Family Burden in Families with a Hemophilic Child*

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The parents of 111 children with hemophilia were interviewed to assess family burden due to the child's chronic condition. A distinction was made between psychological distress and the burden caused by daily practical problems. The latter were perceived as minor burdens by the majority of parents. Scores on the psychological distress scale, however, showed a large variance, and were associated with fear of the occurrence of bleeding, concern about the child's future, and support from the partner in childrearing.

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A CHILD with a chronic illness requires extra care and attention, which may lead to a rearrangement of tasks and responsibilities within the family and, more generally, to psychological distress for the parents. The illness affects family functioning because it may have an impact on daily activities, on the parent-child relation, on other family relations, and on external relations. The influence of a child's chronic condition on family functioning may be explained by the interdependence of roles and behavior that is characteristic of close-knit groups such as the modern Western family. This "system" approach to family functioning was developed by the family sociologists Hansen and Hill (7) for research on families under stress. For chronic illness as a special category of stressful situations, the "system" approach was elaborated by, among others, Bruhn (3), Schwenk and Hughes (23), Penn (19), and Doherty (5).

The reactions evoked by a stressful event or the illness of a family member may differ considerably between families. The ABC-X model, which stresses the importance of "the definition of the situation," accounts for differences in this respect (7): A, the stressful event, interacts with B, the resources the family disposes of when it tries to cope with this stressful event, and C, the

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definition of the event. This results in the outcome X, the family burden or family dysfunctioning. Although several reviews of studies on the consequences of illness for the family advocate the ABC-X model (13, 24, 31), the "definition of the situation," that is, how the family members interpret the disease, is, in this theoretical notion, a rather rudimentary concept. However, central to the theoretical framework is the notion that an illness does not have only an objective medical meaning; it has several subjective meanings, and the family reacts accordingly (11). This means that parents who interpret an illness as a threat will react with anxiety, parents who interpret the illness as a loss will react with feelings of depression, and parents who interpret it as a challenge will react with both feelings of hope and anxiety (24). It follows almost naturally from this approach that illness may also have positive consequences for family functioning, for instance, a positive influence on family solidarity (8, 13).

Family Burden

Hemophilia is an X-linked, hereditary bleeding disease that predominantly affects males. Most mothers of patients are carriers, which may be a source of feelings of guilt toward their child. Hemophilia is characterized by a partial or complete lack of clotting factor VIII or IX activity. Depending upon the residual activity, a distinction is made between severe hemophilia (0–1% of the normal activity), moderately severe (1–5%), and mild (5–40%) hemophilia. Patients with severe and moderately severe hemophilia have spontaneous hemorrhages in the joints and muscles, which in the long run may lead to irreversible damage and disability. In all cases, hemorrhages may occur after trauma and surgery.

Nowadays, bleedings are treated with intravenously administered clotting factor concentrates made out of human blood (substitution treatment). Patients with frequent hemorrhages may receive prophylactic therapy consisting of two or three infusions per week. Many patients are on home treatment. A minority among the hemophiliacs experience problems with this substitution therapy because they develop antibodies against the administered blood products.

The modern replacement treatment was introduced at the end of the 1960s and has led to a considerable improvement of the medical and social situation of hemophiliacs. The life expectancy is approaching that of the general male population (12, 21); sick leave from school and work has decreased dramatically (26, 28); and it is presumed that the generation that has been treated with clotting factor products since childhood will be less disabled than the adult patients of today (26). A serious side effect of the treatment with products prepared from human blood is the possibility of the transmission of viral infections, for example, the hepatitis virus and the human immunodeficiency virus (HIV). Prior to 1985, 17 to 24% of the Dutch hemophiliacs became infected with HIV (20, 30). In 1985, measures were introduced that reduced the risk of HIV infections considerably.

Hemophilia differs from many other chronic conditions in its abrupt character. The bleedings are mostly unexpected and require immediate treatment; they may be life-threatening. The risk of bleedings places the parents of young patients in a dilemma. They want to protect the child as much as possible against falls and other accidents, but they do not want to limit the child at play or hinder the child in the development of its personality.

Several studies have been made of families with a hemophilic child. Browne, Mally, and Kane (2) interviewed parents in the period before modern substitution therapy was available. They describe most mothers as depressed, worried women for whom childrearing was a matter of life and death.

