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HEMOPHILIA FROM THE PARTNERS' PERSPECTIVE: BURDEN AND IMPACT ON THEIR LIVES

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This study was conducted to assess hemophilia-related burden in partners and to assess the impact of hemophilia on the partners' lives and on their well-being (i.e. satisfaction, self-esteem and health). The influence of patient and partner characteristics on these outcome variables was also assessed. The study applies to 348 patients and their female partners. Six domains of burden (i.e. problem areas) were distinguished: consequences of hemophilia, social response, virus infections, dependency, coping of patients, and heredity. Most burdening aspects to partners were 'pain in patients' and 'the risk of virus infections due to treatment'. Apart from physical characteristics of patients, the following partner variables added to the prediction of total burden: more caregiving tasks, less optimism, negative social interactions, insufficient information on virus infections and a high perceived risk of AIDS. Hemophilia-related burden seemed to influence many aspects of the partners' lives, in particular the relationship and family life. The partners' satisfaction and health were directly influenced by the patient's health, but there was no additional effect of the hemophilia-related burden or impact on the partners' well-being.

KEY WORDS: Hemophilia, partners, burden, impact, predictors, well-being.

INTRODUCTION

Hemophilia and Partners

Hemophilia is a hereditary clotting disorder which predominantly affects males. The deficiency of clotting factor in the blood (factor VIII in type A, and factor IX in type B) causes hemorrhages, particularly in joints and muscles. Bleeding occurs spontaneously in severe hemophilia, and mildly affected patients experience hemorrhages after trauma, dental procedures, or surgery. Factor concentrates are administered in a hospital or at home by the patient himself, his parents or partner to treat or prevent bleeding. The latter so

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called prophylactic treatment is indicated in severe hemophilia to prevent joint damage. Transmission of viruses (HIV and hepatitis) occurred by treatment with contaminated factor concentrates and had devastating effects on mortality in the hemophilic population (Triemstra, Rosendaal, Smit, Van der Ploeg and Briët, 1995a). Hemophilia can put a serious burden on partners or relatives because of its unpredictable nature, its long-term effects and the treatment-related virus infections.

Few studies have addressed the impact of hemophilia on partners. In the five relevant studies that were found in an extensive literature search on this subject in Medline (1966–1995) and PsycLIT (1974–1995) the number of subjects under investigation was small (14 to 40 partners). The earliest publication on patients with hemophilia and their partners ($n = 14$) reported that the marital relationships tended to have a more demanding and less supportive character than relationships with parents (Bruhn, Hampton and Philips, 1971). The other four studies on partners were conducted after 1980, when the consequences of HIV infections and AIDS became apparent. Psychological variables were studied and no difference in psychological distress of partners was found according to the HIV status of the patient (Dew, Ragni and Nimorwicz, 1991; Klimes, Catalan, Garrod, Day, Bond and Rizza, 1992) or the severity of hemophilia (Klimes *et al.*, 1992). Furthermore, Dew *et al.* (1991) found no difference between psychiatric symptom levels of 36 wives of hemophilic men and those reported for a normative community sample. Klimes *et al.* (1992), however, concluded from their study among 36 female partners of men with hemophilia that partners suffered from higher levels of psychological distress than comparable women in the population. Studies on the consequences of HIV for the relationship showed that sexually active couples worried about sexual transmission of HIV (Agle, Gluck and Pierce, 1987), that changes in the sexual relationship could occur (Agle *et al.*, 1987; Wilson and Wasserman, 1989) and that marriages could become seriously threatened because of AIDS (Wilson and Wasserman, 1989).

Caregiving Burden

Most studies on burden in caregivers have been conducted in the field of gerontology and focused on partners or family members of elderly persons who suffered from dementia or Alzheimer's disease. In general, caregivers are defined as 'primary care providers', often referring to the partner, parent, sibling, or child. Burden can be defined as 'distress in response to caregiver experiences' (Vitaliano, Russo, Young, Becker and Maiuro, 1991a; Vitaliano, Russo, Young, Teri and Maiuro, 1991b).

