# ORIGINAL ARTICLE

# Prenatal screening for major congenital heart disease: assessing performance by combining national cardiac audit with maternity data

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## ABSTRACT

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**Objective** Determine maternity hospital and lesionspecific prenatal detection rates of major congenital heart disease (mCHD) for hospitals referring prenatally and postnatally to one Congenital Cardiac Centre, and assess interhospital relative performance (relative risk, RR).

**Methods** We manually linked maternity data (3 hospitals prospectively and another 16 retrospectively) with admissions, fetal diagnostic and surgical cardiac data from one Congenital Cardiac Centre. This Centre submits verified information to National Institute for Cardiovascular Outcomes Research (NICOR-Congenital), which publishes aggregate antenatal diagnosis data from infant surgical procedures.

We included 120 198 unselected women screened prospectively over 11 years in 3 maternity hospitals (A, B, C). Hospital A: colocated with fetal medicine, proactive superintendent, on-site training, case-review and audit, hospital B: on-site training, proactive superintendent, monthly telemedicine clinics, and hospital C: sonographers supported by local obstetrician. We then studied 321 infants undergoing surgery for complete transposition (transposition of the great arteries (TGA), n=157) and isolated aortic coarctation (CoA, n=164) screened in hospitals A, B, C prospectively, and 16 hospitals retrospectively.

**Results** 385 mCHD recorded prospectively from 120 198 (3.2/1000) screened women in 3 hospitals. Interhospital relative performance (RR) in Hospital A:1.68 (1.4 to 2.0), B:0.70 (0.54 to 0.91), C:0.65 (0.5 to 0.8). Standardised prenatal detection rates (funnel plots) demonstrating inter-hospital variation across 19 hospitals for TGA (37%, 0.00 to 0.81) and CoA (34%, 0.00 to 1.06).

**Conclusions** Manually linking data sources produced hospital-specific and lesion-specific prenatal mCHD detection rates. More granular, rather than aggregate, data provides meaningful feedback to improve screening performance. Automatic maternal and infant record linkage on a national scale, requires verified, prospective maternity audit and integration of health information systems.

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**Solution** Linked

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### INTRODUCTION

In 1999, Bull's report for the British Congenital Cardiac Association documented the proportion of infants with major congenital heart disease (mCHD) undergoing surgery or intervention diagnosed prenatally.<sup>1</sup> Bull published antenatal diagnosis rates by postcode areas from unverified databases held in individual surgical centres. The Society of Cardiothoracic Surgeons established an unverified voluntary congenital database in 1977. Bull reported UK average antenatal diagnosis of 23%, with wide variation and a north-south divide. This study predated the Central Cardiac Audit Database (CCAD),<sup>2</sup> now called National Institute for Cardiovascular Outcomes Research-Congenital Audit (NICOR-Congenital).<sup>3</sup> CCAD was established in 2000, as a recommendation of the Bristol enquiry into children's heart surgery. Annually validated regional information on whether an infant undergoing surgical or transcatheter treatment for mCHD had an antenatal diagnosis (ves/no) has been available on the NICOR-Congenital public portal since 2004. Submission is mandatory for all congenital cardiac centres, but many obstetricians and regional congenital malformations registries are unaware that it records antenatal diagnosis.

NICOR-Congenital uses figures submitted within defined healthcare regions but lacks maternity hospital data, thus, hospital-specific audit cannot be generated and fed back to lead clinicians—preventing 'naming and shaming' which could drive appropriate provision of resources and training to improve performance.

Obstetric ultrasound anomaly screening is offered routinely to pregnant women between 18 and 20+6 weeks. Fetuses with suspected mCHD are referred for specialist opinion to a fetal medicine centre or cardiac unit for fetal echocardiography. Outcome measures include screen-positive confirmed cases of mCHD resulting in live birth, termination of pregnancy or spontaneous intrauterine demise. A proportion of screen-positive referrals are thought normal at specialist review, and a few found normal, postnatally, following specialist confirmation (usually coarctation or small ventricular septal defects (VSD) closing spontaneously). Screen-negative cases undergo postnatal clinical evaluation which may soon include pulse-oximetry. Fetuses screened in teaching hospitals may have prenatal detection rates >80%, but overall in England, two of three babies with mCHD are undiagnosed prenatally and the current newborn examination fails to detect one in three with lifethreatening mCHD before they leave hospital.<sup>4</sup>

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Their diagnosis is usually made following admission to an intensive care unit with presumed initial diagnosis of infection.

