assessment by the HV, and this response pattern was similar to that of the secondary and tertiary professionals. While there was no agreement for a child with amber BDA to be under the care of the community paediatrician, there were also conflicting opinions over whether or not the PEC or paediatrician is equipped to assess child development and hence undertake the recommended re-assessment. This may reflect lack of clarity between health professionals from different sectors as to each others roles and skills. In addition, the initial lack of consensus about who should make the referral to the community paediatrician in the case of a child with a red BDA may reflect the lack of familiarity with the way local services operated across sectors. Furthermore, bearing in mind that there are significant regional and sub-regional variations in the delivery of healthcare, the process for the re-assessment of a child with amber BDA needs to be locally/regionally defined, and adapted to local resource availability. A particular hallmark of child development in those with CHD is that this is an issue that spans sectors and hence may fall through the gaps, and the responses

of the Delphi survey were consistent with this, although eventually reaching consensus as to responsibilities for each sector/professional group.

Strengths and Limitations

This is the first time a consensus has been developed using the Delphi process to outline referral pathways for potentially abnormal neurodevelopment from a group of primary, secondary and tertiary care professionals looking after children with CHD. The Delphi process does not involve any face-to-face contact unlike a consensus development conference or a structured discussion. While every attempt was made to enrol experts into the Delphi panel from primary healthcare, there were few GPs and HVs who accepted the invitation. While every attempt was made to enrol experts into the Delphi panel from primary healthcare, there were few GPs and HVs who accepted the invitation. Despite this, the pattern of responses was similar across the groups. Furthermore, utilising only two virtual scenarios—one example each of amber and red BDA—may have limited the generalisability of the survey, and offering more scenarios may have generated a heterogeneous response but achieving consensus may have been difficult, and may have negatively impacted on the response rate.

CONCLUSIONS

The Delphi process has provided the initial platform for developing consensus on a national pathway for the

management of children with CHD who have either equivocal or delayed neurodevelopment. The consensus supported the vital role of the PEC as a coordinating link between the primary cardiologist and the community services. The HV as a close link to community services in liaison with the PEC was felt to be the point of contact for re-assessment of the child with borderline or equivocal results. Having developed and validated the BDA as an early recognition tool, and having established consensus for the referral pathway within primary, secondary and tertiary care sectors for a child with suspected neurodevelopmental problems based on the BDA results, the next step will be a formal healthcare evaluation of the BDA. It is hoped that referral of children in accordance of the consensus reached in this study will ensure earlier identification of neurodevelopmental problems and timely interventions to address neurodevelopmental deficits.

Twitter Aparna Hoskote @aparnahoskote

Acknowledgements We would like to acknowledge Ms Jana Gurasashvili for the administrative help with the study, Dr Roz Ullman, Senior researcher, Riches & Ullman LLP as the independent expert in Delphi methodology for her help in the initial development of the survey and all the Delphi panellists for their participation in the iterative rounds and the support from the NIHR Great Ormond Street Hospital Biomedical Research Centre, Great Ormond Street Institute of Child Health, Great Ormond Street Hospital NHS Trust, University College London, London.

Contributors Conceptualisation and study design: AH, JW, KB and ML. Administration of the survey and drafting of survey questions for the iterative rounds of Delphi: AH, JW, KB and ML. Analyses of the study data: VB, AH, JW, KB and ML. Write-up: AH, JW, KB and ML. Editing and final approval of the final manuscript: AH, JW, VB, KB and ML.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement All data relevant to the study are included in the article or uploaded as supplementary information.

Open access This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>.