There is a lack of theoretical cohesion and consistency in the conceptualization and measurement of carer burden (see Miller, McFall, and Montgomery, 1991; Nolan, Grant and Ellis, 1990). Burden has been studied as a one-dimensional and as a multidimensional concept. The unidimensional approach assumes that an additive effect of the problems encountered is more important than the type of problems experienced. But the reliance on summary scores may conceal dimension-specific patterns of caregiving appraisal and impact (George and Gwyther, 1986). For example, different types of impairments may have different types of impact on the caregiver (Poulshock and Deimling, 1984). In order not to mask the relative contribution made by specific domains of burden to the overall burden scores, a multidimensional approach might be useful in evaluating the impact of caring.

Most authors distinguished at least two dimensions of caregiving burden: psychological and physical malaise (Nolan *et al.*, 1990), objective and subjective burden (Carlson and Robertson, 1990; Vitaliano *et al.*, 1991a), personal and interpersonal burden

(Miller *et al.*, 1991), or physical and emotional distress (Nyamathi, Jacoby, Constancia and Ruvevich, 1992).

Poulshock and Deimling (1984) proposed the term burden to be restricted to subjective interpretations of events (e.g. the patient's impairment) and suggested caregiving outcomes (i.e. impact) to be established in terms of 'objective changes' in carers' circumstances. They postulated a three-stage model in which burden acted as an intervening factor between levels of impairment in the dependant and the impact on the carer (Impairment → Burden → Impact). This model will serve as a basis for our study and it will be expanded with factors which are found or expected to influence the consequences of caregiving (see 'Objectives of the study').

Burden Correlates and Outcomes

The social and personal life of caregivers can be highly disrupted as a consequence of caring (Chenoweth and Spencer, 1986; Hooyman, Conyea and Montgomery, 1985) and a change in well-being is likely to occur over time (Evans, Bishop and Ousley, 1992; Given, Stommel, Given, Osuch, Kurtz and Kurtz, 1993). Studies conducted among carers of elderly and handicapped persons often identified a deterioration in emotional health as the most prevalent and pervasive consequence of caring (Bell, Gibbons and Pinchen, 1987; Bowling, 1984; Briggs, 1983; Brody, 1985; Cantor, 1983; Gwyther and George, 1986; Hirschfield, 1983; Parker, 1985; Thompson, 1987; Worcester and Quayhagen, 1983).

Many factors are found to influence the level of burden and adjustment of the caregiver: the patients' physical status, the caregivers' sex, the type of relationship, psychological characteristics of both the patient and the caregiver and psychosocial resources. Relevant findings with respect to these factors are presented below.

According to Miller *et al.* (1991) the impairment and global health of patients affected the level of burden in caregivers, although this effect was rather indirect, operating through stress and involvement. Somatic symptoms in patients were also found to predict caregiver depression, operating through patient depression (Kurtz, Kurtz, Given and Given, 1995). Several authors mentioned that women report higher levels of caregiving stress than men, regardless of the amount of care provided or the level of impairment (Day, 1985; Horowitz, 1985; Horowitz, Silverstone and Reinhardt, 1991; McKinlay and Tennstedt, 1986; Miller *et al.*, 1991; Reis, Gold, Andres, Markiewcz and Gauthier, 1994; Stone, Cafferata and Sangl, 1987). The type of relation with the patient determines the level of involvement, which in turn might influence the perceived stress. Partners report higher levels of stress than other family caregivers, regardless of the level of disability (George and Gwyther, 1986; Miller and McFall, 1989; Miller *et al.*, 1991; Young and Kahana, 1989; Zarit, Reever and Bach-Peterson, 1980). The psychological adjustment of partners to a disease seems to be influenced by the quality or closeness of the relationship (Manne and Zautra, 1990; Spaid and Barusch, 1994) and caregiving duration (Evans *et al.*, 1992). Psychological characteristics of caregivers also seem to play a role in the perception of burden. Depression might result in high levels of burden (Lawton, Moss, Kleban, Glicksman and Rovine, 1991; Pruchno, Kleban, Michaels and Dempsey, 1990; Stommel, Given and Given, 1990) whereas optimism is likely to enhance the caregivers adjustment to the burden of caring (Kurtz *et al.*, 1995). Finally, psychosocial resources of caregivers (e.g. social support) might mediate the effects of impairment on burden (Biegel, Milligan, Putnam and Song, 1994; Miller *et al.*, 1991). A lack of family support and adverse financial consequences could increase the psychological malaise in caregivers (Nolan *et al.*, 1990).