Poor antenatal diagnosis rates reduce the opportunity for a comprehensive fetal examination, pregnancy counselling, including genetic testing and optimal perinatal management for confirmed abnormalities by a multidisciplinary team. Failure to recognise and institute appropriate treatment for mCHD is associated with significant morbidity and mortality<sup>5</sup> <sup>6</sup> and is recognised as a major quality-of-care issue. The effectiveness of antenatal mCHD screening is one component of NHS England's congenital cardiology commissioning dashboard.<sup>7</sup> By contrast, less common congenital malformations, such as spina bifida, have a 90% predicted detection rate.<sup>8</sup>

This study links maternal and infant surgical data to inform training initiatives in two ways:

- 1. Prospectively derive hospital-specific detection rates to compare three models of prenatal congenital cardiology screening support.
- 2. Retrospectively derive and compare two lesion-specific detection rates in 19 screening hospitals (three informed by prospectively collected maternity audit).

### **METHODS**

In this study, prospective maternity screening ultrasound data of 120 198 unselected pregnant women was acquired from three maternity hospitals receiving differing models of cardiac support on referred cases from specialists working in one congenital cardiac surgical centre:

- ► Hospital A: colocated with fetal medicine unit; ready access to second opinion; proactive superintendent; received on-site training and regular audit feedback.
- ► Hospital B: received on-site training, proactive superintendent; monthly telemedicine clinics with a perinatal cardiologist.
- ► Hospital C: sonographers supported by local obstetricians with scanning expertise.

On-site training in mCHD screening provided by Tiny Tickers (http://www.tinytickers.org) was based on a five-transverse view protocol.<sup>9</sup> Hospitals A and C used obstetric sonographers throughout 2000–2011, but hospital B used research fellows after 2006, who did not provide data.

Maternity data included the outcomes of:

- ► Screen-positive mCHD, comprising liveborns, false positives, terminations of pregnancy and intrauterine demise.
- Screen-negative mCHD (false negatives) undergoing cardiac surgery and recorded for submission to NICOR-Congenital.

The place of screening was tracked by a combination of surname, postcode and expected date of delivery or date of birth linkage, and confirmed by supportive data from admissions data, transfer letters and prenatal diagnostic spreadsheets.

# ABNORMALITIES DETECTABLE ONLY BY SCREENING THE GREAT ARTERIES

The study also looked at prenatal detection of transposition of the great arteries (TGA; screening years 2000–2011) and coarctation of the aorta (CoA; screening years 2003–2011) from A, B, C and retrospectively from 16 additional hospitals. Data were tracked back from infant mCHD cases recorded in NICOR-Congenital from one congenital cardiac surgical centre. We only included cases screened in hospitals whose usual fetal referral centre was ours. These two well-defined lesions were selected to reflect screening proficiency of the outflow tracts, rather than four-chamber screening alone. We estimated the expected detection rates of malformations from each hospital's screening volume and prevalence of malformation in the population.

### Definitions

Antenatal diagnosis: data derived solely from yes/no field in NICOR-Congenital database;

Prenatal detection: rate derived by linking maternity and infant intervention data;

Undiagnosed: mCHD not detected at the fetal anomaly scan;

TGA: concordant atrioventricular and discordant ventriculoarterial connections, including small VSDs. Antenatal diagnosis only attributed where a verified abnormality of the great arteries had been detected at screening;

CoA: concordant atrioventricular and ventriculo-arterial connections, including small VSD, bicuspid aortic valve or persistent left superior caval vein. Large, associated VSDs were excluded as it is ambiguous whether CoA was considered at screening.

NICOR-Congenital define mCHD as requiring surgical or catheter intervention within the first year after delivery, following Bull.<sup>1 3</sup>

The data were anonymised before collation and analysis. Table 1 shows diagnostic categories created by combining morphology with expected postnatal management, for example shunt placement versus initial complete repair. Minor congenital heart disease (including small VSDs and mild valvar stenosis), and normal variants (including isolated right aortic arch or persistent left superior caval vein) requiring no intervention were excluded. Secundum atrial septal defects, patent foramen ovale and persistent patency of the arterial duct were classified as not detectable prenatally. Fetuses with arrhythmias but structurally normal hearts and those screened outside the national screening protocol were excluded.