More recent studies present a more positive picture. The majority of families has adapted to the illness within a few years after diagnosis (22) and has learned to accept the inevitability of bleedings (17). Nevertheless, mothers of hemophilic children are reported to suffer more psychological distress than mothers of non-hemophilic children (10). Findings about the influence of hemophilia on the marital relationship differ widely. Salk, Hilgartner, and Granich (22) concluded that in half of the families hemophilia caused a psychological alienation between the husband and the other family members. In other families, the parents felt that the illness had contributed to an improvement of their marriage. Markova, Macdonald, and Forbes (16) found that, if hemophilia influenced the marital relationship, it was a positive influence, though they had doubted the parental claims in this respect. Some fathers go through a period of blaming the mother and withdrawing from childrearing before they become actively involved (17). Markova et al. (16) found that mothers, who had discussed with their partner the risk of hemophilia before the child was born, were more often supported by their husbands in the care of the child than mothers who had not done so. Feelings of guilt were reported by Browne et al. (2) but, according to Madden and colleagues (14), feelings of guilt for most mothers are a minor problem compared with fear of the possible consequences of bleedings. Chilcote and Baehner (4), Simon (25), and Pawlowsky (18) reported on case studies and point to the fact that the mother's feelings of guilt often give rise to overprotective behavior on her part and disengagement on the father's part.

There are some reports of the positive influence of resources. Well-informed mothers show less psychological distress (10). Consulting other parents of hemophilic children, relatives with hemophilia, and professionals, reduces the fear of bleedings and of the consequences of bleedings (14). Home therapy is reported to have been beneficial for family relationships (15).

All the above-mentioned studies are small-scale studies (at most 30 families). The purpose of the present study is to assess in a large number of families the extent to which a child with hemophilia is an extra burden for the family. Using the ABC-X model, we examined the influence of (A) the objective medical situation, of (B) the resources the parents dispose of, and of (C) the psychological reactions of the parents to the illness. Family burden was measured as experienced by the mother. A distinction was made between: 1) burden caused by daily practical problems, that is, disturbance of activities and relations caused by the extra care for the hemophilic child, and 2) psychological distress.

METHODS

Subjects

In 1987, 126 mothers of children with hemophilia in the age group 0 to 12 were asked to participate in the study. Their addresses were taken from a large address file used for a survey carried out in 1985 among the Dutch hemophilia patients. Moreover, large hemophilia treatment centers were asked to report patients born after 1983. One hundred and ten mothers participated in the study; in one family, the father participated because he was primarily responsible for child-rearing. Thirteen families had more than one hemophilic child. In these cases, the parents were asked about their experiences with the youngest child.

Concepts and Instruments

To a large extent, measurement instruments assessing family burden and psychological reactions were derived from Dutch research on families with a handicapped child or a child with a chronic disease (1, 6,

9, 27; see also Table 1 and Table 3). In Likert-format scales, items with an item-total correlation of less than .20 were omitted, and the internal consistency reliability was determined using Cronbach's alpha.

Family Burden

The following areas of daily practical problems were measured: 1) housekeeping and other daily activities, 2) leisure activities, 3) external relations, and 4) holidays. For each of the activities or relations, we asked whether the extra care for the hemophilic child impeded these activities or relations, often, sometimes, or almost never. "Almost never" was rated as 0, "sometimes" as 1, and "often" as 2, after which the total score was calculated.

Psychological distress was measured using six items (see Appendix 1). Three extra items applied only to intact families with two or more children. For that reason they were not included in the scale. They were, however, included in the questionnaire to allow comparison with a control group of another study.

The influence of hemophilia on the marital relationship was assessed by asking whether the partners had grown closer to one another or had grown apart due to the illness of their child.

The Objective Medical Situation

Severity of hemophilia was taken as a measure of the objective medical situation. A distinction was made between severe, moderately severe, and mild hemophilia. Furthermore, a more comprehensive measure for the objective medical situation was developed, consisting of six variables: severity, number of bleedings, antibodies against administered blood products, HIV-seropositivity, hospitalization due to hemophilia, and bed rest due to hemophilia. Therefore, we transformed the raw scores of these variables into *z*-scores. Using the principal components analysis, one underlying dimension appeared, called "the general medical situation." The reliability coefficient (Cronbach's alpha) was .53.

Psychological Reactions

The following psychological reactions were measured: shame, feelings of guilt, concern about the child's future, fear of the occurrence of bleedings, fear of disability, and AIDS-related anxiety. The "AIDS-related anxiety" scale, measuring how often the respondents thought about the possibility that AIDS would manifest itself, had no Likert-format answers. The answer categories were: never, sometimes, regularly, often, and always. Number of items and reliability coefficients are presented in Table 1.

