**Infant deaths in the UK community following successful cardiac surgery - building the evidence base for optimal surveillance, a mixed methods study**

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Katherine Brown, Rodney Franklin, David Barron and David Cunningham are on the Steering Committee of the Congenital Heart Audit NCHDA.
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## Scientific Summary

### BACKGROUND

Early post-operative outcomes for children undergoing paediatric cardiac surgery have improved over the last decade, due to many small incremental adjustments to the patient journey. Furthermore, such early post-operative outcomes for paediatric cardiac operations are subject to considerable scrutiny, especially in the UK. In contrast, post discharge outcomes for infants with congenital heart disease (CHD) have to date been much less well understood, as have the performance of health services and post discharge processes that contribute to longer-term survival. The motivation for this project was to explore and understand both the risk factors for poor post discharge outcome for infants undergoing cardiac interventions and the health care processes underpinning them, and hence to make a series of recommendations for improvement, the original study questions being:

1. Can a suitable surveillance program or complex intervention be designed with the objective of decreasing mortality associated with infant cardiac surgery, by averting unexpected deaths in the community, subsequent to discharge after 'successful' surgery?
2. Can linkage of individual data from existing routine sources including both clinical and social information, from National Congenital Heart Diseases Audit (NCHDA) and Paediatric Intensive Care Audit Network (PICANET), improve our understanding of why some infants die or collapse at home following cardiac surgery?
3. Can the parents of infants with heart disease and professionals involved in post discharge care better inform the follow up and surveillance processes for infants in the community who have undergone cardiac surgery and help to identify barriers, which may be impairing their access to health care?

### METHODS

#### Systematic reviews:

The following two systematic reviews of the literature were undertaken:

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1) Unexpected deaths and unplanned readmissions in infants discharged home after major surgery for congenital anomalies: a systematic review of potential risk factors. Protocol registration (PROSPERO; CRD42013003483).
2) A systematic review of non-invasive interventions for infants discharged from hospital after major surgery for congenital anomalies. Protocol registration (PROSPERO; CRD42013003484).

**Quantitative analysis of national audit data:**

Records for all UK infants undergoing intervention for CHD between 01/01/2005 and 31/12/2010 were identified from NCHDA and linked to those individuals' intensive care admission records in PICANET. The procedure and admission based datasets from the two national audits were converted into a patient based dataset. A total of 115 children who had an excluded catheter procedure only, 765 premature babies who had ligation of patent ductus arteriosus only and 24 transplant patients were excluded from the analysis. A further 505 patients with unknown life status were removed, leaving 7976 remaining patients.

Logistic regression was used to develop risk models for:
Outcome 1: out of hospital death or death following emergency admission within 1-year following discharge,
Outcome 2: the combination of out of hospital death within 1-year following discharge and emergency readmission to intensive care ending in either survival or death.
Classification and regression tree (CART) analysis was used to identify patient groups differentiated by Outcome 2.

**Qualitative analysis:**

**Helpline staff interview (HLI)** semi structured interviews were conducted with ten congenital heart charity staff.
**Online discussion forum (OF)** 73 participants joined an online discussion forum hosted by the user group Children's Heart Federation (CHF).
**Family interviews (FI)** semi structured interviews were conducted with 20 families that had either lost an infant post discharge following paediatric cardiac surgery or had an infant readmitted to intensive care as an emergency.

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**Health professional interviews (HPI)** semi structured interviews were conducted with 25 tertiary HP and 13 primary and secondary care HP.
Qualitative analysis of these study data was performed using the Framework approach.

**Intervention development**

An expert advisory group was established to review evidence and propose interventions for improving services. It comprised professionals from three tertiary cardiac centres, representatives from primary care, secondary care and patient groups, as well as academics from the disciplines of psychology, statistics, epidemiology and operational research. Three members are trained in quality improvement methodologies. The group met on five occasions (each 2-3 hours) between March 2013 and June 2014. The suggestions for service improvement were discussed at a workshop consisting of parents that participated in the FI.
Through a facilitated process at the final meeting, the group generated a list of evidence-based interventions for future implementation or evaluation.

## RESULTS

### Systematic reviews

Despite a broad search strategy for both reviews, studies meeting inclusion criteria pertained only to patients with CHD, in particular complex single ventricle conditions. Studies were predominantly from the USA.

**Systematic review 1)**

Fifteen studies were eligible for inclusion. Risk factors identified as having a significant association with higher mortality or unplanned readmission were non-Caucasian ethnicity, lower socioeconomic status, co-morbid conditions, age at surgery, operative complexity and procedure type, and post-operative feeding difficulties.

