Psychosocial resilience in parents of children with congenital anomalies: A prospective longitudinal study.

Master's thesis

Author: A.W. (Suzanne) van Eijk
E-mail: aw.vaneijk@gmail.com

Student number: 3159191
Date: 21 June 2011

Master of Clinical and Health Psychology, Utrecht University.

Supported by:

Dr. P. Honig-Mazer
Erasmus MC – Sophia children's hospital,
Department of Pediatric Surgery.

Dr. S. Doosje
Utrecht University,
Department of Clinical and Health Psychology.

Universiteit Utrecht

Table of contents

Preface 3

Abstract (Nederlands) 4

Abstract (English) 5

Introduction 6

Methods 9

Setting 9

Participants 9

Design 10

Procedure 10

Instruments 11

Statistical analyses 12

Results 12

General characteristics 12

Parenting stress 15

General health status 15

Scores at clinical range on PSI-SF and SF-36 16

Discussion 17

References 20

Preface

This study is part of my master Clinical and Health Psychology at Utrecht University. I conducted this study on the pediatric surgical department of the Erasmus MC – Sophia children's hospital, in the surgical long-term follow-up team. Working on this study was a process of changing from student to scientist: I have experienced all aspects of doing a study and writing a scientific article. Moreover, I have learned about functioning in a professional multidisciplinary team. I have noticed that sometimes it is not possible to function without the occasional suggestions of my colleagues. In my view, cooperation with colleagues of the team functioned very well. I have gained a lot of skills in this cooperation, not only regarding research skills but also on social intercourse and practical matters. I really enjoyed doing this study. I have even been dreaming about this study for several nights. In general, I considered it a positive way of spending my time.

In the first place I want show my gratitude to Petra Honig-Mazer, my supervisor at the surgical long-term follow-up team of the Erasmus MC – Sophia children's hospital. She has supported me during the whole process of doing this study and she has made the first moves in the direction of this study during her doctoral research. Her way of supporting was very motivating for me and I felt myself appreciated and professionally supported by her. I always had the chance to ask when I was feeling uncertain about my work. Furthermore, I want to thank Sibe Doosje, my supervisor at the department of Clinical and Health Psychology, Utrecht University. He gave me space to accomplish my 'own' study and gave me a high extent of independence. However, I could always contact him for support. This amount of independence gave me the autonomy to develop myself in a pleasant manner. Finally, I want express my appreciation to the whole surgical long-term follow-up team for their support in several ways. It was a pleasant cooperation; I could always ask them for help. In particular, I want to thank Kim Gevers for assisting me during the data collection phase. She listed a lot of questionnaires for my study and helped me to find structure the data

The theme of the study, psychosocial resilience (Masten, 2001), fascinates me because until recently strength-focusing perspectives were not frequently used in scientific literature on the field of psychology. That is, until recently most studies were mainly focused on weaknesses or psychopathological symptoms instead of strengths of people. Through the implementation of strength-focusing perspectives parents can be trained or counselled to prevent negative psychosocial effects instead of treating them afterwards (Gudmundsdottir, Schirren, & Boman, 2011). I hope you share my fascination, strengthening the strength-focusing perspective in scientific literature. If you have any questions or comments, please feel free to contact me.

Abstract (Nederlands)

Achtergrond: Het doel van deze studie was het onderzoeken van de effecten van het hebben van een kind met een aangeboren afwijking 12 maanden, 24 maanden en 5 jaar na de geboorte op de psychoscociale gemoedstoestand van ouders op de lange termijn. Mogelijke invloeden van de soort aangeboren afwijking op deze verbanden worden beschreven.

Methoden: 12 maanden, 24 maanden en 5 jaar na de geboorte, zijn 156 moeders en 149 vaders van 157 kinderen met een ernstige aangeboren afwijking gevraagd om de Parental Stress Index – verkorte vorm en de SF-36 in te vullen. De mediaan van de leeftijd van de moeders bij de geboorte van hun kind was 31 jaar en dat van de vaders was 33 jaar. Twee overkoepelende schalen kunnen afgeleid worden van de SF-36: lichamelijke gezondheid en mentale gezondheid. De scores zijn vergeleken met Nederlandse normgroepen. Veranderingen over tijd en consensus tussen ouders zijn gemeten.

Resultaten: Alleen 12 maanden na de geboorte rapporteerden moeders meer mentale problemen dan de normgroep. Bij 24 maanden ervoeren vaders zelfs minder mentale problemen. Bij 5 jaar rapporteerden de moeders minder stress gerelateerd aan opvoeding. Andere resultaten met betrekking tot mentale en lichamelijke gezondheid en stress gerelateerd aan opvoeding verschilden niet significant met de populatienormen. Bij 12 en 24 maanden ervoeren moeders significant meer mentale problemen dan vaders. Bij 5 jaar zijn geen significante verschillen gevonden tussen vaders en moeders. Stress gerelateerd aan opvoeding nam bij vaders significant af over de tijd. Bij moeders was de afname in stress gerelateerd aan opvoeding niet significant. Gezondheidsproblemen namen niet significant af over de tijd.

Conclusie: De resultaten van deze studie laten zien dat ouders van kinderen met aangeboren afwijkingen op de lange termijn in staat zijn om om te gaan met het feit dat zij een ernstig ziek kind hebben. De resultaten van deze studie bevestigen het bestaan van veerkracht bij ouders van kinderen met een aangeboren afwijking op de lange termijn. De resultaten van deze studie bevestigen ook de suggestie van verscheidene experts op het gebied van chronische ziektes in de kindertijd, dat variatie in de impact op families meer gerelateerd zouden zijn aan de kenmerken van de conditie in plaats van aan de diagnose alleen.

Abstract (English)

Background: The purpose of this study was to assess long-term parental psychosocial outcome, to measure the impact of having a child with congenital anomalies (CA) 12 months, 24 months and 5 years after birth of the child. Possible influences of the kind of CA on these relations are described.

Methods: At 12 months, 24 months and 5 years after birth, 156 mothers and 149 fathers of 157 children with severe CA were asked to complete the Parental Stress Index – short form and the SF-36. The median age of the mothers at delivery was 31 years; that of the father 33 years. Two summary measures can be derived from the SF-36: physical health and mental health. Scores are compared with Dutch norm groups and changes over time are measured. Consensus between parents was tested.

Results: Only at 12 months after birth, mothers perceived more mental problems than the norm group. At 24 months, fathers even perceived less mental problems. At 5 years mothers experienced less parenting stress than the norms. Other results on mental and physical health and parenting stress did not differ significantly from population norms. At 12 and 24 months, mothers experienced significantly more mental problems than fathers. At 5 years, no significant differences between mothers and fathers were found. Parenting stress declined significantly over time in fathers. In mothers the decline in parenting stress was not significant. Problems concerning health did not decline significantly over time.

