

Original Article

A multicentric study of disease-related stress, and perceived vulnerability, in parents of children with congenital cardiac disease

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Abstract Parents of children with congenitally malformed hearts can suffer from stress as a result of the medical condition of their child. In this cross-sectional study, we aimed to describe levels of parental stress, and perceived vulnerability, in parents of children who underwent major cardiac surgery, by using both generic and disease-related measures for assessment. We included parents of children who underwent open-heart surgery over the period 2002 through 2007 in the Center for congenital Anomalies Heart Amsterdam/Leiden, abbreviated to provide the acronym CAHAL. In total, we assessed 114 mothers and 82 fathers of 131 children, using the Pediatric Inventory for Parents, short form, General Health Questionnaire, Parental Stress Index-Short Form, State-Trait Anxiety Index and the Child Vulnerability Scale. Compared to the reference groups of the instruments used, parents of children with congenitally malformed hearts did not report higher generic nor disease-related stress scores, and parenting levels of stress were also comparable to reference groups. State anxiety levels, however, were higher in mothers of children with congenitally malformed hearts. Both fathers and mothers reported significantly higher rates of perceived vulnerability than did parents of healthy children. Risk factors for increased anxiety and perceived vulnerability were found in the number of surgical procedures, the time past since the last procedure, and ethnicity. Severity of the lesion did not influence parental levels of stress, but parents of children with hypoplastic left heart syndrome did report higher levels of stress than other parents. Psychosocial screening of parents of children with congenitally malformed hearts is important in order to provide appropriate counselling to those parents most in need.

Keywords: congenitally malformed hearts; psychological problems; overprotection; children

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MAJOR ADVANCES IN SURGICAL INTERVENTIONS for patients with congenitally malformed hearts have led to increasing rates of survival¹. Neurological functioning, health, and

quality of life of these children can be compromised.² As a result, parents of children with chronic health problems are at risk for experiencing ongoing stress³. Parents report feeling less competent in their parenting ability, to be more permissive in their style of parenting,^{4,5} and to have lower expectations for their children.

Parents of children with congenitally malformed hearts have also been found to report more

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psychological problems than parents of healthy children, such as higher levels of depression and anxiety,⁶ higher rates of posttraumatic stress disorder,⁷ and lower scores relating to quality of life, especially in the area of personal development.^{8,9} Parents worry about the future of their offspring,⁴ report feelings of distress and hopelessness,^{3,10} and experience financial, familial, social, and personal strain.¹¹ More than one third of the mothers of children with congenitally malformed hearts meet criteria of poor adjustment.¹² Risk factors for poor adjustment are the frequent use of palliative strategies for coping, such as self-blaming and avoiding emotions, stress stemming from daily hassles¹² and being a mother,⁶ or belonging to an ethnic minority.⁷ The behavioural difficulties of the children are related to parental levels of stress,³ and the parental burden of providing care. Dissatisfaction with care, and social isolation, have also been associated with an increased risk of long-standing psychosocial morbidity in a recent longitudinal study.¹⁵

Perceived social support can serve as a resilience factor between family stress and family and parental coping.¹⁴ The severity of the congenital cardiac lesion has been shown not to predict psychosocial functioning of parents over time in numerous studies,^{5,12,13} but one study⁴ did show that parents of children with hypoplastic left heart syndrome reported more negative impact of the illness on the family, and more parenting stress, than did parents of children with transposed arterial trunks.

Some articles report less parenting stress than the normative population,¹⁵ and lower levels of distress than reference groups, at least 7 years after surgery.¹⁶ Whether these findings were due to the desire to 'leave things in the past', the stable condition of the majority of the children, or denial¹⁷ remains unsure. Timing of assessment matters. Close to the surgical procedures, levels of stress proved to be high for many parents, but a significant reduction in the level of distress was found 12 months after surgery.^{18,19}

In our present study, therefore, we aimed, first, to assess whether levels of generic and disease related stress, anxiety and parenting stress of parents of congenitally malformed hearts were higher than of parents of healthy children, second, to assess perceptions of the vulnerability of the children in this group of parents, and third, to determine potential predictors of parental and parenting stress and parental perceptions of vulnerability of their child.

Method

Participants

Parents of children who underwent major surgery for congenitally malformed hearts, recruited in

three academic medical hospitals cooperating with one group of congenital cardiac surgeons were eligible for this study. We randomly selected parents of 300 children who underwent open-heart surgery in the years 2002 through 2007. We invited parents of 50 children, randomly selected from each year, to participate.