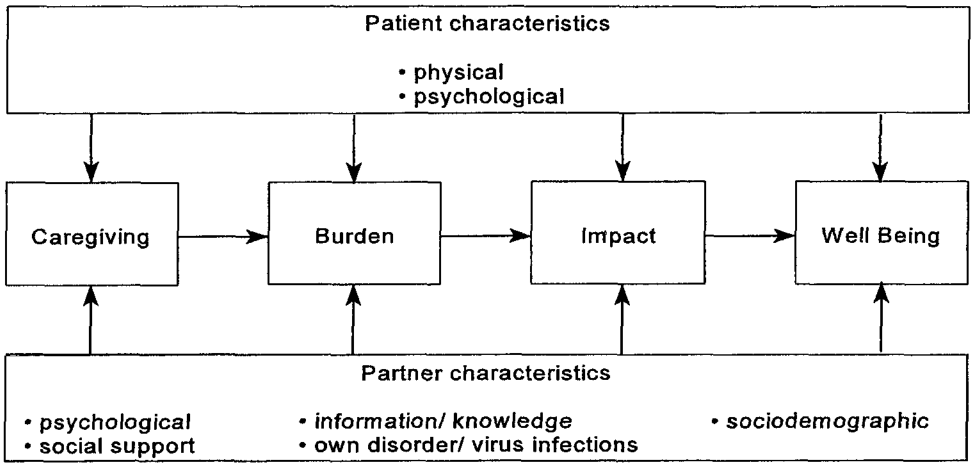

Figure 1 Conceptual model for studying hemophilia-related burden and the impact of hemophilia on partners' lives in association with well-being or other outcome measures in partners.

Objectives of the Study

This paper reports on predictors of hemophilia-related burden in partners and the impact of hemophilia on partners' lives. Studying the process whereby an illness produces a burden on the caregiver should include both patient and caregiver characteristics (Biegel, Sales and Schulz, 1991), together with situational aspects and mediating resources (see Cattanach and Tebes, 1991; Schulz, Tompkins, Wood and Decker, 1987; Vitaliano, Maiuro, Ochs and Russo, 1989; Vitaliano *et al.*, 1991b; Young and Kahana, 1989). Therefore, the concepts 'burden' and 'impact' will be placed within a model which proposes that differences in levels of stressors, personal dispositions and psychosocial resources directly and indirectly influence adverse, stress-related outcomes (cf. House, Umberson and Landis, 1988; Lazarus and Folkman, 1984; Pearlin, Menagham, Lieberman and Muller, 1981; see also Miller *et al.*, 1991; Nolan *et al.*, 1990; Poulshock and Deimling, 1984). The model used in this study is presented in Figure 1.

Patient characteristics are defined as primary antecedents of the perceived problems (i.e. burden) in partners, and burden is expected to be the most relevant predictor of the impact of hemophilia on partners' lives. We will assess the additive contribution of partner variables to the prediction of 'burden' and 'impact' by hierarchical multiple regression. The effects of hemophilia on the partners' well-being (i.e. satisfaction ratings, health and self-esteem) are also evaluated and, finally, predictors of the partners' subjective evaluation of hemophilia (i.e. perceived severity) are assessed.