Conclusion: The results of this study indicate that parents of children with CA are capable of coping with a situation of having a severely ill child on the long term. The results of this study support the notion of resilience in parents of children with CA in the long run. The results of this study also support the suggestion of several experts on childhood chronic conditions that variability in family impact may be more related to characteristics of the condition rather than diagnosis per se.

Introduction

The birth of a child with major anatomical congenital anomalies (CA) might alter family functioning in several ways. Parents have to abandon their expectations of a healthy child and have to cope with the painful experience of raising a severely ill child, either temporarily or life-long (Nicholas & Lewin, 1986). These circumstances place a heavy financial, emotional and family burden on parents (Connor, Kline, Mott, Harris, & Jenkins, 2010; Hauser-Cram et al., 2001; Poley, Stolk, Tibboel, Molenaar, & Busschbach, 2004). The frequent hospital visits and the uncertain outcome are only some of the many potential stressors. Once the child is home, they may have to perform special care giving tasks such as nasogastric tube feeding, enterostomy care and giving medication. As a consequence, parents of children with CA are expected to be at risk for experiencing excessive stress (Majnemer et al. 2006). Still, so far there is no generally accepted theory on the psychosocial consequences for becoming parents of children with congenital anomalies.

A study that evaluated parental burden one year after birth of a child with CA found that mothers and fathers experienced similar feelings of burden and grief (Hunfeld, Tempels, Passchier, Hazebroek, & Tibboel, 1999). Mothers, however, reported significantly more personal strain than fathers. Foreknowledge from prenatal diagnosis about the anomaly, a low perceived functional health status of the child, and the child having more than one anomaly were associated with a larger burden and more grief (Hunfeld et al., 1999). However, Skreden et al. (2010) showed that prenatal diagnosis is associated with significantly increased psychological distress among parents in the acute phase, but there was no long-term increase.

In scientific literature, different effects of having a child with CA on parental psychosocial health and their suggested causes are described. A study of Uzark and Jones (2003), measuring parents of children with congenital heart disease, showed that these parents were more likely than the norm group to report excessive parenting stress: approximately 1 in 5 parents expressed clinically significant levels of stress. However, the majority of parents (82%) had a total score in the normal range. The levels of parenting stress were unrelated to the severity of the heart disease. Several experts on childhood chronic conditions (e.g. cystic fibrosis, meningomyelocele) suggested that variability in family impact may be more related to characteristics of the condition, such as age of onset, prognosis, course, or type of incapacitation rather than diagnosis per se (Stein, Bauman, Westbrook, Coupey, & Ireys, 1993; Perrin, et al., 1993). Other experts suggested that variability in family impact may be more dependent of parental factors related to personality, like depression, anxiety and sense of coherence (Glavin, Smith, Sorum, & Ellefsen, 2010; Gudmundsdottir, Schirren, & Boman, 2011; Misri et al., 2010) and parents' intrapersonal resources of positive affectivity (Vermaes, Janssens, Mullaart, Vinck, & Gerris, (2008). Vermaes et al. (2008) found that the severity of the child's physical dysfunction was positively associated with parenting stress in parents of children with spina bifida, and extraversion (in mothers) and emotional stability and agreeableness (in fathers) were negatively related to parenting stress. Parents' personality traits explained the largest proportions of variance in

parenting stress. Furthermore, the quality of the mother-child relationship appears more critical to successful adaption than the severity of illness (DeMaso et al., 1991). Moreover, parental perceptions appear to play a role in the variability in family impact. Maternal perceptions accounted for 33% of the variability in adjustment, while the medical severity accounted for less than 3% of the variability in adjustment (DeMaso et al., 1991). In high-stress mothers, in the development of depressive feelings, the role of perceptions of defeat or entrapment appears to be higher than the role of parenting stress (Willner & Goldstein, 2001). In another study (Vrijmoet-Wiersma, Ottenkamp, Van Roozendaal, Grootenhuis, & Koopman, 2009) parents of children with congenitally malformed hearts did not report higher generic stress scores and parenting levels of stress were also comparable with norm groups. However, both fathers and mothers reported significantly higher rates of perceived vulnerability, regardless of time since surgery or severity of the disease, and mothers had higher state anxiety levels. Parents of children who underwent surgery more than 3 years ago reported lower scores on all measures than parents of children who underwent surgery less than 3 years ago. Perceived vulnerability was found to be a predictor for parental disease-related stress. That is in a study of Vrijmoet-Wiersma, Egeler et al. (2009) general distress scores in parents of children who underwent stem cell transplantation were comparable with the reference group, while perceived vulnerability did diminish over time but remained high in the years after treatment. Besides, Brosig, Mussatto, Kuhn and Tweddell (2007) showed that the level of parental stress was positively correlated with parental perception of total impact of the illness on the family and personal strain. Further, infant temperament also plays a role in the experience of high levels of stress: the demands of parenting an irritable infant with CA puts mothers at risk for high levels of stress (Torowicz, Irving, Hanlon, Sumpter, & Medoff-Cooper, 2010). A study of Warfield (2005) measured predictors of parenting stress as well. Having fewer children in the family predicted less stress for both parents, household income and interaction between child behaviour problems and work interest were significant predictors in mothers, and greater difficulty in finding reliable child care predicted higher levels of parenting stress for fathers.

There have also been studies that view the psychological health of parents with children with CA within the concept of adaptation. For instance, parental adaptation has been described for children with spina bifida, Duchenne or other chronic disability (Barakat & Linney, 1992; Chen & Clark, 2007; Cleve, 1989). Adaptation to a child's disability shows stages of grief and chronic sorrow, eventually leading to acceptance. Kovacs et al. (1990) found that mothers' initial distress following the diagnosis of insulin-dependent diabetes mellitus in their children diminished over the following 6 years. In the other, Dahlquist et al. (1996), found that mothers' anxiety following their children's cancer diagnosis had decreased 2 years later to normal levels. A study of Spijkerboer et al. (2007) supports the idea of the experience of lessened distress. In this study, parents of children treated for congenital heart disease showed lower levels of distress in comparison with norm groups. A study of Visconti, Saudino, Rappaport, Newburger and Bellinger (2002), measuring parenting stress in parents of children with congenital heart disease 1 and 4 years after birth, showed that these parents experienced

less stress and more social support, compared with normative samples. Parents with less social support reported more parenting stress at both 1 and 4 years after birth. Therefore, social support appears to play a crucial role in the experience of parenting stress. More favourable outcomes on coping were found: parents of children with congenital heart disease showed a weaker tendency to use styles of coping such as reassuring thoughts and expressed negative emotions less often. The expression of negative emotions could predict physical and mental health at a later time point (Stroebe, Hansson, Stroebe, & Schut, 2001). Mothers appeared to seek social support more often than fathers (Spijkerboer et al., 2007). Studies showed that even in the face of adverse life conditions, some families not only do all right, but actually become stronger with family communications improving and parental sharing of responsibilities (Masten, 2001; Patterson, 2002; Rodrigues & Patterson, 2007). They gain meaning to their experiences and rebuild their lives (Lalor, Begley, & Galavan, 2009). Therefore, results of studies evaluating the consequences of having a child with CA can be distinguished in results supporting the vision that having a child with CA causes negative psychosocial consequences and the vision that having a child with CA does not definitely cause negative consequences, or even leads to resilience. Resilience is defined as the capacity to resist adverse psychological reactions when suffering risk experiences (Rutter, 2006).