Procedure

The study was approved by the Medical Ethics Committees of the three medical centers. Eligible parents received information about the study and an informed consent form. The parents who agreed to participate received a login number and password to fill in the questionnaires through the Internet on a specially designed, secured website. Parents were requested to fill in the questionnaires separately from each other, and not to consult each other.

Measures

The *Pediatric Inventory for Parents*, in its short form, is a 15-item self-reporting questionnaire, derived from the full *Pediatric Inventory for Parents*, developed by Randi Streisand,²⁰ that measures parental stress related to the serious illness of their child with respect to, first, communication with the child and the medical team, second, emotional distress, third, medical care, and fourth, role function. Each of the 15 items is rated on two 5-point Likert-type scales. Parents need to respond to the items twice. The first scale assesses the frequency of each stressor, while the second scale assesses how difficult the issue has been for the parent. Parents are asked to consider last week when responding to each item. Adequate internal consistency ($\alpha = .80-.96$) and construct validity of the original version of the measure have been reported, and total scores have been found to correlate significantly with a general non-illness specific measure of state anxiety and parenting stress.²⁰

The *Pediatric Inventory for Parents-Short Form* was developed by two of the authors, specifically C.M.J. Vrijmoet-Wiersma and H.M. Koopman, and consists of the 15 items of the original questionnaire with the highest item-total correlations for both frequency and difficulty, and with the highest clinical relevance. The *Pediatric Inventory for Parents Short Form* correlated highly with both total frequency and total difficulty of the full Dutch *Pediatric Inventory for Parents*, scoring at .95 and .94 respectively, in our sample. Internal consistency of the Short Form in our sample was .96.

The *General Health Questionnaire*, 12-item version, is a self-report measure of psychological symptoms that can be used as a general measure for psychological distress. The psychometric properties

of the Dutch version of the scale are reported to be satisfactory,²¹ and the questionnaire has been used frequently in national and international studies.^{22,23} The cut-off score of the General Health Questionnaire is 2, meaning a total score of 0 or 1 is interpreted as no psychological morbidity, and a score of 2 or higher is interpreted as possible psychopathology. The alpha value in this study is .92.

The *State Trait Anxiety Index, state and trait version* is a 40-item questionnaire that measures both the transitory emotional condition of stress, representing the state, and the general inclination towards anxiety, or the trait, of the respondents. Dutch norm group data are available,²⁴ as well as information about reliability and validity. The alpha value in this study is .80 for state anxiety, and .77 for trait anxiety.

The *Parental Stress Index, Short Form, Dutch version* measures parenting stress associated with raising children in general.²⁵ The short form is derived from the full 123-item Dutch Parental Stress Index. It is a reliable and valid measure, and contains 25 items that are scored on a five-point continuum from strongly agree to strongly disagree. The form differentiates well between clinical and non-clinical groups, and has been used in various studies.²⁶ The alpha value in this study is .93.

The *Child Vulnerability Scale*²⁷ is an instrument used to identify parental perceptions of child vulnerability. The scale is an 8 item self-report measure with a 4-point response scale ranging from definitely false to definitely true. The Dutch version of the scale²⁸ has good reliability and validity, albeit that the results of the validating study have yet to be published. For this reason, we used the American reference group for comparisons.²⁷ The internal consistency of the measure in the present study is .79.

Demographic and clinical information

We recorded the gender, age, marital state, ethnicity, and educational level of the parent, as well as the gender and age of the target child. Furthermore, the medical diagnosis, current state of treatment, number of surgical procedures, and the number of weeks since the last procedure, were traced from the medical files.

Analyses

Differences between responders and non-responders and differences between parental scores and reference groups were calculated through independent T-tests. Possible differences concerning time since the last procedure, and gender and ethnicity of the parents, were determined, along with the extent to which the number of surgical procedures affected

parental stress scores and perceived vulnerability. ANOVA were used to assess differences in levels of stress between parents of children in different categories of disease. Forced entered regression analysis was performed to determine the predictors of perceived vulnerability and parenting stress.

Results

Participants

Of all 300 families who were included in our study, 11 could not be traced, and 4 children had died. A total of 285 eligible families were invited to participate, and 231 parents of 152 children filled in the questionnaires. After examining the data, 35 parents were excluded because their questionnaires contained only demographic data.

A total number of 196 parents of 132 children, made up of 64 children of whom both parents, and 68 children of whom one parent participated, took part in this study. The low response rate, at 34%, was primarily due to the fact that the illness and treatment of the children was perceived to be too long ago, to inconvenience associated with completing questionnaires, to problems with language, and because some parents were too busy with work. Non-participating parents did not differ with regard to time since the last procedure, number of surgical procedures, or gender and age of the child. The percentage of parents with lower education, however, and those of non-Dutch origin, was higher. Table 1 provides a detailed description of the cohort of children.