### Ethics

The study proposal was discussed with local and regional ethics committees. Both decided it was an audit of practice and did not require ethical approval. The audit was registered with the Royal Brompton and Harefield Hospital NHS Foundation Trust Audit Office.

### Statistical analysis

All data analyses were conducted using Microsoft Excel (Microsoft, Redmond, Washington, USA) or SAS V9.2 (SAS Institute, Cary, North Carolina, USA). Statistical significance was set at  $\alpha \leq 0.05$ .

To assess screening performance across three maternity hospitals (A, B, C), data were stratified by hospital and Mantel– Haenszel relative risk (RR) estimates calculated for each and compared for all years. As hospital B data was only available until end of 2006, any significant differences observed over the entire 11-year period should be considered conservatively. Summary statistics describe fetal outcomes for hospitals A, B, C, and within-hospital trends were assessed using 2-year moving averages. Rates of fetal outcomes were calculated for each hospital, and Mantel–Haenszel RR estimates for live births, including and excluding terminations, with prenatal detection were calculated to allow interhospital comparison and determine prenatal detection rates of TGA and CoA.

For the 19 hospitals referring prenatally and postnatally (including A, B, C), prenatal detection rates could be estimated and compared with expected mCHD prevalence of 3.5/1000 anomaly scans, including 5% TGA and 6% isolated CoA.<sup>10</sup>

Tab	le 1 Prenatal detection a	and its in	Ifluence	on outc	ome of p	oregnanc	у								
Hospital parameters /mCHD category		1	2	3	4	5	6	7	8	9	10	Total	RR of PD (95% CI)	RR of liveborn with PD (95% CI)	RR of liveborn with PD (95% Cl terminations excluded
A	mCHD	22	40	18	16	22	5	18	55	6	6	208	1.68 (1.40 to -2.00)	0.78 (0.69 to 0.89)	1.97 (1.60 to 2.40)
	Prenatally detected	22	28	17	14	12	4	10	43	6	6	162			
	Liveborn	4	34	7	15	14	5	18	41	6	4	148			
	Termination	16	5	10	1	3	0	0	12	0	2	49			
	Intrauterine demise	2	1	1	0	5	0	0	2	0	0	11			
	Liveborn with PD	4	22	6	13	8	4	10	31	6	4	108			
	Prenatal detection(%)	100	70	94	88	55	80	56	78	100	100	78			
	(Liveborn with PD)/PD(%)	18	79	35	93	67	100	100	72	100	67	67			
В	mCHD	10	7	5	6	6	1	7	23	3	2	70	0.70 (0.54 to 0.91)	2.90 (1.63 to 5.47)	1.09 (0.93 to 1.30)
	Prenatally detected	9	2	4	4	3	0	2	8	0	1	33		,	
	Liveborn	4	6	5	6	6	1	7	21	3	2	61			
	Termination	6	0	0	0	0	0	0	1	0	0	7			
	Intrauterine demise	0	1	0	0	0	0	0	1	0	0	2			
	Liveborn with PD	3	1	4	4	3	0	2	6	0	1	24			
	Prenatal detection(%)	90	29	80	67	50	0	29	35	0	50	47			
	(Liveborn with PD)/PD(%)	33	50	100	100	100	0	100	75	0	100	73			
С	mCHD	10	22	5	6	9	3	17	29	4	2	107	0.65 (0.50 to 0.80)	2.44 (1.54 to 3.86)	1.22 (1.02 to 1.47)
-	Prenatally detected	10	10	5	0	4	1	2	12	3	2	49			
	Liveborn	4	20	3	6	8	3	17	25	3	2	91			
	Termination	6	0	2	0	1	0	0	4	1	0	14			
	Intrauterine demise	0	2	0	0	0	0	0	0	0	0	2			
	Liveborn with PD	4	8	3	0	3	1	2	8	2	2	33			
	Prenatal detection(%)	100	45	100	0	44	33	12	41	75	100	46			
	(Liveborn with PD)/PD(%)	40	80	60	0	75	100	100	67	67	100	67			

1. Hypoplastic left heart syndrome (HLHS), 2. Left heart obstruction, excluding HLHS and UVH, 3. Functionally univentricular heart, excluding HLHS (UVH) with high pulmonary blood flow, 4. Transposition of the great arteries physiology, excluding UVH, 5. Balanced atrioventricular septal defects (including 2 with aortopulmonary window), 8. Low pulmonary blood flow, including UVH, tetralogy of Fallot and pulmonary atresia, 9. Total or partial anomalous pulmonary venous connections, 10. Unclassified /miscellaneous, mCHD, major congenital heart disease; PD, prenatal detection, RR, relative risk.