METHODS

Procedure

A survey among partners of hemophilia patients in 1993 continued a survey among patients in 1992 (Triemstra, Smit, Van der Ploeg, Briët and Rosendaal, 1995b). The participants in 1992 represented approximately 72% of the total hemophilia population in

The Netherlands. Patients who were contacted for the partner study were all men with hemophilia who had participated in the 1992 study, who were then at least 18 years of age (born before 1974), and who had no objection to participation in further research. Of the 663 eligible patients, 580 could be contacted for the partner study, after exclusion occurred for the following reasons: patient had no partner or relative who lived in the same home ($n=47$), patients moved and new address was unknown ($n=18$), patient died ($n=16$), or partner was ill ($n=2$).

Patients were sent a questionnaire with a cover letter directed to their partner or a relative. A total of 411 (71%) partners and significant others completed the questionnaire anonymously and returned it in a prepaid envelope. Because sex and the type of relationship were expected to influence the reported level and content of burden, and in order to obtain a homogeneous study sample, we decided to exclude family members ($n=58$) and male partners ($n=5$) from analysis. The findings of this study are based on data of the remaining 348 male patients and their female partners.

Measures

Patient characteristics. Physical characteristics of patients were assessed with scores for the severity of hemophilia, ADL-dependency and pain. Three levels of severity were distinguished, depending on the clinically assessed residual clotting factor activity in the blood: 1 = mild hemophilia (5–40% of normal activity); 2 = moderately severe hemophilia (1–5%); 3 = severe hemophilia (less than 1% of normal activity). An Activities of Daily Living (ADL) index was used to assess the degree of functional impairment. The index was specifically composed for this study and highly comparable with the OECD indicator for long term disability (McWinnie, 1982). Nine tasks, which referred to self-care, domestic activities, or outdoor activities (e.g. dressing, climbing stairs, shopping, walking long distances), were rated on a 5-point scale of dependency: 1 = without difficulty; 2 = with some difficulty; 3 = with much difficulty; 4 = only with assistance; 5 = does not perform, total dependency. Sum scores were calculated, with a high score indicating a high level of disability and dependency. Cronbach's alpha for the ADL-scale in this study was 0.93. The frequency in which patients experienced pain due to hemophilia was assessed with a 5-point scale ('never' to 'very often').

Patient and partner characteristics. The following variables were assessed in patients and partners: subjective health, HIV status, psychological characteristics, educational level and social participation. Subjective health was assessed with the single item 'How is your health overall?' (1 = excellent; 5 = poor). Answers were reversely scored. With respect to HIV, patients and partners were asked to indicate whether they were tested (0 = no, 1 = yes) and what the test result was (0 = seronegative, 1 = seropositive). Both patients and partners were asked to fill out scales of a personality inventory (Dutch Self-Assessment Questionnaire-Nijmegen, Van der Ploeg, 1989), representing Dutch adaptations of Spielberger's Trait Anxiety Inventory (1983; 20 items), the Trait Anger Scale of Spielberger, Jacobs, Russell and Crane (1983; 20 items), Zung's Self-rating Depression Scale (1965, 1973; 10 items) and the Life Orientation Test (Scheier and Carver, 1987; 8 items). Answers were rated on a 4-point scale, ranging from 'almost never' to 'almost always'. The sum score for each subscale was computed, with low scores indicating a low frequency of the affect. Validity and reliability estimates for the Dutch scales were previously reported to be satisfactory or good (Mook, Kleijn and

Van der Ploeg, 1989; Van der Ploeg, Defares and Spielberger, 1980; 1982). Cronbach's alphas found in this study for the patient and partner group were respectively 0.92 and 0.91 for the anxiety scale, 0.90 and 0.86 for the anger scale, 0.80 and 0.78 for the depression scale, and 0.84 and 0.86 for the optimism scale. The educational level referred to the highest level attained (1 = primary school, 5 = academic training). With respect to social participation, patients and partners were divided into 3 categories, depending on their answers to a question on their main daily activity: 1 = not employed; 2 = part-time employed/volunteer; 3 = full-time employed/volunteer.