Parental stress and anxiety and parenting stress

Parents of children with congenitally malformed hearts scored in a fashion comparable to parents of children with cancer²⁹ on the disease-related questionnaire. Items on the Pediatric Inventory for Parents-Short Form with the highest perceived difficulty scores were: 'Feeling scared that my child could get very sick or die', with 39% of all parents answering that they find this very difficult or extremely difficult, 'Feeling helpless over my child's condition', answered by 28%, 'Seeing my child sad or scared', answered by 24%, 'Feeling uncertain about the future', answered by 23%, and 'Being with my child during medical procedures', answered by 22%.

Both fathers and mothers of children with congenitally malformed hearts provided scores comparable to the instrument norm groups of the generic stress questionnaire in the General Health Questionnaire. Of all parents, 7% scored higher than the cut-off point of the General Health Questionnaire. This score is significantly lower than the percentage in the Dutch reference group.²¹

Table 1. Characteristics of the cohort.

Characteristics of the 132 children	Mean (SD)	Range
Age at assessment (months)	45.3 (20.6)	10–97
	N	%
Sex		
Male	75	57
Female	56	43
Time since last operation (months)	33 (24.2)	2–80
Number of operations	2.1 (1.7)	1–13
Underlying heart disease		
Ventricular septal defect	29	22
Transposition	28	21
Hypoplastic left heart syndrome	21	16
Tetralogy of Fallot	20	15
Atrioventricular septal defect	8	6
Common arterial trunk	5	4
Aortic stenosis	4	3
Pulmonary stenosis	4	3
Totally anomalous PVC	2	1
Atrial septal defect	1	.07
Miscellaneous lesions	10	8
Parent characteristics (N = 196)	Mean (SD)	Range
Age (years)	35.9 (4.5)	25–52
	N	%
Sex		
Male	82	42
Female	114	58
Paternal educational level		
Lower*	16	21
Middle**	27	33
Higher***	38	46
Maternal educational level		
Lower*	19	17
Middle**	36	33
Higher***	57	50
Ethnicity		
Dutch	183	93
Non-Dutch	13	7

PVC – pulmonary venous connection.

*Lower = primary school, elementary education or secondary school for lower general education \approx O level.

**Middle = senior general secondary education, pre- university education (higher secondary education) \approx A level or intermediate vocational education.

***Higher = higher professional education or university education.

When comparing state and trait anxiety scores of the parents of children with congenitally malformed hearts group to the Dutch reference group²⁴, no significant differences were found for fathers. State anxiety scores of mothers of those with congenitally malformed hearts, however, were significantly higher than those of mothers in the reference group, as shown in Table 2 ($p < .05$). Parenting stress levels as shown using the Parenting Stress Index-Short Form were comparable to the reference groups.

Perceived child vulnerability

Compared to the reference group of Dutch parents of healthy children²⁸, both fathers and mothers scored significantly higher on the Child Vulnerability Scale ($p < .001$ for mothers and fathers). Of all parents, 21% scored above the cut-off point of 10, compared to 10.1% of the parents in the reference group²⁷ (see Table 2).

Correlates of parental stress

Demographic and disease-related characteristics such as time since the last procedure, the number of procedures, and gender and ethnicity of the parent, significantly affected parental scores for stress. Parents of children who underwent surgery more than 3 years ago reported lower scores on all measures that were used (all $p < 0.05$) than parents whose child underwent surgery less than 3 years ago. Parents of children who underwent two or more open-heart procedures reported higher state anxiety levels than parents of children who had only a single procedure ($p < .05$). Mothers reported higher generic stress scores ($p < .01$), parenting stress scores ($p < .01$), and perceived vulnerability ($p < .007$) than did fathers. Non-Dutch parents reported higher perceived vulnerability scores than Dutch parents ($p < .001$).

Severity

We compared stress levels of parents of children whose heart was completely restored after surgery and who have minimal restrictions, considered as healthy, with parents of children who still suffer from mild restrictions, and a third category of parents of children deemed to have severe restrictions. We found no differences on any of the instruments used to measure parental stress or perceived vulnerability (see Table 3).

Examination on a more detailed level showed that parents of children with hypoplastic left heart syndrome reported higher levels of disease related stress ($p < 0.05$) on the disease-related stress and lower levels of trait anxiety ($p < .02$) on trait anxiety than parents of children with ventricular septal defect. Comparison with parents whose child was born with transposition showed that parents of children with hypoplastic left heart syndrome scored higher on generic stress ($p < .03$). We found no differences between parents of children with tetralogy of Fallot and parents of children with hypoplastic left heart syndrome and children with ventricular septal defect.