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Standardised referral and prenatal detection rates with corresponding 95% CIs were calculated for each hospital for TGA and CoA and depicted in funnel plots.

#### RESULTS

# Three models of support and predicting perinatal cardiovascular support

We documented 385 pregnancies complicated by mCHD from the screened population of 120 198, equivalent to 3.2/1000 screened fetuses, indicating good case ascertainment. Figure 1 illustrates linear trends for prenatal detection of mCHD at the three hospitals with annual and summary data in table 2. There were no discernable year-on-year trends in the detection rate of all affected fetuses, including terminations and intrauterine demise. Calculated interhospital relative performance (RR) for prenatal detection of mCHD was significantly higher in A compared to B or C. Ninety-one percent (349/385) of affected pregnancies were screened at the time recommended (18-20 +6 weeks). Associated aneuploidy and extracardiac anomalies were documented (see online supplementary material). Outcome was known for all but three: two with suspected CoA moved abroad before delivery and one undiagnosed case could not be successfully tracked. The dataset contains 243 true screen-positive results and 13 false positives (10 suspected CoA), where cardiologists agreed there was probable mCHD, but postnatal scans proved normal.

Table 1 records prenatal detection and termination impacting on the predicted perinatal support requirements within grouped mCHD categories. Pregnancies in category 1 (hypoplastic left heart syndrome) were more likely to end in termination than other categories, with between 18% and 40% of prenatal diagnoses resulting in a live births. By contrast, in category 4 (TGA), most pregnancies continued. The decision to terminate a pregnancy in the non-critical categories 5 (atrioventricular septal defect) and 8 (mCHD with low pulmonary blood flow) were influenced by high rates of aneuploidy and malformations. Across the three hospitals, mothers screened in A were more likely to have a live birth following prenatal detection when terminated pregnancies were eliminated from comparison.

# Prenatal detection of TGA and CoA referring prenatally and postnatally to one centre

157/323 TGA and 164/219 CoA cases undergoing surgery during 2000-2010 and 2003-2010, respectively, fulfilled the

inclusion criteria. NICOR-Congenital data recorded 49/157 (31%) infants with antenatal diagnosis of TGA, and 61/164 (37%) infants with antenatal diagnosis of CoA. For the 19 maternity hospitals referring prenatally and postnatally, observed prenatal detection was 37% TGA and 34% CoA with wide variation (0-100%). Standardised referral rates for TGA and CoA showed only one hospital referred significantly fewer TGA cases than expected and four fewer for CoA (see online supplementary table). Only five of the 19 hospitals achieved the expected standardised prenatal detection rate for TGA, and only eight for CoA, ranging from 0.00 to 0.81 detection for TGA and 0.00 to 1.06 for CoA. Figure 2 demonstrates 2-year moving averages in the prenatal detection of TGA and CoA for these 19 hospitals demonstrating peaks but no consistent improvement, and figures 3A,B are funnel plots demonstrating interhospital performance of prenatal detection of TGA and CoA. The observed detection rate of TGA and CoA in the three models of support is shown in table 3.

### DISCUSSION

We have improved on Bull's important findings by adding maternity screening data over a 9–12-year period, to provide hospital-specific rather than aggregate audit.<sup>1</sup> Our study shows the feasibility of combining postnatal surgical data with maternity, admissions and paediatric intensive care data to produce maternity hospital-specific and lesion-specific detection rates for mCHD. NICOR-Congenital only includes infants who had an intervention, so omits terminations of pregnancy with mCHD and those dying prenatally or in a neonatal intensive care unit prior to referral or transfer, similar to data collected by Bull.<sup>1</sup>

Hospital and lesion-specific data from three hospitals comprising 120 198 unselected women (2000–2010), shows that over 90% of women received their anomaly scan at the recommended gestational age,<sup>8</sup> but prenatal detection showed no clear trends. There were no important differences in numbers of anomaly scans (table 2), nor are we aware of differing investment by the hospitals—all had received new ultrasound equipment for first trimester nuchal translucency screening.