**Systematic review 2)**

Eight studies were eligible for inclusion. The interventions of interest were home monitoring programmes (HMP). Controls were based on historic patient data in all studies. A range of clinical outcome measures (one year outcome, inter stage mortality, detection of clinical deterioration) showed improvement with HMP in different studies.

### Quantitative data

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Of 7976 patients meeting inclusion criteria, 333 (4.2%, 95% confidence interval (CI) 3.7-4.6) died within their index admission period and were excluded from our analyses, leaving a final dataset comprising 7643 infants discharged alive from their index admission for paediatric cardiac surgery. Of these, 246 (3.2%, 95% CI 2.8-3.6) experienced Outcome 1 and 514 (6.7%, 95% CI 6.2-7.3) experienced Outcome 2.

Using multiple logistic regression analysis, risk factors for death within 1-year following discharge (Outcome 1) were identified as: age at procedure, weight z-score, cardiac procedure, cardiac diagnosis, non-cardiac congenital anomaly, clinical deterioration, prematurity (<37 weeks gestation), ethnicity, and length of stay in specialist centre. When additionally including emergency readmissions to intensive care (Outcome 2), pre-procedure clinical deterioration was not significant, whilst neurodevelopmental conditions and acquired diagnoses were. Model discriminations for Outcomes 1 and 2 were very similar, with area under the receiver operating characteristic curves of 0.78, 95% CI (0.75, 0.82) and 0.78 (0.75, 0.80) respectively.

The CART analysis identified six patient groups differentiated by Outcome 2 and defined in terms of the following patient characteristics (high risk characteristics in bold):
(1) Neurodevelopmental conditions [24% Outcome 2];
(2) No neurodevelopmental conditions; low risk cardiac diagnosis (VSD/Other); congenital anomalies[†]; length of stay in specialist centre (LOS) > 1 month [24% Outcome 2];
(3) No neurodevelopmental conditions; High risk cardiac diagnosis (hypoplastic left heart syndrome (HLHS), other types of functionally univentricular heart (UVH) or pulmonary atresia (PA)) [15% Outcome 2];
(4) No neurodevelopmental conditions; low risk cardiac diagnosis (VSD/Other); no congenital anomalies; LOS > 1 month [9% Outcome 2];
(5) No neurodevelopmental conditions; low risk cardiac diagnosis (VSD/Other); congenital anomalies; LOS < 1 month [8% Outcome 2];
(6) No neurodevelopmental conditions; low risk cardiac diagnosis (VSD/Other); no congenital anomalies; LOS < 1 month [3% Outcome 2].

**Qualitative data**

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This information is presented as a synthesis of the four qualitative data sources list in methods.

*1. Training & information for families pre-discharge*

**Information overload:** It is difficult for families to understand and absorb all of the information they are given. [FI, OF]

**Poor timing:** Information is often rushed before discharge. [HPI, FI]

**Insufficient training on "Signs, symptoms, responses":** These are often missed, vague or unstructured, and no written material is given to take away. [HPI, FI]

**Barriers for non-English speakers:** There is limited access to interpreters and most resources are only available in English. [HPI]

**Some families miss out:** Limits to the availability of resources may influence the content of training and information provided. [HPI]

*2. Discharge & transferring to non-specialist services*

**Poor access to local support services:** It is difficult for specialist centres to know what local and community services are available and how to contact them, particularly where links not well established. Community teams are often short of resources. [HPI]

**Inadequate planning:** Discharge may occur at short notice and the content of a discharge package in may be strongly influenced by the availability and accessibility of local resources, leading to variation across the country in terms of who is offered what follow-up care. [HPI]

**Poor quality discharge letters/summaries:** These are often very delayed, do not reach all HPs, contain too much specialist information and terminology and often do not include: basic information; what to look out for and how to respond. [HPI]

**Ad-hoc planning for high risk patients**: In some centres there is no protocol in place for identifying high risk babies and the (extra) care that is offered to them. [HPI]

*3. Medical follow-up services*

**Problems with clinics:** Clinics are often full and running late. Outreach clinics may not incorporate paediatricians and specialist nurses. [HPI, FI]

**Inconsistent specialist support between clinics:** Many families (particularly high risk) get regular calls from cardiac nurses (CLN), but some do not and can find it hard to get in touch with them. [FI, HPI]

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**Variability & resource challenges:** There are not enough paediatricians with expertise in cardiology (PEC) and often newly trained or less experienced community nurses / health visitors (HV) attend visits. Infants must have a medical need to get a community nurse but it can be difficult to maintain regular home visits from HV, as the baby may not be considered high priority. [HPI]