Predictors of perceived vulnerability and parenting stress

A forced entered regression analysis was performed to determine which factors are predictors of heightened perceived vulnerability. We found that

Table 2. Comparison of parents of children with congenitally malformed hearts to reference groups.

	114 Mothers of children with congenitally malformed hearts M (SD)	82 Fathers of children with congenitally malformed hearts M (SD)	Reference group Mothers M (SD)	Reference group Fathers M (SD)
PIP-SF total	55.9 (19.9)	51.9 (16.3)	54.4 (19.3)	48.5 (16.4)
GHQ	1.9 (2.8) ^a	1.1 (1.6)	2.2 (.5)	1.6 (.5)
STAI state	53.9* (9.6)	44.0 (8.2)	38.8 (13.2)	36.4 (10.3)
STAI trait	46.6 (6.6)	44.1 (7.8)	39.4 (11.2)	37.3 (10.3)
PSI-SF	52.3 (23.6) ^a	45.9 (15.4)	54.4 (19.3)	48.5 (16.4)
CVS	7.1 (5.1)** ^a	5.9 (4.1)**	2.2 (2.2)	2.1 (2.2)

PIP-SF = Pediatric Inventory for Parents, short form; GHQ = General Health Questionnaire; STAI = State Trait Anxiety Index; PSI-SF = Parenting Stress Index, short form; CVS = Child Vulnerability Scale.

*Significant difference between parents of children with congenitally malformed hearts and reference group ($p < .05$).

**($p < .001$).

^aSignificant difference between mothers and fathers.

Table 3. Parental stress scores classified by health restrictions and diagnosis.

	PIP-SF M (SD)	GHQ M (SD)	STAI state M (SD)	STAI trait M (SD)	PSI-SF M (SD)	CVS M (SD)
'Minimal' (N = 69)	55.5 (22.8)	2.7 (.3)	44.5 (7.5)	46.1 (6.7)	53.2 (22.4)	6.9 (.6)
'Mild' (N = 28)	58.3 (20.1)	1.4 (.5)	45.8 (7.2)	46.6 (5.7)	51.6 (21.9)	7.8 (.8)
'Severe' (N = 26)	59.5 (16.7)	2.2 (2.9)	46.9 (7.8)	45.6 (5.7)	48.4 (17.7)	4.4 (.9)
HLHS (N = 21)	62.9 (14.5) ^a	2.5 (3.0) ^b	45.9 (8.3)	44.6 (10.0) ^a	49.5 (15.5)	5.3 (3.8)
VSD (N = 29)	53.8 (25.5)	1.9 (2.9)	44.8 (9.1)	47.4 (6.5)	59.5 (26.5)	7.0 (5.3)
TGA (N = 28)	54.5 (21.2)	1.3 (2.0)	44.1 (5.4)	45.0 (5.8)	50.0 (18.1)	6.0 (5.2)
Fallot (N = 20)	57.6 (18.9)	2.5 (3.0)	44.9 (7.9)	46.1 (6.4)	56.2 (23.3)	8.0 (4.7)

'Minimal': those with children have ventricular septal defects (VSD), atrial septal defects, pulmonary and aortic stenosis, transposition (TGA), and totally anomalous pulmonary venous connection; 'Mild': those with children having atrioventricular septal defect and tetralogy of Fallot; 'Severe': those with children having hypoplastic left heart syndrome (HLHS) and common arterial trunk.

PIP-SF = Pediatric Inventory for Parents, short form; GHQ = General Health Questionnaire; STAI = State Trait Anxiety Index;

PSI-SF = Parenting Stress Index, short form; CVS = Child Vulnerability Scale.

^aSignificant difference between HLHS and VSD ($p < .05$).

^bSignificant difference between HLHS and TGA ($p < .05$).

disease-related parental stress and parenting stress combined accounted for a total of 26% of the variance in perceived vulnerability in both mothers and fathers. Forced entered regression analyses revealed that perceived vulnerability accounted for 14% of parenting stress in mothers and 16% in fathers. Time since the surgical procedure, and the number of procedures, did not add to the explained variance.