The funnel plots in NICOR-Congenital database do not show any areas falling outside the 95% CI, nor do they demonstrate the wide variation between geographically colocated obstetric screening units in prenatal detection of mCHD who are liveborn and undergo intervention. Our funnel plots clearly show this wide interhospital variation with several hospitals having



**Figure 1** Linear trends for prenatal detection of major congenital heart disease in three screening hospitals over 11 years showing a steady and high detection rate in Hospital A, improvement in Hospital B (data only between 2000–2006), and a steady but poorer detection rate in hospital C.  $\diamond$ : Hospital A; **=**: Hospital B;  $\diamond$ : Hospital C, and linear trends for Hospital A: .....; Hospital B: ------ and Hospital C —

Hospital/year	2000	2001	2002	2003	2004	2005	2006	2007	2008	2009	201
Fetal anomaly scans	4098	4132	4601	4767	4822	4873	4906	4926	5097	5089	5094
mCHD	14	16	21	28	22	21	15	17	9	23	22
# prenatally detected	10	13	15	23	13	16	13	15	7	20	17
Prenatal detection (%)	70	81	71	82	59	76	87	88	78	87	77
Fetal anomaly scans	3880	3886	3679	4015	3998	4106	4260	-	-	-	-
mCHD	16	10	12	12	3	6	11	-	-	-	-
# prenatally detected	2	6	4	6	1	4	10				
Prenatal detection (%)	13	60	33	50	33	67	91	-	-	-	-
Fetal anomaly scans	3300	3298	3341	3402	3409	3435	3426	3957	4048	4202	4151
mCHD	7	12	2	5	10	6	9	9	13	17	17
# prenatally detected	4	6	0	2	5	3	8	1	7	5	8
Prenatal detection (%)	57	50	0	40	50	50	89	11	54	29	47

mCHD, major congenital heart disease; #, number.

zero detection of TGA and CoA over a decade (see online supplementary table). Sharing this audit with individual hospitals brought realisation of their poor performance. Current presentation of aggregate data (NICOR-Congenital) does not provide individual screening hospitals with the granularity required to understand their deficits and institute actions to improve performance.

The most recent NICOR-Congenital data shows increasing detection rates across England: 28% (2004–2008), 31% (2009–

2010), 33% (2010–2011) and 35% (2011–2012).<sup>3</sup> This has been achieved by local and regional training initiatives: the Antenatal Update published by the National Institute of Health and Clinical Excellence<sup>11</sup> recommended routine screening of Outflow Tracts and NHS Fetal Anomaly Screening Programme (FASP) commissioned hands-on, on-site training in England in 2010–2012 (after the period covered by our report).<sup>8</sup> Tiny Tickers was invited to train about 70% of England, and other regions had local trainers. Recent NICOR-Congenital antenatal



Figure 2 2-year moving averages for the detection of (A) complete transposition of the great arteries (TGA) and (B) isolated coarctation of the aorta (CoA).

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Figure 3 Funnel plots for standardised prenatal detection of (A) complete transposition of the great arteries (TGA) and (B) isolated coarctation of the aorta (CoA) with upper and lower 95% CIs (95% CI).

Table	3	Prenatal	detection of transposition of the great arteries
(TGA)	and	isolated	coarctation of the aorta (CoA) at screening in
the th	ree r	nodels o	f support

Maternity hospital	Cases	PD	Detection rate (%)	RR	Lower limit	Upper limit
CoA	2003–2011					
A	11	9	82	5.3	1.44	19.6
В	5	1	20	0.38	0.06	2.31
с	8	1	13	0.2	0.03	1.3
TGA	2000–2011					
A	9	7	78	5.19	1.73	15.58
В	6	3	50	1	0.38	2.6
С	5	0	0	0	0	0

diagnosis funnel plots suggest the north-south divide  $^{1-3}$  is not statistically apparent, although it remains evident in general health measures.  $^{12}$ 

The increase in prenatal detection rate of CoA over a decade in a single congenital cardiac centre exceeds previous single centre and regional reports of  $19\%^{6}$ <sup>10</sup> and suggests training programmes have improved diagnostic competency for this lifethreatening lesion. From knowledge of each hospital we can draw further conclusions. The prenatal detection rates in three geographically distinct maternity hospitals (A, B, C) show highest detection rates in central London (Hospital A), whereas funnel plots for CoA and TGA highlight wide differences between geographically close maternity hospitals-for example two of A's neighbouring hospitals, F and H, had almost zero detection of either lesion (figure 3A,B and see online supplementary table). The good performance of Hospitals A (colocated within a tertiary fetal medicine unit) and E is likely due to a cardiologist on staff providing easy access to expertise and second opinion, and enhanced training opportunities from the high number of mCHD referrals. Other ways of increasing expertise include extending the training of paediatricians with expertise in cardiology to include fetal cardiology to support local prenatal screening and to train a local sonographer 'champion' to ensure education and quality control as in other national programmes.<sup>13</sup>