Partner characteristics. Variables which were assessed only in partners concerned the categories 'caregiving', 'social support', 'information/knowledge', 'own disorder and virus infections', and 'sociodemographic characteristics'. Outcome variables in partners were: burden, impact, indicators of well-being (i.e. satisfaction, self-esteem and subjective health), and their perceived severity of hemophilia.

Caregiving. Partners were asked if and in which respects they considered themselves as a 'caregiver' of the patient. Caregiving tasks concerned: assistance with treatment of hemophilia or daily activities, housekeeping/domestic activities, supporting the patient, understanding the patient, providing advice and information. Sum scores were computed to express the number of caregiving tasks. If none of the answering alternatives did apply to the partner, they could answer 'I never perceive myself as his caregiver' and the variable 'Perception as caregiver' was scored as '0' (else '1' = yes). Partners were also asked whether they provided help with ADL tasks (listed in the ADL-index), and how often they provided help to the patient (1 = never, 2 = sometimes, 3 = often or always).

Social support. Partners could choose from 12 social categories to indicate with whom they talked about hemophilia: patient, parents (in-law), siblings, children, extended family, friends, acquaintances/neighbours/colleagues, members of the Hemophilia Society, health care providers, or others. Another question, with nine answering categories, assessed those from whom the partner received most support in coping with hemophilia or its related concerns. Partners could choose more than one social category or indicate an alternative answering category ('no interest' or 'no support'). Sum scores were calculated to express the total number of categories involved.

To measure recently received support, a Dutch adaptation of the Social Support Questionnaire (SSQ, Revenson, Wollman and Felton, 1983) by Tempelaar, De Haes, Van den Heuvel, Van Nieuwenhuijzen and Pennink (1987) was used. Partners were asked to rate positive and negative social interactions that occurred in the preceding week. Eight positively worded items referred to experiences that provided support, affirmation or aid, and 8 items described non-supportive interactions. The answering format was a 4-point frequency scale running from 'never' to 'often'. In this study, the reliability of both subscales proved to be good (Cronbach's $\alpha = 0.87$ and 0.79).

The Marital-scale (10 items) of the Maudsley Marital Questionnaire (MMQ, Crowe, 1978; Dutch version: Arrindell, Boelens and Lambert, 1983a) was used to measure the quality of the emotional bond. The answering format was a 9-point scale (0–8). The MMQ was designed to assess the degree of favourableness of attitude towards one's own marriage. The scale demonstrated to have a high internal consistency (Cronbach's $\alpha = 0.90$) and sufficient test-retest reliability and validity (Arrindell *et al.*, 1983a; Arrindell, Emmelkamp and Bast, 1983b; Arrindell and Schaap, 1985). A total score was computed, with high scores indicating a high level of relationship problems.

Information/knowledge. Partners were asked whether they received sufficient information about AIDS or hepatitis. Answers ranged from 'not at all sufficient' (1) to 'amply sufficient' (4). Their knowledge of hemophilia and heredity was rated on scales ranging from 'nothing' (1) to 'much' (5).

Own disorder and virus infections. Partners were asked whether they suffered from a chronic disease, disorder or handicap (0 = no, 1 = yes). Also, apart from a question on being tested for HIV, they were asked to indicate whether they perceived their risk of AIDS or hepatitis as being 'lower' (1), 'equal' (2) or 'higher' (3) than comparable others in the general population.

Sociodemographic characteristics. Apart from the educational level and social participation (see 'Patient and partner characteristics'), partners were asked about their age, the duration of their relationship and number of children.