Resources

A distinction was made between personal or family characteristics (educational level, socioeconomic status, family situation, number of children, and support from the partner in childrearing), and illness-related characteristics (home treatment, prophylactic treatment, knowledge about hemophilia, information given by physicians, and occurrence of hemophilia in the family). Family situation refers to whether or not it was a single-parent family. Support from the partner was assessed with the item "I feel that the rearing of this child falls upon my shoulders instead of upon both of us" (9).

Knowledge about hemophilia was measured by asking the respondents whether or not they agreed with 24 statements about hemophilia and the treatment of hemophilia, half of which were incorrect. For every correct answer, a score of 1 was assigned. The average score was 22.

Information given by the physician was assessed by asking which of a number of eleven topics related to hemophilia, the treatment of hemophilia, child-rearing, and

TABLE 1

Scales Measuring Psychological Distress and Other Psychological Reactions: Number of Items, Reliability Coefficient Alpha, and Authors of Items

Scales	Items	Alpha	Author(s)*
Psychological distress	6	.79	S + GG
Psychological reactions			
Shame	2	.38	GG
Feelings of guilt	1	—	V
Concern about the future	5	.68	S
Fear of bleedings	5	.84	V + S**
Fear of disability	2	.84	V
AIDS-related anxiety	6	.77	R

*S = Suurmeijer (27); GG = Gresnigt & Gresnigt-Strengers (6); B = Bos (1); V = Varekamp et al., 1989; R = Rosendaal (20)

**items somewhat rephrased

education had been discussed. Every topic discussed was rated as one point.

RESULTS

General Characteristics

The general characteristics of the population are shown in Table 2. Compared with the total group of Dutch hemophilia patients, there were relatively more children with severe and moderately severe hemophilia (20). One in three mothers with hemophilia in their families had not realized that they were at risk to transmit the disease to their children.

Burden of Daily Practical Problems

Table 3 shows that "almost never" was the most frequent answer to the question of how often activities or relations were disturbed because of the extra care for the hemophilic child. Activities and relations most often disturbed were housekeeping, regularity in the family's schedule of daily activities, sufficient relaxation after work, time for hobbies, going out with the partner, and family holidays. The average score for the total burden caused by daily practical problems was 2.4 (range 0–34; SD = 3.7).

Psychological Distress

The mean psychological distress score was 15.9 (range 6–30; SD = 6). For intact families with two or more children in which the hemophilic child was age 11 or 12, the score of the 9-item version of the psychological distress scale was also assessed to allow comparison with another study that made use of control groups (27). Thirteen families belonged to this category of families.

TABLE 2

General Characteristics of the Survey Population (N = 111)

Family situation	
intact families	93%
single-parent families	7%
Severity of hemophilia	
severe	51%
moderately severe	26%
mild	23%
Hemophilia treatment*	
home treatment	36%
prophylactic treatment	37%
Knowledge about risk of having a hemophilic child	
knew about risk	36%
was ignorant of risk	18%
not applicable; no prior hemophilia in the family	46%

*categories may overlap

TABLE 3

Burden Caused by Daily Practical Problems: Disturbance of Activities and Relations Caused by Extra Care for the Hemophilic Child (n = 110)

	% Disturbance		
	often	sometimes	almost never
Housekeeping, other daily activities			
be finished with the housekeeping	3	20	77
perform household work properly	1	13	86
have meals undisturbed	—	12	88
shopping	—	7	93
regularity in the family's daily activities	1	19	80
a tidy house	2	9	89
design of the house	1	3	96
Leisure activities and recreation			
sufficient relaxation after work	2	19	79
sufficient time for hobbies	3	15	83
doing what you want in the evenings	3	9	88
External relations of parents			
go out together	2	17	81
pay a visit	1	9	90
have visitors	—	4	96
finding a baby-sitter when going out together	5	6	89
participation in club activities	1	5	95
contact with family, neighbors, and acquaintances	—	4	96
Holidays			
go on holiday with the family	6	16	79

*items derived from Gresnigt and Gresnigt-Strengers (6)

The mean score for these families was 24.5 (range 9–45; SD = 5.5).

Marital Relationship

Fifty-two % of the respondents stated that hemophilia had not influenced their marriage; 45% stated that they had grown closer to one another; 4% stated that they had grown apart somewhat because of the illness. These were answers of the respondents who had not been divorced. Four of the nine respondents who had been divorced thought that their child's hemophilia had contributed to their divorce.