ORCID ID

Aparna Hoskote <http://orcid.org/0000-0003-4760-1199>

REFERENCES

- Mackie AS, Alton GY, Dinu IA, *et al*. Clinical outcome score predicts the need for neurodevelopmental intervention after infant heart surgery. *J Thorac Cardiovasc Surg* 2013;145:1248–54.
- Hövels-Gürich HH, Konrad K, Skorzewski D, *et al*. Long-Term behavior and quality of life after corrective cardiac surgery in infancy for tetralogy of Fallot or ventricular septal defect. *Pediatr Cardiol* 2007;28:346–54.
- Miatton M, De Wolf D, François K, *et al*. Neuropsychological performance in school-aged children with surgically corrected congenital heart disease. *J Pediatr* 2007;151:78 e1.:73–8.
- Hövels-Gürich HH, Konrad K, Skorzewski D, *et al*. Long-Term neurodevelopmental outcome and exercise capacity after corrective surgery for tetralogy of Fallot or ventricular septal defect in infancy. *Ann Thorac Surg* 2006;81:958–66.
- Walker K, Holland AJA, Winlaw D, *et al*. Neurodevelopmental outcomes and surgery in neonates. *J Paediatr Child Health* 2006;42:749–51.
- Wernovsky G, Shillingford AJ, Gaynor JW. Central nervous system outcomes in children with complex congenital heart disease. *Curr Opin Cardiol* 2005;20:94–9.
- Bellinger DC, Wypij D, duPlessis AJ, *et al*. Neurodevelopmental status at eight years in children with dextro-transposition of the great arteries: the Boston circulatory arrest trial. *J Thorac Cardiovasc Surg* 2003;126:1385–96.
- Andropoulos DB, Ahmad HB, Haq T, *et al*. The association between brain injury, perioperative anesthetic exposure, and 12-month neurodevelopmental outcomes after neonatal cardiac surgery: a retrospective cohort study. *Paediatr Anaesth* 2014;24:266–74.
- Beca J, Gunn JK, Coleman L, *et al*. New white matter brain injury after infant heart surgery is associated with diagnostic group and the use of circulatory arrest. *Circulation* 2013;127:971–9.
- Andropoulos DB, Easley RB, Brady K, *et al*. Neurodevelopmental outcomes after regional cerebral perfusion with neuromonitoring for neonatal aortic arch reconstruction. *Ann Thorac Surg* 2013;95:discussion 54-5.:648–55.
- Undar A, Wang S, Guan Y, *et al*. Brain protection during pediatric cardiac surgery: a multidisciplinary team approach at Penn state hershey children's hospital and pediatric cardiovascular research center. *Artif Organs* 2012;36:A10.
- Walker K, Holland AJA, Halliday R, *et al*. Which high-risk infants should we follow-up and how should we do it? *J Paediatr Child Health* 2012;48:789–93.
- Andropoulos DB, Easley RB, Brady K, *et al*. Changing expectations for neurological outcomes after the neonatal arterial switch operation. *Ann Thorac Surg* 2012;94:discussion 55-6.:1250–6.
- Gunn JK, Beca J, Hunt RW, *et al*. Perioperative amplitude-integrated EEG and neurodevelopment in infants with congenital heart disease. *Intensive Care Med* 2012;38:1539–47.
- Wernovsky G. Neurodevelopmental outcomes: scope of the problem and current challenges. *Artif Organs* 2010;34:A1.
- Gaynor JW, Gerdes M, Nord AS, *et al*. Is cardiac diagnosis a predictor of neurodevelopmental outcome after cardiac surgery in infancy? *J Thorac Cardiovasc Surg* 2010;140:1230–7.
- Hövels-Gürich HH, Bauer SB, Schnitker R, *et al*. Long-Term outcome of speech and language in children after corrective surgery for cyanotic or acyanotic cardiac defects in infancy. *Eur J Paediatr Neurol* 2008;12:378–86.
- Marino BS, Lipkin PH, Newburger JW, *et al*. Neurodevelopmental outcomes in children with congenital heart disease: evaluation and management: a scientific statement from the American heart association. *Circulation* 2012;126:1143–72.
- Brown KPC, Ridout D, Wray J, *et al*. Selection, definition and evaluation of important early morbidities associated with paediatric cardiac surgery: a mixed methods study. *NIHR Journals Library*. In Press 2019.
- Pagel C, Brown KL, McLeod I, *et al*. Selection by a panel of clinicians and family representatives of important early morbidities associated with paediatric cardiac surgery suitable for routine monitoring using the nominal group technique and a robust voting process. *BMJ Open* 2017;7:e014743.
- Hoskote VB AU, Kakat S, Ridout, M Lakanpaul DA, *et al*. Neurodevelopmental status and follow-up in pre-school children with heart disease in London UK.. *Archives of Disease in Childhood in review* 2019.
- Wray J, Brown KL, Ridout D, *et al*. Development and preliminary testing of the brief developmental assessment: an early recognition tool for children with heart disease. *Cardiol Young* 2018;28:582–91.
- Brown KL, Ridout DA, Pagel C, *et al*. Validation of the brief developmental assessment in pre-school children with heart disease. *Cardiol Young* 2018;28:571–81.
- Management of children and young people with an acute decrease in conscious level – clinical guideline, 2019. Available: <https://www.rcpch.ac.uk/resources/management-children-young-people-acute-decrease-conscious-level-clinical-guideline>
- Management of the child with decreased conscious level – a nationally developed evidence based guideline for hospital practitioners., 2003. Available: <https://www.nottingham.ac.uk/paediatric-guideline/Tecdoc.pdf>
- The brain pathways guideline: a guideline to assist healthcare professionals in the assessment of children who may have a brain tumour, 2017. Available: https://assets.headsmart.org.uk/live/media/filer_public/8e/c3/8ec36f34-1c0d-4c3c-bc5f-706570b7af34/brain_pathways_guidelines_final.pdf [Accessed 09 January 2020].
- Murphy MK, Black NA, Lamping DL, *et al*. Consensus development methods, and their use in clinical Guideline development. *Health Technol Assess* 1998;2:1-88.:i–iv.
- Robson VK, Stopp C, Wypij D, *et al*. Longitudinal associations between neurodevelopment and psychosocial health status in



- patients with repaired D-Transposition of the great arteries. *J Pediatr* 2019;204:38–45.
- 29 Rometsch S, Greutmann M, Latal B, *et al.* Predictors of quality of life in young adults with congenital heart disease. *Eur Heart J Qual Care Clin Outcomes* 2019;5:161–8.
- 30 Kasmi L, Bonnet D, Montreuil M, *et al.* Neuropsychological and psychiatric outcomes in Dextro-Transposition of the great arteries across the lifespan: a state-of-the-art review. *Front Pediatr* 2017;5:59.
- 31 Anim C. Health visitors change lives - so why are numbers decreasing Royal College of Nursing, 2016[Report]. Available: <https://www.rcn.org.uk/news-and-events/blogs/health-visitors-change-lives>
- 32 Report.. Investment needed in specialist children's nurses. Health visitors and school nurses report shift understaffing as specialist nurse numbers drop. Royal College of Nursing; 2018.
- 33 Jozwiak G. Health Visitor numbers 'below safe level' Children & Young People Now, 2018[Report].. Available: <https://www.cypnow.co.uk/cyp/news/2006032/health-visitor-numbers-below-safe-level>
- 34 Minkovitz CS, Hughart N, Strobino D, *et al.* A practice-based intervention to enhance quality of care in the first 3 years of life: the healthy steps for young children program. *JAMA* 2003;290:3081–91.