Burden. To measure the extent of burden, a list was employed giving 19 potential problems partners might face with respect to hemophilia. The problem items concerned hemophilia treatment; characteristics of hemophilia (i.e. hemorrhages, disability, pain, heredity); the risk of virus infections and transmission; dependency on partner, other persons, health care providers or financial resources; social and medical contacts; the partners' incapacity to pay enough attention to hemophilia; and coping of the patient with hemorrhages, virus infections or increasing levels of joint impairment. Partners were asked to consider if they experienced a particular problem (i.e. stressor, related to hemophilia) in their present situation and, additionally, to indicate the degree of actual stress they perceived each stressor to provoke (1 = not at all/ not applicable; 4 = extremely). For each respondent a total score was calculated to express the level of burden. The reliability of the burden scale was good (Cronbach's $\alpha = 0.93$).

Apart from a summary score, we distinguished six meaningful domains of burden: (1) *Consequences of hemophilia*: physical deterioration and caregiving demands (6 items, $\alpha = 0.80$); (2) *Social response*: misunderstanding, adverse reactions and lack of knowledge (3 items, $\alpha = 0.82$); (3) *Virus infections*: sexual transmission, infections due to treatment, coping of patient (3 items, $\alpha = 0.66$); (4) *Dependency on external resources*, i.e. others, professional caregivers, formal services, financial resources (4 items, $\alpha = 0.65$); (5) *Coping of patient* with hemorrhages and joint impairment (2 items, $\alpha = 0.64$); (6) *Heredity of hemophilia* (1 item). Pearson's intercorrelations of the burden domains ranged between 0.12 and 0.52.

Impact. The impact on partners was indicated by an index of influence. Partners were asked to indicate in what way hemophilia affected the following areas of their lives: (1) *Relationship with patient*, (2) *Family life*, (3) *Social contacts*, (4) *Daily activities*, (5) *Leisure*, (6) *Activities together*, (7) *Partner's health*, (8) *Attention for own problems*, (9) *Sexual relationship*. Answers were coded: 0 = no influence/not applicable, 1 = positive, 2 = positive and negative, 3 = negative. A total score was computed to obtain an overall measure of impact, with high scores indicating a more pervasive and negative influence of hemophilia. Cronbach's alpha for the impact scale was 0.89. The impact items showed, in general, moderately strong associations (Pearson correlations varied between 0.30 and 0.62), except for the association between leisure and activities together ($r = 0.79$).

Well-being. Partners were asked to rate their satisfaction 'in general' with nine aspects of their lives: social contacts, partner/relationship/family life, leisure, daily activities, financial and social status, physical condition, friends and acquaintances, society, their housing and life as a whole. The response format ranged from 'extremely satisfied' (1) to 'rather unsatisfied' (5). Cronbach's alpha for the scale was 0.86.

Self-esteem, i.e. a favourable attitude towards oneself, was measured with Rosenberg's Self Esteem Questionnaire (Rosenberg, 1965; 9 items). A Dutch translation of the scale was obtained from Sanders (1977), and, in this study, the internal consistency of the scale was satisfactory (Cronbach's $\alpha = 0.87$).

The subjective health of partners was used as a third component of well-being (see 'Patient and partner characteristics').

Perceived severity of hemophilia. Partners were asked to evaluate the severity of hemophilia (1 = not serious at all, 5 = very serious disorder).

Data Analyses

Statistical analyses were conducted with SPSS 5.0 (Norusis, 1992). Hierarchical multiple regression analyses were employed to assess predictors of burden, impact and other outcome scores. Independent variables were entered in blocks. Within each block the order of entry was determined by stepwise regression (p-criteria for entry and removal limits were respectively 0.05 and 0.10). Model fit was evaluated by the adjusted R square. Pairwise deletion of missing data was applied.