Factors Associated With Family Burden

Psychological distress was on average 12.6, 16.5, and 16.9 for mild, moderately severe, and severe hemophilia, respectively ($F = 5.3$; $p = .01$). The burden caused by daily practical problems rose from 1.7, in case of mild hemophilia, to 2.2 in case of moderately severe hemophilia, and to 2.9 in case of severe hemophilia (not statistically significant; $F = 1.0$; $p = .37$).

For three age categories (0–3, 4–10, 11–12), mean psychological distress scores were 17.1, 14.8, and 17.0, respectively, and mean scores for the burden caused by daily practical problems were 3.3, 2.0, and 2.3, respectively. Neither of these differences reached statistical significance ($F = 2.1$; $p = .13$ and $F = 1.2$, $p = .30$, respectively).

Table 4 shows the correlation matrix for variables measuring family burden, and variables measuring the objective medical situation, psychological reactions to hemophilia, and resources, respectively. Psychological distress was more strongly correlated with several psychological reactions than with the severity of hemophilia and

TABLE 4

Correlations between Dependent Variables and Independent Variables

Independent Variables	Dependent Variables: Psychological distress	Dependent Variables: Burden caused by daily practical problems
Objective medical situation		
1. severity of hemophilia	.27**	.14
2. general medical situation	.34**	.08
Resources		
3. educational level	−.00	−.10
4. socioeconomic status	−.01	−.07
5. family situation	−.01	.04
6. number of children	−.10	.16*
7. support from partner in childrearing	−.32**	−.12
8. home treatment	.09	.06
9. prophylactic treatment	.17*	.15
10. knowledge about hemophilia	.05	.06
11. information given by doctors	.03	−.01
12. hemophilia in the family	.11	.05
Psychological reactions		
13. shame	.30**	.26**
14. guilt	.17*	.09
15. concern about the future	.47**	.26**
16. fear of bleedings	.49**	.24**
17. fear of disability	.39**	.19*
18. AIDS-related anxiety	.25**	.12

* = $p < .05$; ** = $p < .01$

the general medical situation. The burden caused by daily practical problems was correlated less strongly with the objective medical situation and the psychological reactions; again, the psychological reactions showed the strongest correlations. Among the "resources," only the support of the partner in childrearing contributed to an alleviation of psychological distress. Some of the "resources" are more easily available in case of more severe hemophilia. For instance, home therapy is for practical reasons not available in case of mild hemophilia. Thus, severity of hemophilia may be a confounding variable affecting the correlation between these resources and psychological distress. However, the relationship between the resources and psychological distress, controlling for severity, yields no statistically significant coefficients, although the sign of the coefficient changed in the expected direction.

The psychological reactions may be mutually correlated. Stepwise multiple regression was used to determine which of the independent variables contributed most to psychological distress. The beta coefficients in Table 5 are the standardized regression coefficients. They indicate how much change in the dependent variable

TABLE 5

*Stepwise Multiple Regression on Psychological Distress**

Independent Variables	beta	*F*	*p*
fear of bleedings	.37	19,08	<.001
concern about the future	.26	8,96	<.001
general medical situation	.24	8,84	<.001

*$R^2 = .39$, $F = 20{,}87$; $p < .001$

psychological distress is produced by a standardized change in the independent variable when the others are controlled. Fear for the occurrence of bleedings contributed most to psychological distress, followed by concern about the child's future and the general medical situation. These three variables account for 39% of the variance. Fear of the occurrence of bleedings was not at all correlated with the number of bleedings per year ($r = -.00$).

Mothers who had discussed the risk of having a hemophilic child with their partners before the child was born, were expected to experience more support from their partner in childrearing than mothers who, knowing about the risk, had not discussed this with their partners. The correlation was, however, not significant, and negative instead of positive ($r = -.23$; $p = .08$; $n = 34$).

The negative correlation of $r = -.28$ between maternal feelings of guilt and perceived support from the partner indicates that the hypothesis that maternal feelings of guilt lead to overprotective behavior of the mother, and to disengagement of the father, may well be true.

CONCLUSIONS

The burden caused by daily practical problems was of minor importance in families with a child with hemophilia. Only a small minority of the parents thought that activities and relations were often hindered. Comparison with studies among families with a mentally handicapped child (6) or a child with a congenital heart condition (1) shows that a mental handicap is much more often felt as a heavy burden, and that hemophilia resembles a congenital heart condition in this respect. We may conclude that hemophilia hardly gives rise to family disturbances in terms of daily practical problems.

