Journal of Pediatric Psychology, Vol. 24, No. 6, 1999, pp. 515–520

Brief Report: Parental Burden and Grief One Year After the Birth of a Child With a Congenital Anomaly

Joke A. M. Hunfeld,[1,2] PhD, ***Angelique Tempels,***[1,2] ***Jan Passchier,***[1] PhD, ***Frans W. J. Hazebroek,***[2] PhD, MD, and ***Dick Tibboel,***[2] PhD, MD

[1]Erasmus University, Medical Psychology and Psychotherapy and [2]University Hospital, Rotterdam, The Netherlands

Objective: **To assess parental burden and grief one year after having a child with a congenital anomaly.**

Method: **Twenty-five couples completed the Impact on Family Scale (IFS) and 22 couples answered the Perinatal Grief Scale (PGS). In addition, 27 mothers completed the Functional Health Status Scale (FSII-R).**

Results: **Mothers and fathers showed no significant differences in overall burden (IFS) and grief (PGS). Regarding the subscales, mothers reported significantly more personal strain. Foreknowledge from prenatal diagnosis about the anomaly, a low perceived functional health status of the child, and multiple congenital anomalies increased the burden and grief.**

Conclusions: **A perinatal counseling team that provides clear and consistent information about the anomalies, the treatment, and the prognosis would help to reduce unnecessary stress and uncertainty, particularly for parents who received prenatal information and whose infant has multiple congenital anomalies.**

Key words: *multiple congenital anomalies; parents; burden; grief.*

In the Netherlands approximately 200,000 infants are born per year. About 8,000 (4%) of them display congenital structural pathology, which is severe in 2.5% of them. Dutch pregnant women are eligible for prenatal diagnosis if they have a known risk for fetal abnormalities, advanced maternal age, a previous affected child, or are a known carrier of a genetic abnormality. They are also eligible when fetal problems are discovered during a routine ultrasound scan, such as severe growth retardation or severe polyhydramnion. In these cases, except for advanced maternal age, Dutch women are invited for prenatal testing in a subsequent pregnancy. Women of advanced maternal age are told about prenatal testing by the Dutch media, the family physician, or midwife. Usually these scans take place between the 18th and 20th week of gestation. Due to rapid progress in pediatric surgical specialties and neonatal intensive care, most anomalies lead not to death but to sometimes long-term morbidity even beyond infancy or childhood.

Although the birth of a child with a defect means grief and burden, few studies have assessed the grief and burden of the parents. Clinical observations show that the mother who, in the majority of cases, is the primary care provider particularly experiences a heavy burden. Further, some authors stress the severity of the illness as an important factor in the parent's burden (Walker, Ford, & Donald,

All correspondence should be sent to Joke A. M. Hunfeld, Erasmus University, Cf308, P.O. Box 1738, 3000 DR Rotterdam, The Netherlands. E-mail: hunfeld@mpp.fgg.eur.nl.

1987), whereas, according to others, the severity may not be crucial to the burden at all (Canning, Harris, & Kelleher, 1996).

Studies on the impact of having a child with a birth defect often used heterogeneous samples with respect to the children's age, varying from 0–18 years (Canning et al., 1996). It is therefore difficult to assess the burden related to a child's medical condition separately from his or her developmental phases. If problems, and factors associated with these problems, in family functioning due to the child's medical condition can be detected at an early stage, intervention can follow and prevent problems from worsening. Therefore, we addressed the following questions: (1) What are the differences in burden and grief between mothers and fathers of a child with a congenital anomaly one year after the child's birth? (2) What is the impact of having foreknowledge from prenatal diagnosis, the nature of the diagnosis (doctor's report), and the child's functional health status (mother's report) on parental burden and grief? We expected that being the mother, having a child with multiple congenital anomalies, and perceiving the health status of the child as low would involve more burden and grief, due to the diversity of problems and the severity of the child's medical condition. We also expected more burden and grief from foreknowledge from prenatal diagnosis because parents have to face the traumatic event twice, in imagination during diagnosis and in reality after birth.

Method

Participants

We approached the parents of all 27 newborns admitted to the Pediatric Surgery Department of Sophia's Children's Hospital for surgical treatment for major congenital anomalies in 1996 and who met the remaining inclusion criteria mentioned below. These anomalies were either isolated (one organ, $n = 18$) or multiple (two or more anomalies of one organ and chromosomal anomalies, $n = 9$, for example meconium peritonitis due to cystic fibrosis). Inclusion criteria were (1) a child in the first year of life; (2) pediatric surgical treatment within the first week of the child's life with (3) a hospital stay of one week or more; (4) sufficient knowledge of the Dutch language to complete the questionnaires.