Only few related studies included the perspectives of both parents. Most research on parental experience with children with chronic conditions and severe illness has relied on mothers' reports because she is viewed as the parent closest to the child. In one of the studies including both parents, mothers and fathers had similar views about the functioning of their families (Knafl & Zoeller, 2000), in another study mothers of children with cancer generally reported more stress than fathers (Sawyer, Antoniou, Toogood, Rice, & Baghurst, 1993) and in another mothers of children with congenital heart disease and other diseases reported higher levels of distress and hopelessness than fathers. However, in the control group of healthy children, mothers also reported higher levels of distress and hopelessness than fathers (Lawoko & Soares, 2002). In a study of Mazer, Gischler, Koot, Tibboel, Van Dijk and Van Duivenvoorden (2008) mothers and fathers of children with severe CA are asked to complete a generic health status questionnaire at 6 weeks and 6 months after birth of the child. At both time points both fathers and mothers clearly perceived lower quality of life than the norm group. They particularly had more problems concerning mental health. Mothers perceived lower quality of life than fathers did, at both time points.

The present study is part of a large follow-up study on long-term medical and psychological outcomes in children with CA treated in a pediatric surgical service of a university children's hospital, and in their parents. The aim of the present study is to assess long-term parental psychosocial outcome, to measure the impact of having a child with CA 12 months, 24 months and 5 years after birth of the child. Central questions are to what extent parents of children with CA experience parenting stress and what kind of problems concerning their health they experience 12 months, 24 months and 5 years after birth of the child. Furthermore, correlations between parenting stress and

problems concerning health and sex differences are analyzed. Possible influences of the kind of CA on these relations are described. Considering earlier studies parenting stress and problems concerning health in parents of children with CA are expected to diminish in the long run. At 12 months after birth, parents are expected to experience more parenting stress and problems concerning their health than the norm groups. However, in the long run, at 5 years after birth, parents are expected to experience parenting stress levels and problems concerning health comparable with the norm groups.

Methods

Setting

The Erasmus MC - Sophia children's hospital is a university hospital with a pediatric surgical department in which all surgical specialties except open-heart surgery are represented. As the traditional monospecialistic approach of children with CA was felt to be inadequate, a multidisciplinary support and follow-up team for the management of children with CA and their parents was instituted in 1999. This team consists of pediatricians, a consultant pediatric surgeon, developmental psychologists, a pediatric physical therapist, a social worker, nursing staff, and a clinical geneticist.

Participants

From January 1999 to December 2004 the parents of a total of 360 children with CA, admitted to the pediatric surgical department, were included in the follow-up program. Parents of 51 children did not participate in the follow-up program, 40 children died before the age of 6 months, 3 died between the age of 6 to 12 months, and 8 children died after the age of 12 months. Parents of 101 children were unwilling to complete questionnaires or completed questionnaires at only one time point. Of the parents who refused to complete questionnaires 68.97% (20/29) were non-native Dutch.

Parents of 157 children participated in the study, that is, returned questionnaires for at least two time points (figure 1). Largely in line with Ravitch's so-called surgical index diagnoses of CA (Ravitch & Barton, 1974) the following categories were distinguished: 1) small intestinal anomaly (SIA), 2) abdominal wall defect (AWD), 3) congenital diaphragmatic hernia (CDH), 4) esophageal atresia (EA), and 5) colorectal diseases (CD).

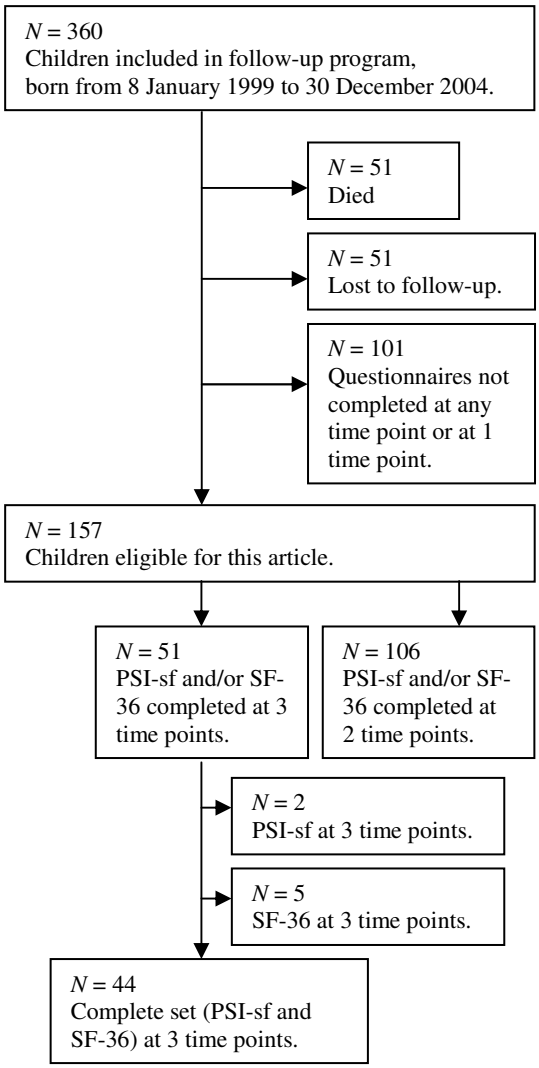

Figure 1. General flow-chart.

Design

This is a prospective, longitudinal cohort study comprising three assessments: at 12 months, 24 months, and 5 years after the birth of the child.

Procedure

The ethical review board agreed with the study, and written parental informed consent was obtained for all subjects. At 12 months, 24 months, and 5 years parents were asked to complete the SF-36 (Coons, Rao, Keininger, & Hays, 2000; Van der Zee & Sanderman, 1993; Ware, 2000) and the Parenting Stress Index – short form (PSI-sf; Abidin, 1983; De Brock, Vermulst, Gerris, & Abidin, 1992). Assessments at 12 and 24 months were corrected for gestational age of the child. Parent couples were explicitly instructed to complete the questionnaires independently. The questionnaires were mailed to the parents. Usually the questionnaires were completed at home and returned on the day of the follow-up appointment or were returned by mail.