The psychological distress showed a larger variance and a relatively higher average score than the burden caused by daily practical problems. Not all psychological distress, however, is attributable to hemophilia. Suurmeijer (27) found an average score of 23.2 among mothers of 11- and 12-year-old epileptic boys, and an average score of 18.6 in a control group, using a 9-item psychological distress scale. In a comparable subgroup of 13 hemophilia families in our study, the average score was 24.5. We may conclude that a considerable group of parents of a hemophilic child, like parents of children with other chronic conditions such as epilepsy, experience more psychological distress than parents of children without a chronic condition.

However, our results compare favorably with those of Browne et al. (2) in 1960, who found that the great majority of mothers of hemophilic patients felt overburdened by the care for their hemophilic son. Modern hemophilia treatment probably has alleviated the burden to a large extent.

If the respondents thought that the quality of their marriage was influenced by their child's hemophilia, they far more often thought that their marriage had changed for the better than for the worse: 45% versus 4%. This corroborates the findings of Markova et al. (16) who found that hemophilia had hardly any detrimental impact, or had brought the parents closer together. However, Salk et al. (22), as well as Pawlovsky (18), found that hemophilia often gave rise to marital or family problems. Several explanations may be given for this inconsistency. Pawlovsky's study is, in fact, an "impressionistic view" of the burden hemophilia may impose on family or marital relations. As this view was based on his experience as a psychologist, a selective bias may have been present in his study. The study of Salk et al. (22) was conducted in a period in which modern replacement therapy was hardly available. In those days, hemophilia may have had more detrimental effects, not only on the patient but also on the family relations.

Furthermore, it is possible that shortly after the diagnosis of a chronic condition, many parents experience marital crises and feelings of being let down, problems that are generally overcome within a couple of years. This is referred to as "the angle of recovery" (7, 29, 31). Because of the small number of families in our survey, in which the diagnosis was made shortly before the study, we could not verify this explanation. Studies about other chronic conditions and literature reviews often mention that the impact of a child's chronic disease on the marital and family relations differs between families, and may be either favorable or unfavorable (1, 5, 8, 13, 29, 31).

In reviews of the literature on the impact of a child's illness on family functioning, it has also been concluded that families vary widely in their response to a chronic illness (23, 24). "A particular disease that affected a family member could enhance a stable family's cohesiveness, disrupt an already dysfunctional family further, or the reverse of either of these, depending on the family's interpretation of the illness event." (23, p 9). From our study, it appears that although the objective medical situation is associated with the psychological distress, the interpretation of the disease in terms of certain psychological reactions often has a greater impact, especially the parental fear of bleedings and the worries about the child's future.

The resources the parents dispose of when coping with hemophilia seldom alleviate distress. The favorable influences of home therapy on family life (15) do not stand out clearly in our study. A possible explanation is that the children who are eligible for these therapies, in view of the frequency of bleedings, actually benefit from them, which makes a proper analysis impossible. A comparison of the situation before and after the introduction of these therapies would have been a more adequate research design. The perceived support from the partner in childrearing is the main resource factor that lessens psychological distress. Consequently, we expected an association between family situation (intact or one-parent families) and psychological distress. This association is absent. It appears that parents are better off without a partner than with a partner who they feel does not support them.

Coming to terms with the fact that a child has a chronic illness or a handicap is for most parents a process without a clear-cut end. Every now and then during the child's development, the parents are confronted with the fact that the child is different from other children, which forces them to make a realistic reinterpretation of the future. Changes in family relations may also lead to recurrent feelings of guilt or shame. This means that there is no one-way causal relation between the C and the X in the ABC-X model, between the psychological reactions and the family burden. Longitudinal studies should be carried out to elucidate the mutual influence of family burden and psychological reactions. In addition, more attention should be paid to the fear of the occurrence of episodes of sudden illness and to the concern about the child's future as main factors causing psychological distress. That this is necessary is shown by the large differences between parents in these respects, the more so since better counseling of the parents may have a positive influence on these concerns (10, 14).

APPENDIX 1

Scale Items Measuring Psychological Distress*

1. Hardly ever does this child bring about stress at home.
2. It is not his fault, but this child puts pressure on me sometimes.
3. There are moments I am not quite able to cope with the situation.
4. Sometimes the care and rearing of this child puts a severe strain on my nerves.

5. Often I am extra concerned about this child.
6. Because of its way of behaving, this child often disturbs the atmosphere at home.

Items Applicable Only to Intact Families with Two or More Children

7. Hardly ever does the rearing of this child give rise to disagreements between the father and the mother.
8. Sometimes the relations between the children are a bit disturbed.
9. The other children are sometimes slightly neglected because of the care of this child.

*derived from Suurmeijer (27)

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