**No protocol for home monitoring programmes (HMP):** Large variation between centres in the provision of HMP and the content thereof. Community professionals may not know how to respond to changes in infant condition. Some families find HMP helpful, others a distraction or too complicated. [HPI, FI]

**Feeding/weight gain:** Many families find this a very stressful aspect of care. Lack of support and conflicting advice between HP, and nasogastric tubes were cited as reasons. [FI]

*4. Non-medical support*

**Practical difficulties:** Families sometimes experience practical difficulties in the community that may not have been identified prior to discharge. These include: child care for siblings, access to transport, financial difficulties due to long hospital stays, debts, loss of earnings and inability to return to work. Some families struggle to adhere to medication regimes and can experience difficultly getting prescriptions because GPs are not always clear what has been prescribed or what to do about off-license medications [OF, HLI, FI]

**Fear & Isolation:** Parents often live in fear of an emergency and the worry of infection isolates them from other parents and support groups in their community. [FI, OF]

**Families lack confidence:** Some families lack the confidence to approach or challenge health professionals, fail to ask questions during appointments for fear of appearing ignorant or incapable, or lack the ability to articulate their concerns (particularly non-English speakers). [OF, HLI, HPI]

**The strain of 'expert parenting**' / **lack of confidence in local services:** Many families have to pass on information about their child's condition to health professionals that do not have specialist knowledge and sometimes (as the holders of knowledge) feel they are battling with local services. Many families take on an 'expert parent' role, which can be alienating and frightening. [OF, FI, HPI]

**Insufficient psychosocial support:** Support offered to families is often purely related to the medical needs of their child with no specific protocol for assessing their psychosocial needs

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and resources harder to get for social support unless they meet criteria for safeguarding. [OF, HPI, FI]

*5. Patient information*

**Poor sharing of patient information:** There are very few shared electronic patient record systems across services. Information is often relayed through the families, although there is inconsistency in the extent to which HPs use Red Books, hand-held records, health booklets etc. [HPI, FI]

**Not flagged or fast-tracked:** Often no formal system for flagging (high risk) babies or for enabling them to have quick access to services. [FI, HPI]

*6. Accessing support when a baby is sick*

**Not knowing 'Signs, symptoms, response':** Parents and all local HPs are often unclear what signs & symptoms to look for and how to respond, with insufficient guidance from specialist centres. [HPI, FI]

**Families not taken seriously:** Families sometimes find it difficult to verbalise their concerns, lack the confidence to seek help or do not feel listened to by HPs when they do. [FI, OF]

**Failing to seek specialist advice:** Sometimes local HPs fail to notify the paediatrician with expertise in cardiology or specialist centre of an incident (deterioration) or contact them when there is a concern. [HPI, FI]

**Conclusions –suggestions for health care improvement**

The following are recommended by the working group:

- All infants may benefit from a nationally standardised structured discharge document available to HPs involved in their care.
- Infants in high-risk groups, which are those with HLHS, UVH or PA, neurodevelopmental conditions and/or LOS >1 month would benefit from 'step-down' care, i.e. discharge via their local hospital.
- Home monitoring may be beneficial for all infants with a primary diagnosis of HLHS, UVH or PA.

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- All families and HP are likely to benefit from the same clear guidance on ‘what is normal’ for that child, signs & symptoms to look for, how to respond and important contact numbers, e.g. in the form of a traffic light tool.
- Anationally standardised checklist in order to plan, deliver and audit the provision of training and information for all families prior to discharge may be helpful to HP.
- Review of all post-discharge deaths of infants outside a specialist centre may be best placed at a mortality and morbidity conference held within the relevant network.
- Peer support with other families e.g. through social media or charity support groups is suggested for those being dicharged with their infant.
- The wider report provides detail of proposed metrics for processes and outcomes for use on the care quality dashboard including additional clinical outcome measures for national audit.

**Conclusions- recommendations for research**

These include:

- Further research and national consensus building is required to establish the optimal protocol, components and inclusion criteria for HMP (if beyond those proposed above), including an assessment of resource implications.
- Additional health care evaluation is required of the best format, applications and effectiveness of the proposed traffic light tool, as well as an evaluation of the proposed structured discharge document, discharge checklist and step down care. Cultural and language barriers should form part of this evaluation.
- Further research to establish the statistical and analytical steps required for routine audit of relevant outcome measures for this population, in particular post discharge mortality rates, which should incorporate adjustment for case mix.

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