Two fathers did not return the Impact on Family Scale (IFS) and another three fathers did not answer the Perinatal Grief Scale (PGS), leaving 25 couples who completed the IFS and 22 couples who answered the PGS. In addition, 27 mothers filled in the Functional Health Status Scale, Short Form (FS II-R). Eight couples had received foreknowledge from prenatal diagnosis that "something was wrong" with their unborn baby. They had, however, no certainty about the diagnosis until after birth.

Maternal age varied from 21 to 37 years ($N = 27$; $M = 29.2$ years; $SD = 4.0$), and paternal age ranged from 24 to 49 years ($N = 25$; $M = 32.7$ years; $SD = 5.5$). The large majority of the sample was Dutch (83%) and married or living together (87%). More than half of the couples had two or three children (59%), and more than one-third of the couples had one child (37%). The large majority of the fathers (96%) and less than half of the mothers (44%) had paid employment.

The children were between 2 months and 1 year old ($M = 6$ months; $SD = 3.26$); most were male (21 boys versus 6 girls). The majority of the anomalies were in the digestive tract, consisting of congenital obstructions at different levels (56%), for example, oesophageal atresia. Several disorders were in the central nervous system (19%), such as meningomyelocéle, and other disorders (22%) included hygroma colli, hairy pigment naevus, or teratoma. Most children were in a good medical condition at the time of the study. Six children still had medical problems, such as urinary retention, obstipation, or periods of drain dysfunction in the case of meningomyelocéle ($n = 5$) and growth retardation due to absorption problems in the case of cystic fibrosis ($n = 1$).

Procedure

We approached all the 27 parents by mail and a subsequent telephone call. First, the mother or father received a telephone interview. In addition, the mother and the father were requested to complete independently the IFS, the PGS, and only the mother was asked to complete the Functional Health Status Scale about the child.

Measures

To measure parental burden as a consequence of having a child with a congenital anomaly, we used the 24-item IFS (Stein & Riessman, 1980) in a Dutch translation. The scale consists of four subscales: (1) financial burden; (2) social impact refers to problems with social interaction; (3) personal strain re-

fers to maladjustment experienced by the primary care provider, for example, constant fatigue; and (4) mastery refers to coping strategies employed by the parents to master the stress of their child's medical condition. In addition, a Total Burden score was obtained by summing all items. The lower the Total Burden score, the lower the impact of the childs medical condition on the parents. The scale has sufficient reliability and validity for American care providers (Stein & Jones Jessop, 1990). Cronbachs α ranged from .60 to .88, with a median of .81. It is the first time that the scale has been used in the Netherlands. We found, however, quite high correspondence between the intercorrelations of the scales for Dutch and American mothers. For example, reliability coefficients of the Total Burden score for Dutch and American mothers were .80 and .88, respectively, and for personal strain .79 and .81, respectively. The scale was positively correlated with the PGS ($r = .65$, $p < .01$), indicating construct validity. In addition, Stein and Riessman found that for American mothers burden was positively associated with the mother's perception that her child was difficult to care for and that the illness had affected her life.

Grief over the loss of a healthy infant was assessed with the 33-item PGS (Potvin, Lasker, & Toedter, 1989) in a Dutch translation. The scale was adapted for parents who had received the diagnosis of a severe congenital anomaly of their baby before or after birth. The scale consists of three subscales: (1) active grief refers to common emotional reactions following a traumatic event; (2) difficulty with coping refers to more complicated emotional reactions, for example, problems with day-to-day functioning; and (3) despair refers to reactions that reflect the potential for serious psychological problems. The Total Grief score consisted of the summation of all items; the lower the Total Grief score, the lower the level of grief. The PGS showed moderate to high correlations with the Symptom Checklist-90 depression subscale (Toedter, Lasker, & Aldaheff, 1988). The reliability and validity of the scale are good for the Dutch population (Hunfeld, Wladimiroff, & Passchier, 1993). Cronbachs α ranged from .84 to .95.

The perception of the mother about the health of her child was assessed with the 14-item FS II-R (Stein & Jones Jessup, 1990). The scale measures behavioral responses to illness that interfere with normal functioning (i.e., mobility, energy, sleep). The overall FS II-R score can range from 0 (indicating total impairment due to illness) to 100 (no impairment due to illness). Reliability and validity are good for the American population (Stein & Jones Jessup, 1990). The scale showed good discriminative validity for Dutch mothers of asthmatic children. These mothers rated the health of their children significantly lower than mothers of a control group rated their healthy children, and Cronbachs α ranged from .81 to .90 (Post et al., 1998).