All demographic and medical data as shown in Table 2 were collected prospectively from the first day of admission. Ethnicity was categorized as being native or non-native to the country. Socioeconomic status (SES) was assessed according to a national classification as 'low', or 'high' (Statistics Netherlands, 1992). Medical characteristics include type of anomaly, total number of minor and major anomalies, duration of admission in the first 6 months, number of medical applicanes at discharge, surgical interventions and additional medical problems in the first 24 months (see Table 2). Medical appliances include: medications, oxygen therapy, tracheostomy, nasogastric tube, enterostomy, heart and respiration monitoring, central venous line, airway suction and other medical appliances (e.g. rectal cannula and enema).

Instruments

Parenting Stress Index – Short Form

The parent-child relationship was assessed using a validated Short Form (SF) of the
Dutch version of the Parenting Stress Index (PSI) (De Brock, et al., 1992) of the Parenting Stress Index (PSI; Abidin, 1983; Loyd & Abidin, 1985). The PSI-SF was designed to assess the degree of stress related to parenting in parents of children ages to 13 years. It consists of 25 items derived from the PSI, which comprise three domains: parental characteristics (distress), (difficult) child characteristics and situational variables (dysfunctional parent-child interactions). The items are scored on a six-point Likert scale from strongly disagree (1) to strongly agree (6). A total score is obtained by calculating the scores on the 25 items; total score thus may range from 25 to 150. The higher the score, the more stress reported. Scores were compared with Dutch population norms for mothers (M = 54.4, SD = 19,3) and fathers (M = 48.5, SD = 16,4) separately. The sums of scores were used in statistical analyses.

SF-36

The SF-36 is a generic health status questionnaire (Coons, et al., 2000; Van der Zee & Sanderman, 1993; Ware, 2000). It consists of 36 questions organized into eight domains: 1) physical functioning (M = 89.5, SD = 17.8), 2) social functioning (M = 90.7, SD = 16.5), 3) role limitations because of physical health problems (M = 82.5, SD = 32.4), 4) role limitations because of emotional problems (M = 86.8, SD = 29.6), 5) general mental health (M = 78.8, SD = 17.5), 6) vitality (M = 69.1, SD = 19.0), 7) bodily pain (M = 84.1, SD = 23.9), and 8) general health (M = 77.5, SD = 19.7).

Two summary measures can be derived: physical health (including domains 1, 3, 7 and 8; M = 83.4, SD = 24.1) and mental health (including domains 2, 4, 5 and 6; M = 81.4, SD = 21.3). Total scores are linearly transformed to range from 0 to 100, with higher scores indicating a better-perceived health status. Dutch population norms adjusted for age (25 - 34 years) were used (Van der Zee & Sanderman, 1993). These two summary measures are mostly used in statistical analyses. A generic

measure, the SF-36 has proven useful in surveys of general and specific populations (Coons, et al., 2000).

In Table 1 an overview of returned questionnaires, separated by questionnaire, time point and gender of the parent are presented. Furthermore, the total amount of couples and complete sets is displayed.

Table 1. *Number of returned questionnaires (N = 157).*

		12 months N (%)	*24 months N (%)*	*5 years N (%)*	*Complete set N (%)*
PSI-SF	Mothers	125 (79.6)	94 (59.9)	125 (79.6)	57 (36.3)
	Fathers	108 (68.8)	86 (54.8)	112 (71.3)	51 (32.5)
	Couples	105 (66.9)	79 (50.3)	111 (70.7)	46 (29.3)
	Total	233	180	237	108
SF-36	Mothers	98 (62.4)	129 (82.2)	92 (58.6)	60 (38.2)
	Fathers	88 (56.1)	109 (69.4)	84 (53.5)	50 (31.8)
	Couples	84 (53.5)	107 (68.2)	82 (52.2)	49 (31.2)
	Total	186	238	176	110

Statistical analyses

As measures of central tendency the means (normal distribution) and medians (non-normal distribution) are presented. The standard deviation (normal distribution) and interquartile range (non-normal distribution) served as measures of dispersion. In case of categorical data the numbers and percentages are presented. The Intra-Class Correlation coefficient (two-way mixed model) is used to estimate the inter-parent agreement. For these correlations the rule of thumb for effect size provided by Cohen (1988) is used: low = .10, moderate = .24 or high = .37.

The paired-sample t-test is used to determine significant differences for paired samples (mothers and fathers) at each time point for the SF-36 only. Between assessments of the same parents across time for both PSI-SF and SF-36 mixed-model ANOVA's are used. All statistical testing is performed at the .05 level of significance (two-tailed). The software program SPSS 15.0 for Windows is used.

Results

General characteristics

Characteristics of the 157 children and parents, 156 mothers and 149 fathers, are presented in Table 2. The median number of major anomalies per child was 1 (IQR: 1-2) and the median number of minor anomalies per child was 0 (IQR: 0-1). Median duration of admission in the first 6 months was 39 days. Children underwent a median of three surgical interventions in the first 12 months (IQR: 1-6), no surgical intervention from 12 to 24 months (IQR: 0-0) and no interventions from 24 months to 5 years (IQR: 0-0). Ten children with major chromosomal or syndromal abnormality were at risk of severe psychomotor delay. One or both parents of 20 children were non-native Dutch, but had enough

command of Dutch language to fill in the questionnaires. The median age of the mothers at delivery was 31 years; that of the father 33 years. In 58% of the families the child with CA was first born.

Table 2. *General characteristics of patients and parents (N = 157).*

		N	*%*	*Mdn*	*IQR*
Patients					
Female		74	47.1		
First born		91	58.0		
Primary Congenital Anomaly	Small intestinal anomaly	54	34.4		
	Abdominal wall defect	33	21.0		
	Congenital diaphragmatic hernia	23	14.6		
	Oesophageal atresia	27	17.2		
	Colorectal diseases	20	12.7		
Chromosomal or syndromal abnormality		10	6.4		
1 major congenital anomaly (number)		111	70.7		
More than 1 major congenital anomaly (number)		46	29.3		
0 to 1 minor congenital anomaly (number)		135	86.0		
More than 1 minor congenital anomaly (number)		22	14.0		
0 to 1 additional medical problem (number)		69	43.9		
More than 1 additional medical problem (number)		88	56.1		
Major congenital anomalies per patient (number)				1	1 to 2
Minor congenital anomalies per patient (number)				0	0 to 1
Additional medical problems (number)				2	1 to 3
Total admission in first 6 months (days)				39	24 to 77
Medical appliances at discharge (number) *				1	0 to 2
Surgical interventions in first 12 months (number)				3	1 to 6
Surgical interventions 12 to 24 months (number)				0	0 to 0
Surgical interventions 24 months to 5 years (number)				0	0 to 0
Parents					
Age mothers at delivery				31	28 to 34
Age fathers at delivery				33	30 to 37
Number of children in the family at delivery				1	0 to 1
1 or both parent(s) non-Dutch		20	12.7		
Socioeconomic status	Low	27	17.2		
	Medium	76	48.4		
	High	53	33.8		
	Missing	1	0.6		

* Medical appliances include: medication, oxygen, tracheostomy, nasogastric tube, enterostomy, heartrate and respiration monitoring, central venous line, airway suction and other medical appliances (e.g. rectal canula and clysmata).