In predicting burden and impact, patient characteristics were entered first in the analyses. This hierarchy was based on the logical consideration that, in general, patient characteristics rather than partner variables are better predictors of hemophilia-related burden and impact scores. The order of entry enabled us to determine if partner variables improved prediction of burden and impact after differences in patient characteristics had been statistically eliminated. To assess the specific contribution of burden and impact on outcomes in partners, the total scores of burden and impact were entered as a final block in the regression model.

RESULTS

Mean age of the 348 female partners in 1993 was 41 years (range 18–76). Patients were on average 3 years older than their partners. The mean duration of the relationship was 20 years (range 1–58). Almost all of the couples (97%) lived together, 83% ($n = 288$) were married, and the mean number of children was 1.5 (range 1–10). 89% ($n = 309$) of the patients had hemophilia A (deficiency of clotting factor VIII) and 11% ($n = 39$) had hemophilia B (deficiency of factor IX). The overall distribution by severity of hemophilia was 40% ($n = 138$) with severe hemophilia, 15% ($n = 53$) with moderately severe hemophilia and 45% ($n = 157$) with the mild form. A total of 20 patients were infected with HIV due to treatment with contaminated clotting factor products before 1985. One partner reported being seropositive. In general, partners of patients with mild hemophilia reported a better health status: 86% had an excellent or good health while this percentage was 79% in case of severe or moderately severe hemophilia.

Caregiving most often consisted of understanding (44%) and supporting (37%) the patient, followed by providing practical help (21%) and help with treatment (21%). 134 partners (39%) said they provided assistance with ADL, and 10% ($n = 36$) of the partners reported helping patients 'often or always' with ADL tasks. Helping patients with common activities was most frequently mentioned ($n = 103$).

Table 1 presents correlations between patient characteristics and partner characteristics for 348 couples. The patients' physical status (i.e. severity of hemophilia, ADL and pain) showed moderately strong associations with characteristics of caregiving in partners, and weaker correlations with information/knowledge of hemophilia-related issues and social support in partners. The psychological characteristics of patients and partners were weakly related, except for anger in partners which was not significantly associated with any of the patient characteristics. All other clusters of variables in Table 1 were weakly or not significantly interrelated.

Table 2 shows correlations of patient and partner characteristics with the outcome measures under study. Strongest correlates of total burden and impact scores were the physical status of patients and caregiving characteristics in partners. Satisfaction and self-esteem, as indicators of well-being in partners, were most strongly associated with psychological characteristics of partners and the quality of the relationship. Self-perceived health in partners was strongest related to having a disorder themselves and their level of anxiety and depression.

Table 3 shows Pearson intercorrelations of domains of burden and impact (subscale and one-item scores, respectively). The total index scores were moderately strong correlated ($r = 0.58$).

In Table 4, results of hierarchical multiple regression analyses are shown, with total scores of burden and impact and outcomes of well-being in partners as criterion variables (maximum $n = 344$). The explained variance varied from 50% for the impact of hemophilia on the partners' lives to 42% for the partners' satisfaction and health.

Burden

Partners of patients with severe or moderately severe hemophilia were more likely to be burdened. A mean of 8.0 problems was reported in cases of a severe form, while partners of patients with mild hemophilia reported an average of 4.8 problems. The total mean was 6.5 problems. The total problem score (i.e. burden) showed a curvilinear association with the severity classification. The mean score for burden was 31.1 in partners of patients with severe hemophilia, 32.6 in case of moderately severe hemophilia and 26.6 in case of the mild form. The total mean for burden was 29.4 (SD 8.1, range 19–61). The five most intense ('rather extreme' to 'extreme') problems mentioned by partners were: pain (39%), virus infections (33%), heredity (29%), increasing level of impairment (24%), and occurrence of hemorrhages (23%).

Total burden was best predicted by pain in patients ($\beta = 0.40$), number of caring tasks ($\beta = 0.22$) and the perceived risk of AIDS ($\beta = 0.18$) (Table 4). Severity of hemophilia showed a negative beta weight ($\beta = -0.16$), although its correlation with burden was positive (Pearson $r = 0.26$, Table 2).

