

REGULAR ARTICLE

Emotional reactivity in infants with congenital heart defects: findings from a large case-cohort study in Norway

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Abstract

Aim: Advances in medical treatment in recent years have led to dramatically improved survival rates of children with severe congenital heart defects (CHD). However, very little is known about the psychological consequences for these children, particularly during and after the early period of invasive treatment. In this study, we investigated the extent to which the severity of the CHD affects the child's emotional reactivity at 6 months of age.

Method: We linked prospective data from the Norwegian Mother and Child Cohort Study, conducted by the Norwegian Institute of Public Health with a nationwide medical CHD registry and identified 212 infants with CHD in a cohort of 61 299 infants. Mothers reported on their child's emotional reactivity at age 6 months by means of a standardized questionnaire.

Results: Infants with severe to moderate CHD had 60% higher odds for severe emotional reactivity (cut-off at the 85 percentile) compared with healthy infants, after controlling for important maternal and child confounders.

Conclusion: Our study is the first to show elevated emotional reactivity in children with moderate to severe CHD, suggesting a need for special parental attention to soothe their distress. Follow-up studies will show whether this emotional reactivity is transient or an early marker of continuing emotional or behavioural problems.

INTRODUCTION

Congenital heart defects (CHD) are the most common congenital malformations and affect about 1% of all live born children (1). The clinical manifestation of CHDs varies greatly because of broad variation in severity and extent of the defect. CHDs may be life threatening and account for approximately 7% of all infant deaths (1,2). Due to advances in medical treatment (3), survival rates among CHD patients have increased dramatically in the last 10 years, and today surgical interventions very early in life are frequent (4,5).

Frequent and prolonged hospitalizations, painful surgical and medical interventions and all the discomforts brought on by the heart defect itself are just some of the stressors that these infants encounter. Whether coping with these stressors affects these infants' tendency to respond intensively (emotional reactivity) in the short and longer term is practically unknown, as recent studies on this issue are lacking. Increased rates of emotional and behavioural problems have been reported in older children and adults with CHDs, however (6–8). Presently, there is only one single study, conducted nearly 20 years ago, that documented that CHD had an effect on infant temperamental dimensions (9). That study showed that 3- to 6-month-old infants with

CHDs were more withdrawn, yet more intense, and had lower thresholds for stimulation compared with healthy infants. No influence of the severity of the CHD on the infants' emotional and behavioural problems was found.

To the best of our knowledge, this study will be the first large case-cohort study of mental health in infants with CHD. Based on extensive medical information on the children's diagnosis, we aim to determine whether emotional reactivity covaries with the severity of the CHD. Moreover, we will control for a broad range of confounders with a documented effect on the child's reactivity. Variables controlled for in the study are the child's gender (10), low birth weight (11), prematurity (12) and mother's level of education (13).

METHOD

Study design and participants

Our study has a longitudinal case-cohort design, where 212 children with CHD were identified within a large prospective epidemiological study (n = 61 299), the Norwegian Mother and Child Cohort Study (MoBa) (<http://www.fhi.no/morogbarn>). The MoBa study has been following mothers and their child from early pregnancy with repeated questionnaire assessments. Pregnant women were

invited to join the study when they received their first free ultrasound examination at more than 50 hospitals across Norway. The participation rate at first assessment was 42.7% (14). Response rates among those who consented to join the study were 95%, 92% and 87% respectively, at gestation weeks 17, 30 and 6 months postpartum (14). Mothers completed questionnaires at each assessment. In addition, we used information on the child's status at birth from the nationwide Medical Birth Registry (MBR) (15). Clinical information on the diagnosis and treatment of children with CHD was available through the nationwide CHD registry at the Department of Pediatrics, Pediatric Cardiology Unit at Rikshospitalet University Hospital, Oslo, Norway, which serves as a national tertiary centre for these children (3). The CHD registry contains information on almost all cases of significant paediatric heart disease in Norway. Every examination, diagnosis, procedure and contact with patients with CHD is entered into the database with assigned dates. To ensure the quality of the registry, only senior paediatric cardiologists are entitled to enter data. CHD was defined in this study as 'a gross structural abnormality of the heart or the intra-thoracic great vessels that is actually or potentially of functional significance (16).' Infants with CHD were then identified by matching the children's personal identification numbers in the CHD registry with those from the database of the MoBa study; 304 infants in the CHD registry were identified as participants in the MoBa study. We lost 19 infants due to attrition. Furthermore, we excluded children with significant co-morbidity and specified syndromes that are likely to affect their behaviour more strongly than the heart defect (e.g. Down's syndrome, anal atresia) (17,18). This left us with 212 infants with CHD.