Table 3. SF-*36 and PSI-sf distinguished by parent across time.*

	12 months						*24 months*						*5 years*					
	Mothers			*Fathers*			*Mothers*			*Fathers*			*Mothers*			*Fathers*		
SF-36	M	SD	N	M	SD	N	M	SD	N	M	SD	N	M	SD	N	M	SD	N
Physical component scale	82.36	18.07	98	85.43	18.21	88	83.14	20.13	129	87.97*	16.68	109	85.81	15.17	92	86.00	17.81	84
Mental component scale	75.58*	17.84	98	82.06	14.73	88	77.26	21.12	129	85.24*	12.81	109	78.66	17.89	92	82.07	14.40	84
Physical functioning	91.48	14.27	98	93.28	14.48	88	90.27	19.92	129	93.85*	15.63	109	94.02*	10.49	92	94.13*	13.17	84
Social functioning	83.29*	22.77	98	89.20	18.29	88	86.82	22.66	129	92.55	14.35	109	88.59	18.50	92	88.39	15.86	84
Role limitations, physical prob.	79.59	37.02	98	85.23	31.40	88	81.46	34.39	129	87.39	27.98	109	87.23	29.76	92	85.71	30.38	84
Role limitations, emotional prob.	84.01	31.47	98	92.05	21.44	88	81.65	35.59	129	91.74	22.75	109	85.87	30.15	92	90.48	28.14	84
General mental health	75.45	16.00	98	79.95	15.22	88	76.95	17.65	129	82.83*	12.16	109	75.53	18.68	92	80.01	14.06	84
Vitality	59.57*	17.84	98	67.03	17.39	88	63.60*	19.45	129	73.85*	16.20	109	64.64	18.49	92	69.40	16.31	84
Bodily pain	83.49	22.79	98	87.50	22.75	88	83.39	26.49	129	90.10*	16.87	109	85.09	20.83	92	86.76	22.10	84
General health	74.90	19.66	98	75.72	19.31	88	77.44	21.54	129	80.56	18.47	109	76.90	18.88	92	77.41	19.08	84
PSI-SF	51.15	16.02	125	46.45	13.92	108	51.59	17.64	94	45.01	14.59	86	47.70*	19.93	125	49.47	18.49	112

*) Significant at the .05 level (two-tailed).

Parenting stress

Comparison with population norms

At 12 months, the mean stress scores of mothers and fathers were 51.2 (SD = 16.0) and 46.5 (SD = 13.9), respectively. At 24 months, the mean scores of mothers and fathers were 51.6 (SD = 17.6) and 45.0 (SD = 14.6), respectively. At 5 years, the mean scores of mothers and fathers were 47.7 (SD = 19.9) and 49.5 (18.5), respectively. At 5 years, mothers experienced less parenting stress than the Dutch population norms, with significantly lower scores on the PSI-SF (t (286) = -2.86, $p < .05$). Other scores did not differ significantly from the Dutch population norms (see Table 3 on page 14).

Parental agreement

The intra-class correlations of PSI-SF scores between mothers and fathers were high: .58 at 12 months, .68 at 24 months and .53 at 5 years.

Changes over time

Analysis on longitudinal evaluative data using mixed-model ANOVA showed changes in PSI-SF scores of fathers over time, both between 12 months and 24 months and between 24 months and 5 years (t (187) = -2.37, $p < .05$; t (193) = -3.73, $p = <.001$, respectively). This analysis did not show changes in PSI-SF scores of mothers over time.

General health status

Comparison with population norms

As shown in Table 3, at 12 months, the mean scores of mothers and fathers on the physical components scale (PCS) were 82.4 (SD = 18.1) and 85.4 (SD = 18.2), respectively, and on the mental component scale (MCS) were 75.6 (SD = 17.8) and 82.1 (SD = 14.7), respectively. At 24 months, the mean scores of mothers and fathers on the PCS were 83.1 (SD = 20.1) and 88.0 (SD = 16.7), respectively, and on the MCS were 77.3 (SD = 21.1) and 85.2 (SD = 12.8), respectively. At 5 years, the mean scores of mothers and fathers on the PCS were 85.8 (SD = 15.2) and 86.0 (17.8), respectively, and on the MCS were 78.7 (SD = 17.9) and 82.1 (SD = 14.4).

At 12 months, mothers perceived more mental problems than the norm group, with lower scores on the MCS (t (319) = -2.53, $p < .05$). Analyzing the scores on the eight domains of the SF-36, mothers perceived more social problems and problems concerning vitality than the norm group (t (319) = -2.90, $p < .05$; t (319) = -4.13, $p < .05$, respectively). At 24 months, fathers perceived less physical and mental problems than the norm group, with higher scores on the PCS and the MCS (t (330) = 2.01, $p < .05$; t (330) = 2.04, $p < .05$, respectively). Analyzing the scores on the eight domains, mothers perceived only more problems than the norm group concerning vitality (t (350) = -2.57, $p < .05$), and fathers perceived less physical problems and problems concerning mental health, vitality and pain than the norm group (t (330) = 2.27, $p < .05$; t (330) = 2.43, $p <.05$; t (330) = 2.36, $p < .05$; t (330)

= 2.63, $p < .05$, respectively). At 5 years, both mothers and fathers perceived less physical problems than the norm group (t (313) = 2.79, $p < .05$; t (305) = 2.48, $p < .05$, respectively).

Parental agreement

At 12 and 24 months, paired measurements showed that mothers scored worse than fathers on the MCS (t (84) = 2.50, $p < .05$; t (107) = 4.19, $p < .001$, respectively). This difference was no longer present at the 5-year interval.

At 12 and 24 months, the intra-class correlations of scores on the PCS and MCS for mothers and fathers were high (.34 and .43; .45 and .44, respectively), and at 5 years, the intra-class correlations were low and high, .15 and .32, respectively.