Data Reduction and Analysis

Question 1 about the differences between mothers and fathers was analyzed with *t* tests for paired samples on burden and grief. The analysis on burden included 25 couples, because 2 fathers had incomplete burden scores, and the analysis on grief included 22 couples, because 5 fathers had incomplete grief scores. To address question 2 regarding the impact of predictors, ANOVAs with repeated measures were carried out with the between-subject factors, "nature of the diagnosis" (isolated versus multiple anomalies, doctor's report) and "parents" (father versus mother) as independent variables and burden and grief, including all subscale summed scores, as successive dependent variables. In the case of significant group and interaction effects, these were further explored by *t* tests for paired or independent samples. The other independent variables, "prenatal diagnosis" (foreknowledge about the child's anomaly) (yes versus no) and "functional health status" were tested with *t* tests for independent samples. The reasons for this were too small sample size and only one independent variable for functional health status. For functional health status, the children were divided into two groups with a summed score of 85 on the FS II-R as the cutoff point, based on the bimodal distribution of the scores.

We used the Bonferroni procedure to correct for multiple comparisons (Holm, 1979). However, both significant Bonferroni corrected and uncorrected *p* values will be presented, since the Bonferroni correction for multiple comparisons is still under debate (Perneger, 1998).

Results

Power analyses showed that our number of subjects was sufficient for demonstrating significance of a moderate effect size (Cohen's $d = .5$) in related groups (parents) and a large effect size (Cohen's $d > 1$) in unrelated groups (nature of the diagnosis, per-

ception of functional health status, and prenatal foreknowledge).

Parental Burden and Grief

Mothers and fathers showed no differences in the reported Total Burden score (i.e., the total summed score of each subscale sum score) of having a child with a congenital anomaly, nor in the specific subscale sum scores of financial burden, social impact, and mastery (see Table I). However, mothers felt significantly more personal strain than fathers (expressed as "I feel more tired" or "I feel like I am living in extremes, one day happy and the other sad"), $t(24)$[1] = 2,99, Bonferroni $p < .05$, uncorrected $p = .003$. There were no differences in grief between mothers and fathers. Pearson Product Moment Correlation coefficients revealed significantly highly positive relationships between the scores of fathers and mothers for Total Burden ($r = .65$, $p = .001$), social impact ($r = .88$, $p < .001$), personal strain ($r = .82$, $p = .001$), and financial burden ($r = .65$, $p < .001$). The same was valid for total grief ($r = .85$, $p < .001$), active grief ($r = .88$, $p < .001$), difficulty with coping ($r = .82$, $p < .001$), and despair ($r = .76$, $p < .001$). These significant correlations between burden and grief scores within a couple indicated that more burden and grief in the mother is paralleled by relatively more burden and grief in the father. In addition,Total Burden correlated highly positively with total grief in the mothers ($r = .68$), $p < .001$). This was not the case for the fathers ($r = .38$, $p = .12$).

The Impact of the Nature of the Diagnosis, Parents, Health Status, and Prenatal Diagnosis

The ANOVAs with the independent variables nature of the diagnosis (isolated versus multiple anomalies, doctor's report) and parents (father versus mother) showed a significant main effect of nature of the diagnosis on personal strain, $F(1, 23) = 8.50$, Bonferroni $p < .05$, uncorrected $p < .008$. *T* tests for independent samples revealed that this occurred because multiple congenital anomalies involved more burden than an isolated anomaly. Other effects of nature of the diagnosis were in the predicted direction, however, not significant after Bonferroni correction. Uncorrected *p* values were for Total Burden, $F(1, 23) = 4.05$, $p = .05$, social impact, $F(1, 23) = 4.71$, $p = .04$, and despair, $F(1, 20) = 4.73$, $p < .05$. Table II shows the corresponding average scores of burden and grief for the primary care provider, that is, the mother.

Although *t* tests on the impact of functional health status and prenatal diagnosis were not significant after Bonferroni correction, many uncorrected *p* values appeared to be <.05 and in the predicted direction. Mothers who perceived their child's performance as low due to the medical condition reported significantly more Total Burden, $t(25) = 1.76$, $p = .05$, heavier social impact, $t(25) = 1.97$, $p < .04$, and more personal strain, $t(25) = 1.66$, $p = .05$.