Hospital B was the only one supported by telemedicine (requiring sonographer involvement in recording and presenting echocardiograms), as demonstrated in several specialities to provide a good method of triage, diagnosis, remote patient monitoring, staff training and opportunity for discussion.<sup>14-16</sup> Figure 1 suggests this model of support may contribute to a steady increase in mCHD detection in B without the need for frequent on-site visits, an ideal training model for more remote maternity hospitals. Hospital C had an above average mCHD detection rate of about 50%, and provision of prospective audit to this initiative may have been a major contributor. However, performance in detection of TGA and CoA was poor and highlights their need for targeted outflow tract training. Barriers to prenatal detection include inconsistent staffing, limited imaging ability and high maternal Body Mass Index.

We believe this study is an excellent demonstration of how quality of care can be improved for any clinical outcome measure using a validated national database which publishes its results on a public, web-based portal, and permits a degree of 'naming and shaming' to drive improvements. This increase in performance following the incorporation of validated 'yes/no' question into the NICOR-Congenital database has implicitly raised the standards of care for babies with mCHD, and supports the need for national comprehensive validated registries.

Others have successfully linked registries to track outcomes<sup>17</sup> <sup>18</sup> but none permit early anonymisation of data, and all depend on rigorous manual examination of data after automated links between databases have derived the initial dataset. Links to academic development and national reference tables of sociodemographic and environmental data,<sup>18</sup> may permit inclusion of developmental and socioeconomic factors, relevant to outcomes in children with mCHD.

This study highlights potential developments of NICOR-Congenital. Lack of automatic mother-to-infant linkage reduces our ability to provide granular audit and makes us reliant on local knowledge and the ability to search confidential patient information. Manual linkage is impractical on a national basis, requiring considerable skilled resources and time. Coverage by congenital malformation registries is patchy, and postmortem rates have fallen, so verified information is difficult to obtain.<sup>19</sup> Only live births feature in the UK's National Congenital Anomaly System, so information on true disease prevalence is incomplete, particularly for conditions with high prenatal detection and termination rates. No verified audit is undertaken in most hospitals, although it has been 'mandatory' in the NHS since 1989.<sup>20</sup>

Although NICOR-Congenital records childhood data reflecting ongoing pregnancies requiring interventional treatment and cannot provide a complete record of prenatal detection of mCHD, it is verified and can be used to compare temporal patterns of prenatal detection assuming similar termination rates. This information has been pivotal in highlighting poor performance publicly, and stimulating local investment in equipment and training infrastructure. Our results support this presumption with prospective evidence showing striking disparity in interhospital detection rates for CoA and TGA where neonates undiagnosed at screening are most vulnerable.

### **Study limitations**

Linking data is complex and time consuming, as the NICOR-Congenital database lacks identifiers to link mother and infant; a problem present in other non-cardiac national databases.<sup>17</sup> <sup>18</sup> Maternal-infant linkage is essential for determining site-specific data and calculating false negative screening rates. Ascertainment was hindered when the place of surgical referral was altered at delivery, but facilitated when antenatal diagnosis was made, or where perinatal collapse was recorded. Postmortem data were sought from perinatal pathology services, but few were traceable and first trimester postmortems were rare.

The small TGA and CoA sample sizes do not permit conclusions to be drawn regarding relative performance of models of care (A, B, C), but the results indicate that enhanced data collection and surveillance are vital towards improving prenatal detection.

### CONCLUSIONS

We believe this contemporaneous study using verified data for hospital and lesion-specific prenatal detection rates is a model that will enable a more comprehensive audit of outcomes than current NICOR-Congenital data alone, to facilitate better analysis and targeted improvements in fetal cardiac screening practices. We recommend the future inclusion of maternal data, such as maternal NHS number, into the NICOR-Congenital dataset to enable mother and infant linkage and tracking, for more targeted training.

### Congenital heart disease

### Key messages

### What is already known on this subject

Current prenatal screening in England fails to detect 2/3 babies born with major congenital heart disease (mCHD). National infant surgical data (NICOR-Congenital) shows prenatal detection of babies undergoing intervention for mCHD is slowly but steadily improving.