Changes over time

Analysis on longitudinal evaluative data using mixed-model ANOVA did not show significant changes in PCS and MCS scores of fathers and mothers over time.

Scores at clinical range on PSI-sf and SF-36

Analyzing the characteristics of the children of parents scoring at the clinical range on the PSI-SF and the SF-36, particular things attract attention. First, all subgroups of CA are represented. None of the subgroups was overrepresented. The specific subgroup, or diagnosis, does not appear to be a valid indicator of severity of the illness. However, other characteristics accompanying CA seem to be a valid indicator of severity of the illness. That is, in general children of parents scoring at the clinical range on the PSI-sf and the SF-36 have more admission days in the first 6 months of their lives, have had more surgical interventions in the first 12 months of their lives, in comparison with children with parents scoring at the normal range on the PSI-SF and SF-36. Moreover, these children have more than one minor or major CA. Furthermore, these children often have more medical appliances at discharge. Therefore, these child characteristics seem to be valid predictors for scores on the PSI-SF and the SF-36 of parents at the clinical range. As a result, the severity of the illness, as presented by the characteristics described above, seems to be a valid predictor of experiencing parenting stress and physical and mental health problems.

Looking at the subgroups of CA, it is noticeable that in some subgroups the numbers of parents scoring at the clinical range are the same on the PSI-SF and the SF-36. However, in other subgroups these numbers on the PSI-SF differ in comparison with the SF-36. This is the case in the subgroup of AWD and the subgroup of EA. A possible explanation for these differences is that in one disease impact is more noticeable in parenting and in the other the impact is more noticeable in physical and mental health.

Discussion

This prospective longitudinal study investigated the effects of having a child with a major anatomical CA on parenting stress and parental general health, aspects which both may influence quality of life. The results of this study indicate that parents of children with CA are capable of coping with a situation of having a severely ill child in the long run. Only at 12 months after birth of the child with CA, mothers perceived more mental problems than the norm group. However, at 24 months, fathers even perceived less mental problems than the norm group. Concerning parenting stress, at 5 years mothers experienced less parenting stress than the Dutch population norms. Other results on mental and physical health and parenting stress did not differ significantly from population norms. It seems that the process of parental acceptance was apparently resolved and parental stress was balanced out. Parenting stress declined over time in fathers but not in mothers. Problems concerning health did not decline over time. Generally, parental agreement on mental and physical health and parenting stress were high. At 12 and 24 months, mothers experienced significantly more mental problems than fathers. At 5 years, no differences between mothers and fathers could be found.

The results of this study support the notion of resilience in parents of children with CA in the long run. The presence of a stressor, such as having to care for a child with a chronic condition, appear to challenge families to use internal resources and to develop internal strengths for managing the situation. It is important to mention that the findings of this study, psychosocial resilience in the long run, cannot be generalized to all parents in this study group. The results of this study support the suggestion of several experts on childhood chronic conditions (Stein, et al., 1993; Perrin, et al., 1993) that variability in family impact may be more related to characteristics of the condition rather than diagnosis per se. The specific subgroup, or diagnosis, does not appear to be a valid indicator of severity of the illness. However, other characteristics going together with the CA, like more admission days in the first 6 months, more surgical interventions in the first 12 months, more than one minor or major CA and more medical appliances, seem to be al valid indicator of severity of the illness.

This study has the strength of measuring long-term impact on parents of children with CA. This has rarely been performed. Moreover, most studies on this impact do not differentiate between fathers and mothers separately. In addition, the study-design not only allowed to measure two-parent families but also allowed separately analyzing the single parents. A limitation of this study might be that the available medical characteristics did not allow predicting an outcome in terms of parenting stress and general health. On the other hand, outcome over time appeared to be within the normal range.

It has been argued that, considering the results of this study, lower rates of parenting stress and problems concerning health in parents of children with CA in the long run in comparison with the norm groups may be explained by the phenomenon of resilience. However, recent studies in the scientific field of the impact of serious life events explained these higher scores on perceived quality of life by the phenomenon of posttraumatic growth and response shift. That is, a study of Colville and

Cream (2009) showed that parents of children admitted to the intensive care unit with being ventilated, scored higher on the Posttraumatic Growth Inventory (Tedeschi & Calhoun, 1996) than parents of children on the intensive care unit who underwent less traumatic interventions. These outcomes indicate that these parents have experienced a positive change due to their experiences. Besides, Ito et al. (2010) showed that differences in scores on perceived quality of life pre and post event may be explained by response shift: the unit of comparison in the participants has been changed by the experience of serious life events. It is possible that one of these relatively new phenomena in scientific literature, posttraumatic growth and response shift, could explain the results of this study, too. Another possibility is that there is conceptual overlap between the phenomena of resilience, posttraumatic growth and response shift. Future research should clarify the conceptual differences between these phenomena.

Future studies should clarify the predicting capacity of extended medical characteristics. Moreover, it is not precluded that parents consulted each other about their responses to the questionnaires, which may have influenced parental agreement. Furthermore, this study presents data of parents who had enough command of language to understand and fill in the questionnaires. Future studies about quality of life of parents of other and more diverse cultures is needed to become aware of their needs and be able to offer support according to those needs.

Experts suggested that variability in family impact may be more dependent of parental factors related to personality, like depression, anxiety and sense of coherence (Misri et al., 2010; Glavin et al., 2010; Gudmundsdottir et al., 2011) and parents' intrapersonal resources of positive affectivity (Vermaes et al., (2008). Furthermore, DeMaso et al. (1991) showed that the quality of the mother-child relationship appears more critical to successful adaption than the severity of illness. Moreover, parental perceptions appear to play a role in the variability in family impact (Willner & Goldstein, 2001; Brosig et al., 2007), and infant temperament plays a role in the experience of high levels of stress (Torowicz et al., 2010). In the present study the influences of parental factors related to personality, infant temperament and differences in perceptions of parents have not been measured. In future studies these variables should be included, because it is plausible that the way parents perceive the whole situation of their child is more determining for the psychosocial condition of the parents than the objective circumstances.