One year after the child's birth, the mothers who had been informed prenatally ($n = 8$) about the child's congenital anomaly reported significantly more Total Burden, $t(24) = 1.73$; $p = .05$, and

Table I. Burden of Having a Child with a Congenital Anomaly, Means and *p* Values of the Differences between the Parents

	Mothers ($N = 25$)		Fathers ($N = 25$)	
Measures	***M (SD)***	**Range**	***M (SD)***	**Range**
Total Burden score	45.0 (11.1)	30–60	44.3 (11.4)	29–66
Financial burden	8.2 (3.0)	4–15	8.4 (2.2)	4–13
Personal strain	13.0 (5.0)*	6–21	11.1 (5.0)	6–22
Social impact	15.2 (5.1)	9–28	16.0 (5.3)	9–28
Mastery	8.8 (2.1)	5–13	9.0 (1.3)	6–11

*Bonferroni $p < .05$; uncorrected $p = .003$.

Table II. Nature of the Diagnosis Associated With Burden and Grief, Means and *p* Values of the Differences Between Isolated or Multiple Anomalies, for Mothers

Measures	**Isolated ($N = 16$)** ***M (SD)***	**Multiple ($N = 9$)** ***M (SD)***
Total Burden score	42.1 (9.0)	50.0 (13.0)*
Financial burden	8.1 (3.0)	8.4 (4.0)
Personal strain	11.0 (4.0)	16.0 (4.3)**
Social impact	14.0 (4.0)	18.0 (6.0)*
Mastery	10.0 (2.2)	8.1 (2.2)
	($N = 13$)	($N = 9$)
Total Grief score	49.0 (11.0)	69.4 (34.4)
Active grief	19.0 (6.0)	26.4 (13.4)
Difficulty with coping	16.0 (4.0)	24.0 (14.3)
Despair	14.3 (3.2)	20.0 (8.0)

*Uncorrected $p < .05$.
**Bonferroni corrected $p < .05$; uncorrected $p < .01$.

[1]The differences in *df*s compared to the original sample size numbers (i.e., 25 couples for the IFS, 22 couples for the PGS, and 27 mothers for the FS II-R data) are due to differences in number of missing responses for the different scales. This note applies to all reported *t* tests and ANOVAs.

a heavier social impact, $t(24) = 1.89$, $p < .04$, than mothers without prenatal foreknowledge ($n = 19$). The former mothers also reported significantly more Total grief, $t(20) = 1.83$, $p = .05$, more difficulty with coping, $t(20) = 2.18$, $p < .04$, and more despair, $t(20) = 2.17$, $p = .03$. In addition, they gave significantly lower FS II-R scores, indicating that they perceived their child's performance as low due to his or her medical condition, $t(23) = 1.93$, $p < .03$. The fathers with, compared to fathers without, prenatal foreknowledge showed only a trend in reporting feelings of financial burden, $t(23) = 1.43$, $p = .08$. Post hoc chi-square tests did not show any significant relationship between the nature of the diagnosis (multiple versus isolated anomalies, doctor's report) and prenatal foreknowledge, indicating that the two variables did not contaminate the effect on burden and grief.

Discussion

Parental Differences

Parents experienced the same low impact of financial burden and mastery problems and similar high limitations in social interactions due to time-consuming and energy-consuming care tasks regarding the index child. After Bonferroni correction, mothers still showed more personal strain: however, the mean scores of this subscale did not reveal any major difference between mothers and fathers. Burden and grief were significantly positively correlated in the mothers, but not in the fathers. This possibly reflects the fact that usually the mother spends more time with the child and is more often confronted with problems related to the child's medical condition than the father, who is still the usual breadwinner.

Factors Associated With Burden and Grief

After Bonferroni correction, nature of the diagnosis (isolated versus multiple anomalies) still had a significant effect on personal strain. Having a child with multiple, compared to isolated, congenital anomalies was associated with more burden and a low health status rating given by the mother. The latter was in line with the child's medical condition at the time of this study, as determined by the pediatrician. The majority of chronic medical problems were seen in children with multiple congenital anomalies. Post hoc chi-square tests showed a significantly positive relationship between the pediatrician's report on the child's medical condition and the mother's report on the functional health status. Our findings therefore disagree with what is generally suggested in the literature, namely, that the severity of the child's medical condition may not be crucial to the adjustment of the family.

Although the other tests did not pass this correction for multiple comparisons, many reached uncorrected p value less than .05 in the predicted direction. Since the debate is still going on about the appropriateness of multiple comparisons correction for correlated hypotheses (Perneger, 1998), we will discuss the effects with an uncorrected $p < .05$ here.