Classification of the cardiac defects according to severity and treatment-related aspects was performed by two senior paediatric cardiologists (HH and LTE). The classification was based on previously accepted methods (19). The CHD patients were assigned to the following two groups:

1. 'Mild congenital heart defect' (n = 92). Children in this group are generally asymptomatic and without the need of treatment. The most common defects are hemodynamically insignificant left to right shunts and minor valvar anomalies (e.g. bicuspid aortic valve).
2. 'Moderate/severe congenital heart defect' (n = 120). This group includes children with a heart defect in need of treatment. These children are often symptomatic, and follow-up through childhood is warranted. Many are severely ill in the newborn period and are subject to early surgical treatment.

The 304 CHD infants represent 0.5% of the MoBa cohort. This percentage is somewhat lower than the expected prevalence in the normal population (1). Mild congenital heart defects account for the majority of CHDs in epidemiological studies (1). These children are not in need of treatment, and if diagnosed at hospitals other than Rikshospitalet

University Hospital, they are not consistently registered in the CHD database at Rikshospitalet.

Measures

Infant's emotional reactivity was assessed at child age 6 months by means of seven items from the Infant Characteristic Questionnaire (ICQ), fussy/difficult subscale (20). The seven items used in this study were selected on the basis of a factor analysis (21). As it is typical for large inter-disciplinary epidemiological studies, the original scale had to be shortened because of space restrictions in the questionnaire. Mothers were asked to indicate whether they agreed or disagreed with a number of assertions about their child's usual mood and temperament (see Box 1). They provided answers on 7-point Likert scales, ranging from 1 = 'totally disagree' to 7 = 'totally agree.' An average score, ranging from 1 to 7, was calculated where higher scores reflected greater infant reactivity. In view of the skewed distribution of the scale, and to identify infants with the most extreme scores, we dichotomized the emotional reactivity scale as close as practically possible to the 85th percentile. This cut-off point was used previously to identify the most extreme cases of difficult behaviour in toddlers (22). Although this cut-off point is arbitrary and not previously tested in infants, our analysis showed that varying the cut-off point or using a continuous score did not yield substantially different results. In this study, the internal consistency of the ICQ was $\alpha = 0.70$.

Box 1 Infant characteristic items

1. The child whimpers and cries a lot
2. The child is usually easy to console when he/she cries
3. The child is easily upset and begins to cry
4. The child usually screams angrily and loudly when he/she cries
5. The child demands a lot of attention
6. The child usually plays well alone when left to himself/herself
7. The child is so demanding that he/she would be a considerable problem for most parents

Confounders

Information regarding the child's birth weight, gender and gestational week at birth was retrieved from the MBR of Norway (15). The child's birth weight was defined as a continuous variable measuring the child's weight in grams. Gestational week was defined as a continuous variable measuring number of weeks from last menstruation to the week of birth.

Length of the mother's education was defined as the number of years of education including college and university studies. The mothers reported the length of their education on the questionnaire completed at the 17th week of gestation.

Statistical analyses

To investigate the relationship between severity of CHD and the degree of emotional reactivity among the

6-month-old infants, logistic regression analysis was carried out in the full sample. We adjusted for the child's birth weight, the child's gender, gestational week and length of mother's education in years. The percentage of missing values in the majority of variables was low (0.8–1.6 %). By contrast, reports on maternal education showed that 8.7% of the values were missing. To substitute the number of missing values in this variable, we performed a maximum likelihood imputation procedure using information from the highly correlated variables of father's level of education as well as the father's and the mother's incomes (23).

RESULTS

Table 1 gives a descriptive overview of the main variables used in the logistic regression analysis. The crude logistic regression analysis revealed that the 6-month-old infants with mild CHD did not show an increased probability for emotional reactivity (odds ratio 0.98, 95% CI: 0.56–1.70). Infants with moderate/severe CHD on the other hand showed a 71% increased probability for emotional reactivity (odds ratio 1.71, 95% CI: 1.13–2.59). The adjusted logistic regression analysis displayed a similar result as the crude analysis. The 6-month-old infants with mild CHD did not show an increased probability for emotional reactivity. Infants with moderate to severe CHD showed a 60% increased probability for emotional reactivity (Table 2).

DISCUSSION

The main goal of this study was to explore whether 6-month-old infants with CHD showed more emotional reactivity than healthy infants, after controlling for potential confounding variables. A further goal was to examine whether infants with more severe heart defects showed more emotional reactivity than infants with mild heart defects or no heart defects respectively.