The results of the study on parents of children with congenitally malformed hearts have shown that rates of perceived vulnerability of the child were higher in these parents in comparison with parents of healthy children, regardless of the time since surgery or severity of the disease (Vrijmoet-Wiersma, Ottenkamp et al., 2009) and higher in parents of children who underwent more severe surgical interventions, like stem cell transplantation (Vrijmoet-Wiersma, Egeler et al., 2009). Furthermore, a study of Fedele et al (2010) showed that continued use of the modified Parenting Stress Index – short form appears appropriate in a pediatric illness population, but some items of the PSI-SF, the majority on the domain of dysfunctional parent-child interactions, are not appropriate or relevant

to parents of children with a chronic illness, or, possibly these parents may generally have difficulty acknowledging problematic interactions with their children. Therefore, perceived vulnerability of the child appears to be a more valid indicator for the experience of excessive amounts of disease-related stress in parents of children with CA. The study of Fedele et al. (2010) showed that the use of the Child Vulnerability Scale (Forsyth, Horwitz, Leventhal, Burger, & Leaf, 1996) appears appropriate in a pediatric illness population. Consequently, measuring perceived vulnerability of children with CA to predict excessive stress in parents of these children appears to be a recommendation for future studies in this specialism.

Although parents of children with CA face many challenges, in the long run they seem capable of coping effectively with the situation of having a severely ill child. It appears that a mentally and physically healthy parent is able to cope with the situation of having a child with CA. Good adaptation strategies make parents less vulnerable to stress. Few studies about the psychosocial effects of having a child with CA have a strengths-focusing orientation. Most studies intend to measure negative effects of having a child with CA, while this is not necessary the case. In future studies measuring the effects of having a child with CA the resilience, strengths-focusing, perspective should be extended, because this may enable training or counselling programs for parents in order to prevent negative psychosocial effects of having a child with CA. Future studies would do well to evaluate coping style in relation to the impact of having a child with CA. Moreover, according to recent scientific literature, measuring negative psychosocial effects of having a child with CA should be done by measuring perceived vulnerability instead of measuring parenting stress. Considering the results of this study, this training or counselling should be focused on parents of children with CA with a high number of admission days in the first months after birth, increased surgical interventions in the first year after birth, more than one minor or major CA and the use of a high number of medical appliances.

References

Abidin, R. R. (1983). *Parental Stress Index. Manual.* Charlottesville: Pediatric Psychology Press.

Barakat, L. P., & Linney, J. A. (1992). Children with physical handicaps and their mothers: the interrelation of social support, maternal adjustment, and child adjustment. *Journal of Pediatric Psychology, 17,* 6, 725-39.

Brosig, C. L., Mussatto, K. A., Kuhn, E. M., & Tweddell, J. S. (2007). Psychosocial outcomes for preschool children and families after surgery for complex congenital heart disease. *Pediatric Cardiology, 28,* 255-262.

Chen, J. Y., Clark, M. J. (2007). Family function in families of children with Duchenne muscular dystrophy. *Family & Community Health, 30,* 4, 296-304.

Cleve, L. V. (1989). Parental coping in response to their child's spina bifida. *Journal of Pediatric Nursing, 4,* 3, 172-6.

Cohen, J. (1988). *Statistical power analyses for the behavior sciences* (2nd Ed.). Hillsdale, New Jersey: Lawrence Erlbaum.

Colville, G., & Cream, P. (2009). Post-traumatic growth in parents after a child's admission to intensive care: maybe Nietzsche was right? *Intensive Care Medicine, 35,* 919-923.

Connor, J. A., Kline, N. E., Mott, S. M., Harris, S. K., & Jenkins, K. J. (2010). The meaning of cost for families of children with congenital heart disease. *Journal of Pediatric Health Care, 24,* 5, 318-325.

Coons, S. J., Rao, S., & Keininger, D. L. (2000). A comparative review of generic quality-of-life instruments. *Pharmacoeconomics,17,* 13-35.

Dahlquist, L. M., Czyzewski, D. I., & Jones, C. L. (1996). Parents of children with cancer: a longitudinal study of emotional distress, coping style, and marital adjustment two and twenty months after diagnosis. *Journal of Pediatric Psychology, 21,* 4, 541-54.

De Brock, A. J. L. L., Vermulst, A. A., Gerris, J. R. M., & Abidin, R. R. (1992). *Nijmeegse Ouderlijke Stress Index. Handleiding* [PSI, Dutch manual]. Lisse: Swets en Zeitlinger.

DeMaso, D.R., Campis, L. K., Wypij, D., Bertram, S., Lipshitz, & Freed, 1991. The impact of maternal perception and medical severity on the adjustment of children with congenital heart disease. *Journal of Pediatric Psychology, 16,* 2, 137-149.

Fedele, D. A., Grant, D. M., Wolfe-Christensen, Mullins, L. L., & Ryan, J. L. (2010). An examination of the factor structure of parenting capacity measures in chronic illness population. *Journal of Pediatric Psychology, 35,* 10, 1083-1092.

Forsyth, B., Horwitz, S., Leventhal, J., Burger, J., & Leaf, P. (1996). The child vulnerability scale: an instrument to measure parental perceptions of child vulnerability. *Journal of Pediatric Psychology, 21,* 89-101.

Glavin, K., Smith, L. S., Sorum, R., & Ellefsen, B. (2010). Redesigned community postpartum care to prevent and treat postpartum depression in women – a one-year follow-up study. *Journal of Clinical Nursing, 19,* 3051-3062.

Gudmundsdottir, E., Schirren, M. & Boman, K. K. (2011). Psychological resilience and long-term distress in Swedish and Icelandic parents' adjustment to childhood cancer. *Acta Oncologica, 50,* 373-380.

Hauser-Cram, P., Warfield, M. E., Shonkoff, J. P., Krauss, M. W., Sayer, A., & Upshur, C. C. (2001). Children with disabilities: a longitudinal study of child development and parent well-being. *Monographs of the Society for Research in Child Development, 66,* 3: i-viii, 1-114; discussion 5-26.

Hunfeld, J. A., Tempels, A., Passchier, J., Hazebroek, F. W., & Tibboel, D. (1999). Brief report: parental burden and grief one year after the birth of a child with a congenital anomaly. *Journal of Pediatric Psychology, 24,* 6, 515-20.

Ito, N., Ishiguro, M., Tanaka, M., Tokunaga, K., Sugihara, K., & Kazuma, K. (2010). Repsonse shift in quality-of-life assessment in patients undergoing curative surgery with permanent colostomy; a preliminary study. *Gastroenterology Nursing, 33,* 6, 408-412.

Knafl, K., & Zoeller, L. (2000). Childhood chronic illness: A comparison of mothers and fathers experiences. *Journal of Family Nursing, 6,* 287-302.

Kovacs, M., Iyengar, S., Goldston, D., Obrosky, D. S., Stewart, J., & Marsh, J. (1990). Psychological functioning among mothers of children with insulin-dependent diabetes mellitus: a longitudinal study. *Journal of Consulting and Clinical Psychology, 58,* 2,189-95.

Lalor, J., Begley, C. M., & Galavan, E. (2009). Recasting hope: a process of adaptation following fetal anomaly diagnosis. *Social Science & Medicine, 68,* 462-472.