#### What this study adds

Creating maternity hospital and lesion-specific prenatal detection rates enables a more meaningful audit of outcomes. NICOR-Congenital contains no maternity data to link mother and infant, therefore, combining verified data from maternity screening with NICOR-Congenital is feasible, but has to be done manually.

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organisations that might have an interest in the submitted work in the previous 3 years. JL Gibbs and RC Franklin are immediate past and current lead clinicians for NICOR-Congenital, and HMG is founder and medical advisor to Tiny Tickers Charity.

**Ethics approval** Local ethics committee felt it was not required, as this was an audit. It was registered as an audit with Royal Brompton Hospital.

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### REFERENCES

- Bull C. Current and potential impact of fetal diagnosis on prevalence and spectrum of serious congenital heart disease at term in the UK. *Lancet* 1999;354:1242–7.
- 2 Gibbs JL, Monro JL, Cunningham D, et al. Survival after surgery or therapeutic catheterisation for congenital heart disease in children in the United Kingdom: analysis of the central cardiac audit database for 2000–1. BMJ 2004;328:611–15.
- 3 https://nicor4.nicor.org.uk/CHD/an\_paeds.nsf/vwContent/Antenatal%20Diagnosis? Opendocument (accessed 18Sep 2013).
- 4 Wren C, Reinhardt Z, Khawaja K. Twenty-year trends in diagnosis of life-threatening neonatal cardiovascular malformations. *Arch Dis Child Fetal Neonatal Ed* 2008;93: F33–5.
- 5 Boyd PA, Armstrong B, Dolk H, et al. Congenital anomaly surveillance in Englandascertainment deficiencies in the national system. BMJ 2005;330:27–9.
- 6 Brown KL, Ridout DA, Hoskote A, et al. Delayed diagnosis of congenital heart disease worsens preoperative condition and outcome of surgery in neonates. *Heart* 2006;92:1298–302.
- 7 http://www.specialisedservices.nhs.uk/info/specialised-services-quality-dashboards (accessed 15 Sep 2013).
- 8 NHS FASP, http://fetalanomaly.screening.nhs.uk/onlineresources#fileid11381 (accessed 17 Sep 2013).
- 9 Yagel S, Cohen SM, Achiron R. Examination of the fetal heart by five short-axis views: a proposed screening method for comprehensive cardiac evaluation. Ultrasound Obstet Gynecol 2001;17:367–9.
- 10 Marek J, Tomek V, Škovránek J, et al. Prenatal ultrasound screening of congenital heart disease in an unselected national population: a 21-year experience. *Heart* 2011;97:124–30.
- 11 http://www.nice.org.uk/CG062fullguideline (accessed 17 Sep 2014).
- 12 Kmietowicz Z. North-South divide in health persists in England. BMJ 2006:333;774.
- 13 Butler A. Report on the evaluation of the Regional Obstetric Ultrasound Coordinator's (ROSCO) role. October 2011. http://fetalanomaly.screening.nhs.uk/ reports#fileid11427 (accessed 17 Sep 2013).
- 14 Mistry H, Gardiner HM. Assessing the cost-effectiveness of prenatal detection for congenital heart disease using telemedicine screening. J Telemed Telecare 2013;19:190–6.
- 15 McCrossan BA, Sands AJ, Kileen T, et al. A fetal telecardiology service: patient preference and socio-economic factors. *Prenat Diagn* 2012;32:883–7.
- 16 Anker SD, Koehler F, Abraham WT. Telemedicine and remote management of patients with heart failure. *Lancet* 2011;378:731–9.
- 17 Rankin J, Silf KA, Pearce MS, et al. Congenital anomaly and childhood cancer: a population-based, record linkage study. Pediatr Blood Cancer 2008;51:608–12.
- 18 Boyd A, Golding J, Macleod J, et al. Cohort profile: the 'children of the 90s'—the index offspring of the avon longitudinal study of parents and children. Int J Epidemiol 2013;42:111–27.
- 19 Boyd PA, Tonks AM, Rankin J, et al. Monitoring the prenatal detection of structural congenital anomalies in England and Wales: register-based study. J Med Screen 2011;18:2–7.
- 20 Secretaries of State for Health, Wales, Northern Ireland and Scotland. Medical audit. Working paper 6. London: HMSO, 1989.



# Prenatal screening for major congenital heart disease: assessing performance by combining national cardiac audit with maternity data

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