Indeed, we found that moderate to severe CHD increased the likelihood of emotional reactivity in 6-month-old infants by 60% after controlling for a range of potential confounders. The only study with which our results can be

Table 2 Logistic regression analyses of congenital heart defect and infant emotional reactivity, adjusted for relevant confounders

Variables	Odds ratio	95% CI
Mild CHD [†]	0.88	0.48–1.58
Moderate/severe CHD [†]	1.60*	1.03–2.47
Gestational week	0.97***	0.96–0.98
Birth weight	0.94***	0.92–0.96
The child's gender [‡]	1.19***	1.14–1.25
Length of education in years [§]	0.97***	0.96–0.97

Odds ratios and 95% CI are shown; *p < 0.05, **p < 0.010, ***p < 0.001.

Reference categories: [†]Healthy children, [‡]Female.

[§]The odds ratio refers to each year increase.

compared could not document such a differential effect (9). However, our study is larger, including a case group of 92 infants with mild CHD and 120 infants with moderate to severe CHD, securing enough power to detect even slight differences in reactivity among the CHD groups. Moreover, it is likely that our sample comprised more severely ill infants, as mortality in children with severe CHD was much higher before the development of today's advanced surgical and treatment methods. Exclusion of significant co-morbidity and additional syndromes may be another explanation of our findings, as many syndromes may blur differences between mild and moderate/severe CHD (if the syndrome is present in both groups). An example of such a syndrome is velocardiofacial syndrome (22q11-deletion), which increases the risk for both CHD and several forms of mental health problems (18).

An important question is by which mechanisms CHD may influence infant emotional reactivity. First, the prolonged exposure to stressful hospital care that moderate/severe CHD brings with it may put the infant in a state of chronic stress, which in turn provokes emotional distress and crying (24). Stress may also affect the parents, resulting in negative circuits of interaction. Another possible mechanism for these behavioural deviations may be physiological effects of the heart disease or its treatment on the brain, e.g. oxygen deprivation pre-, peri- or postoperatively. Impaired oxygen delivery to the brain was found previously to have an impact on behaviour problems in the young (25). A third possibility is that a common genetic link disposes for both CHD and emotional reactivity. No such mechanism has been found with respect to CHD but has been found in a related syndrome (18).

The stability of the emotional reactivity measured at 6 months of age raises another question. Mapping of developmental trajectories through childhood could be important to get a complete picture of the effects of CHD on child behaviour. In this context, the timing of assessments is important, because infants and toddlers display very different behaviours at different ages that are linked by complex developmental pathways (26). Whether the association between emotional reactivity in infants with CHD increase or decrease with age is difficult to answer. Both scenarios are possible. Studies on what predicts a specific behavioural development in infants with CHD will be important to target the best strategies for intervention.

Table 1 Characteristics of the Cohort

	Control (n = 61 299)	Mild CHD (n = 92)	Moderate/ Severe CHD (n = 120)
The child's gender			
Boys	31251 (51)	36 (39.1)	73 (61.3)
Girls	30046 (49)	56 (60.9)	46 (38.7)
Gestational week M ± SD	39.40 ± 1.89	38.73 ± 2.92	38.69 ± 2.51
Birth weight in grams M ± SD	3586 ± 586	3592 ± 828	3338 ± 827
Infant emotional reactivity ≥85th percentile	10198 (16.7)	15 (16.3)	30 (25.4)
Length of education in years M ± SD	14.47 ± 2.51	14.40 ± 2.45	14.50 ± 2.40

Case numbers and within group percentages in parentheses are shown.

Our study has some limitations that must be taken in to consideration when interpreting the results. First, we had access to maternal reports of the infant behaviour only. It would have been desirable to have a professional rating of the infant behaviour, as psychological factors in mothers may impact their responses (27). Another limitation lies in the use of a short version to map infant behaviour. A more comprehensive measure would possibly reveal stronger effects than what we have found.

Clinical implications

Signs of emotional distress in infants with moderate to severe CHD deserve the clinician's attention. Parents should be informed early that their infant may show signs of emotional distress and be in need of more comforting than healthy infants. Moreover having parents check off a short questionnaire on the child's emotional reactivity and soothability or enquiring about such symptoms during follow-up consultations could open up opportunities for detecting infants at risk for continued emotional problems. In such cases, early interventions may be key to obtain better psychological treatment outcomes (28).

CONCLUSION

Our findings revealed an increased probability of emotional reactivity already at 6 months of age in infants with moderate/severe CHD after controlling for a range of potential confounders. Infants with mild forms of CHD showed no increased probability of emotional reactivity at 6 months of age. This information may be useful in the counselling of parents of children with CHD and in the planning of supportive care.

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