Lawoko, S., & Soares, J. J. F. (2002). Distress and hopelessness among parents of children with congenital heart disease, parents of children with other diseases, and parents of healthy children. *Journal of Psychosomatic Research, 52,* 193-208.

Loyd, B. H., & Abidin, R. R. (1985). Revision of the Parenting Stress Index. *Journal of Pediatric Psychology, 10,* 2, 169-177.

Majnemer, A., Limperopoulos, C., Shevell, M., Rohlicek, C., Rosenblatt, B., & Tchervenkov, C. (2006). Health and well-being of children with cardiac malformations, and their families, following open-heart surgery. *Cardiology in the Young, 16,* 157-164.

Masten, A. (2001). Ordinary magic: Resilience processes in development. *American Psychologist, 56,* 227-38.

Mazer, P., Gischler, S. J., Koot, H. M., Tibboel, D., Van Dijk, M, & Van Duivenvoorden, H. J. (2008). Impact of a child with congenital anomalies on parents (ICCAP) questionnaire; a psychometric analysis. *Health and Quality of Life, 6,* 102.

Misri, S., Kendrick, K., Oberlander, T. F., Norris, S., Tomfohr, L., Zhang, H., & Grunau, R. E. (2010). Antenatal depression and anxiety affect postpartum parenting stress: a longitudinal, prospective study. *La Revue canadienne de psychiatrie, 55,* 4, 222-228.

Nicholas, A. M., & Lewin, T. J. (1986). Grief reactions of parental couples: congenital handicap and cot death. *Medical Journal of Australia, 144,* 6, 292-5, 8.

Patterson, J. (2002). Integrating family resilience and family stress theory. *Journal of Marriage and Family, 64,* 349-60.

Perrin, E. C., Newacheck, P., Pless, I. B., Drotar, D., Gortmaker, S. L., Leventhal, J., et al. (1993). Issues involved in the definition and classification of chronic health conditions. *Pediatrics, 91,* 4, 787-93.

Poley, M. J., Stolk, E. A., Tibboel, D., Molenaar, J. C., & Busschbach, J. J. (2004). Short term and long term health related quality of life after congenital anorectal malformations and congenital diaphragmatic hernia. *Archives of Disease in Childhood, 89,* 9, 836-41.

Ravitch, M. M., & Barton, B. A. (1974). The need for pediatric surgeons as determined by the volume of work and the mode of delivery of surgical care. *Surgery, 76,* 5, 754-63.

Rodrigues, N., & Patterson, J. M. (2007). Impact of severity of a child's chronic condition on the functioning of two-parent families. *Journal of Pediatric Psychology, 32,* 4, 417-26.

Rutter, M. (2006). Implications of resilience concepts for scientific understanding. *Annals of the New York Academic Science, 1094,* 1–12.

Sawyer, M. G., Antoniou, G., Toogood, I., Rice, M., & Baghurst, P. A. (1993). A prospective study of the psychological adjustment of parents and families of children with cancer. *Journal of Paediatrics and Child Health, 29,* 5, 352-6.

Skreden, M., Skari, H., Malt, U. F., Haugen, G., Pripp, A. H., Faugli, A., & Emblem, R. (2010) Long-term parental psychological distress among parents of children with a malformation – a prospective longitudinal study. *American Journal of Medical Genetics Part A, 152A,* 2193-2202.

Spijkerboer, A. W., Hebling W. A., Bogers, A. J. J. C., Van Domburg, R. T., Verhulst, F. C., Utens, E. M. W. J. (2007). Long-term psychological distress, and styles of coping, in parents of children and adolescents who underwent invasive treatment for congenital cardiac disease. *Cardiology in the Young, 17,* 638-645.

Statistics Netherlands (1992). *Standaard Beroepenclassificatie [Standard Occupational Classification].* Voorburg: CBS.

Stein, R. E., Bauman, L. J., Westbrook, L. E., Coupey, S. M., & Ireys, H.T. (1993). Framework for identifying children who have chronic conditions: the case for a new definition. *Journal of Pediatrics, 122,* 342-7.

Stroebe, M., Hansson, R. O., Stroebe, W. & Schut, H. (Eds.) (2001). *Handbook of bereavement research: Consequences, coping and care.* Washington: American Psychological Association.

Tedeschi, R., Calhoun, L. (1996). The Posttraumatic Growth Inventory: measuring the positive legacy of trauma. *Journal of Traumatic Stress, 9,* 455-471.

Torowicz, D., Irving, S. Y., Hanlon, A. L., Sumpter, D. F., & Medoff-Cooper, B. (2010). Infant temperament and parental stress in 3-month-old infants after gurgery for complex congenital heart-disease. *Journal of Development & Behavioral Pediatrics, 31,* 3, 202-208.

Uzark, K., & Jones, K. (2003). Parenting stress and children with heart disease. *Journal of Pediatric Health Care, 17,* 163-168.

Van der Zee, K. I., & Sanderman, R. (1993). *Het meten van de algemene gezondheidstoestand met de Rand-36: een handleiding [SF-36, Dutch manual].* Groningen: Centrum voor gezondheidsvraagstukken.

Vermaes, I. P. R., Janssens, J. M. A. M., Mullaart, R. A., Vinck, A., & Gerris, J. R. M. (2008). Parents' personality and parenting stress in families of children with spina bifida. *Child: care, health and development, 34,* 5, 665-674.

Visconti, K. J., Saudino, K. Y. J., Rappaport, L. A., Newburger, J. W., & Bellinger, D. C. (2002). Influence of parental and social support on the behaviour adjustment of children with transposition of the great arteries. *Development and Behavioral Pediatrics, 23,* 5, 314-321.

Vrijmoet-Wiersma, C. M. J., Egeler, R. M., Koopman, H. M., Bresters, D., Norberg, A. L., & Grootenhuis, M. A. (2009). Parental stress and perceived vulnerability at 5 and 10 years after pediatric SCT. *None Marrow Transplantation, 1-7.*

Vrijmoet-Wiersma, C. M. J., Ottenkamp, J., Van Roozendaal, M., Grootenhuis, M. A., & Koopman, K. M. (2009). A multicentric study of disease-related stress, and perceived vulnerability, in parents of children with congenital cardiac disease. *Cardiology in the Young, 19,* 608-614.

Ware, J. E. (2000). SF-36 Health Survey Update. *Spine, 25,* 3130-9.

Warfield, M. E. (2005). Family and work predictors of parenting role stress among two-earner families of children with disabilities. *Infant and Child Development, 14,* 155-176.

Willner, P., & Goldstein, R. C. (2001). Mediation of depression by perceptions of defeat and entrapment in high-stress mothers. *British Journal of Medical Psychology, 74,* 473-485.