Prenatal diagnosis showed the most comprehensive effect on burden, grief, and the child's functional health status as perceived by the mother. Probably this is because prenatal diagnosis stirs up strong emotions of fear and uncertainty about what may happen. In a 4-year follow-up study, many bereaved mothers remembered how they were treated rather than the actual words that were said during the unfavorable prenatal ultrasound diagnosis of a lethal anomaly, which had led to the death of their infant (Hunfeld, Wladimiroff, & Passchier, 1997). This suggests that "the first (earliest) cut is the deepest," particularly for the mother. The view that prenatal diagnosis helps the parents to prepare themselves mentally for the birth of a child with a congenital anomaly is therefore debatable. To optimize the information transmission between the parents and the physician subsequent to prenatal testing, we started a video study to relate factors of the conversation and physician to the knowledge and satisfaction of the parents (Hunfeld et al., in press).

Our group sizes were rather modest. Replication of the positive findings can be recommended. On the other hand, the number of significant results was too large to be explained by Type I errors.

Several studies (Forrest, Standish, & Baum, 1982; Lee & Slade, 1996) showed a positive effect of specific (versus the usual ad hoc) hospital care on the emotional well-being of parents in a situation of threatening pregnancy loss. Our findings suggest that a perinatal counseling team might also be needed for parents following the prenatal diagnosis of a severe fetal anomaly. The obstetrician, pediatrician, consulting specialists, nurses, and a social worker all play an important role. The pediatrician

should help coordinate the activities of the team members. In this way parents can be provided with consistent information about the nature of the diagnosis, the prognosis, the treatment, and instructions for daily care at home. This seems particularly indicated for parents of a child with multiple congenital anomalies that were already detected in the prenatal period.

Received February 12, 1998; accepted October 14, 1998

References

Canning, R. D., Harris, E. S., & Kelleher, K. J. (1996). Factors predicting distress among caregivers to children with chronic medical conditions. *Journal of Pediatric Psychology, 21,* 735–749.

Forrest, G. C., Standish, E., & Baum, J. D. (1982). Support after bereavement. *British Medical Journal, 285,* 1475–1479.

Holm, S. (1979). A simple sequentially rejective multiple test procedure. *Scandinavian Journal of Statistics, 6,* 65–70.

Hunfeld, J. A. M., Leurs, A., de Jong, M., Lesnik-Oberstein, M. S., Tibben, A., Wladimiroff, J. W., Wildschut, H. I. J., & Passchier, J. (in press). Prenatal consultation after a fetal anomaly scan: Videotaped exploration of physician's attitude and patient's satisfaction. *Prenatal Diagnosis*.

Hunfeld, J. A. M., Wladimiroff, J. W., & Passchier, J., et al. (1993). Reliability and validity of the Perinatal Grief Scale. *British Journal of Medical Psychology, 66,* 295–298.

Hunfeld, J. A. M., Wladimiroff, J. W., & Passchier, J. (1997). Prediction and course of grief four years after perinatal loss due to congenital anomalies: a follow-up study. *British Journal of Medical Psychology, 70,* 85–91.

Lee, C., & Slade, P. (1996). Miscarriage as a traumatic event: A review of the literature and new implications for intervention. *Journal of Psychosomatic Research, 40,* 235–244.

Perneger, T. V. (1998). What is wrong with Bonferroni adjustments. *British Medical Journal, 316,* 1236–1238.

Post, M. W. M., Kuyvenhoven, M. M., Verheij, Th. J. M., De Melker, R. A., & Hoes, A. W. (1998). The Dutch Functional Status II (R): a questionnaire to assess functional health status in children. *Ned Tijdschr Geneesk, 142,* 2675–2683.

Potvin, L., Lasker, J. N., & Toedter, L. J. (1989). Measuring grief: A short version of the Perinatal Grief Scale. *Journal of Psychopathology and Behavioral Assessment, 11,* 29–45.

Stein, R. E. K., & Riessman, C. K. (1980). The development of an Impact-on-Family Scale: Preliminary findings. *Medical Care, 18,* 465–472.

Stein, R. E. K., & Jones Jessop, D. J. (1990). Functional Status II–R: A measure of child health status. *Medical Care, 28,* 1041–1055.

Toedter, L., Lasker, J., & Aldaheff, J. M. (1988). The PGS: Development and initial validation. *American Journal of Orthopsychiatry, 58,* 435–449.

Walker, L. S., Ford, M. B., & Donald, W. D. (1987). Cystic fibrosis and family stress: effects of age and severity of illness. *Pediatrics, 79,* 239–246.