Discussion

The results of our study showed that levels of distress in parents of children with congenitally malformed hearts were largely comparable to reference groups, with the exception of state anxiety, which was higher in mothers of children with congenitally malformed hearts. This finding is in line with a comparable study on parents of children who underwent surgery at least 7 years ago¹⁶ and with another study¹⁵ reporting lower rates of parent-related stress than parents of healthy children

and comparable levels of child-related stress. Our findings, however, are not consistent with other studies⁵ using the Parenting Stress Index-Short Form, in which significantly higher parenting stress compared to the normative sample was reported. This difference in results is difficult to interpret.

Rates of perceived vulnerability were significantly higher in parents of congenitally malformed hearts than in parents of healthy children, regardless of time since surgery or severity of the disease. Rates of parenting stress and disease-related stress combined predicted levels of perceived vulnerability in our sample. High perceived vulnerability can lead to the so-called vulnerable child syndrome,⁴ which develops when parents have been in a situation in which they almost lost their child. This attitude can lead to overprotective parenting, excessive parental concerns, and overindulgence. As a result, the child can become overly dependent, disobedient, irritable, argumentative, and uncooperative. Separation anxiety and underachievement are often also seen in these children.^{30,31} Perceived vulnerability may

explain the heightened vigilance,³² and the finding that mothers of young children with congenitally malformed hearts find it difficult to determine what is normal for their child.³ Parents of children with congenitally malformed hearts are more insecure about what to expect from their child, and about the amount of freedom they can allow their child to discover the world, because they may still carry an unconscious fear of losing their child, even if their doctors will give normalization advice, such as telling them that they should treat their child as any other child.³² Future research should aim to study the effect of heightened perceived vulnerability on the health related quality of life and self-esteem of the children.

In line with other studies,^{6,7} non-Dutch parents reported significantly higher stress levels than did Dutch parents, which may be due to the availability of fewer resources,⁷ or problems with communication. Severity of the malformation did not seem to play an important role in parental stress and well-being, although we did find that parents of children with hypoplastic left heart syndrome reported higher levels of stress than the other parents of children with congenitally malformed hearts, but, surprisingly, lower anxiety levels than parents of children with ventricular septal defect. It appears that the perception of the illness suffered by their children is far more important than the objective clinical severity^{6,33}.

Strong points of our study are that we were able to include a large number of fathers, and that we used a combination of generic and disease related questionnaires. Furthermore, we managed to build and use a secured website to administer the questionnaires. Web-based assessment has the advantages of decreased experimenter demand and social desirability effects, reduced missing data, avoidance of errors in entering data, and savings of money and time³⁴.

Despite the satisfactory results of the present study, there are some limitations that need mentioning. First of all, the response rate of the study was unexpectedly low. Assessment through the Internet has very likely affected rates of response. A possible explanation could be that some parents may not have Internet access, or may not feel confident enough in using it. One could also hypothesize that a paper-and-pencil version in the form of a booklet could serve as a tangible reminder to parents to fill in the questionnaire, whereas a letter with a login code and password could easily get out of sight and out of mind. Other research that assessed participants through the Internet showed inconclusive results with regards to differences in response rates between the Internet and paper-and-pencil versions. A study that did not incorporate a reminder procedure found a large

difference between the Internet group, scoring at 19%, and paper-and-pencil group, achieving 72%, whereas studies that did remind the participants found more equal rates of response in the two groups³⁵.

Furthermore, many parents, when reminded by telephone to fill in the questionnaires, mentioned to us that the surgery had been so long ago that they were wondering if their answers would be relevant for our study. This attitude could have led to over-reporting of parental levels of stress, if more parents with high levels participated than those with low levels. On the other hand, non-responders could also mainly have been parents with many concerns who did not want to be reminded of the surgical procedures, possibly because of posttraumatic stress symptoms such as avoidance or denial, as has been suggested by Spijkerboer et al.¹⁶ Possible denial could have led to underreporting of parental levels of stress.

We had an overrepresentation of parents with higher education, and an underrepresentation of non-Dutch parents, which limits the generalization of our results. It is possible that more of the non-Dutch parents did not have Internet access, while others might have had language problems. Lastly, a more general question that should be asked is whether the assessment mode generally used by paediatric psychologists, that is self-report questionnaires, is adequate for parents from other cultural backgrounds, or if should we make more use of qualitative measures, such as semi-structured interviews or narrative techniques.

To conclude, parental stress, 2 to 6 years after their children have undergone major cardiac surgery, is comparable to reference groups, with the exception of higher levels of anxiety in mothers of children with congenitally malformed hearts. The perceived vulnerability, nonetheless, is very high in this group of parents, and can lead to overprotective parenting and behavioural problems in the children. Parental stress and parenting issues need to be assessed at follow-up visits, in order to provide adequate support, information, and psychological education to these parents.

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