Multiple regression analyses with burden domains entered as criterion variables showed the contribution of patient and partner characteristics to specific problem areas. Burden regarding the consequences of hemophilia was predominantly predicted by pain in patients ($\beta = 0.34$), frequency of caregiving ($\beta = 0.21$) and anxiety in partners ($\beta = 0.17$). 'Social response' was most strongly associated with the negative social interactions experienced by partners ($\beta = 0.29$), their level of education ($\beta = -0.20$) and pain in patients ($\beta = 0.18$). Burden regarding virus infections was mainly associated with the partners' perceived risk of hepatitis ($\beta = 0.34$), the severity of hemophilia ($\beta = -0.26$) and the patients' HIV status ($\beta = 0.24$). 'Dependency' was more burdening in case of

Table 1 Pearson correlations[a] between characteristics of 348 hemophilia patients and characteristics of their partners

	Patient characteristics									
	Physical						*Psychological*			
	Severity classification	*Perceived severity*	*ADL-score*	*Pain*	*HIV status*	*Health*	*Anxiety*	*Anger*	*Depression*	*Optimism*
Partner characteristics										
Caregiving										
No. caring tasks	0.39	0.40	0.44	0.37	(−0.01)	−0.13	(0.06)	(−0.04)	(0.2)	(−0.09)
No. ADL tasks	0.45	0.37	0.70	0.45	(−0.01)	−0.16	(0.10)	(−0.02)	(0.11)	(−0.08)
Frequency	0.47	0.44	0.64	0.50	(0.04)	−0.19	0.20	(0.06)	0.16	−0.14
Perception as a caregiver	0.28	0.29	0.31	0.29	(−0.05)	−0.11	(0.04)	(−0.03)	(0.01)	(−0.08)
Psychological										
Anxiety	(0.03)	0.13	(0.03)	0.12	(0.02)	−0.18	0.24	0.15	0.21	−0.18
Anger	(−0.01)	(0.05)	(−0.07)	(0.07)	(−0.07)	(−0.04)	(−0.02)	(0.03)	(−0.02)	(0.03)
Depression	(0.03)	0.14	(0.06)	(0.08)	(0.05)	−0.21	0.24	0.20	0.23	−0.21
Optimism	(−0.07)	−0.17	(−0.07)	−0.13	(−0.02)	0.19	−0.22	(−0.11)	−0.22	0.25
Social support										
No. talking categories	0.24	0.18	(0.10)	0.26	(0.04)	(−0.08)	(0.03)	(−0.05)	(0.05)	(−0.02)
No. support categories	0.27	0.23	0.22	0.29	(0.10)	−0.16	(0.06)	(−0.09)	(0.10)	(−0.06)
Positive interactions	0.15	(−0.01)	(−0.00)	(0.03)	(0.11)	0.17	−0.13	−0.14	−0.16	(0.09)
Negative interactions	(0.07)	0.19	0.06	0.15	(0.00)	−0.16	(0.11)	(0.09)	0.16	(−0.04)
Relationship problems	(0.04)	0.11	0.13	0.12	(0.04)	−0.14	0.19	0.21	0.15	−0.15

Information/knowledge										
Hemophilia	0.47	0.33	0.37	0.39	(0.09)	(−0.07)	(0.02)	(0.02)	(−0.01)	(−0.02)
AIDS	0.23	0.15	0.15	0.13	(−0.02)	(0.06)	−0.13	−0.14	(−0.11)	0.14
Hepatitis	0.33	0.18	0.22	0.26	(−0.00)	(−0.01)	(−0.10)	−0.13	−0.15	0.17
Heredity	0.37	0.21	0.26	0.31	(0.02)	(0.02)	(−0.01)	(0.01)	(−0.07)	(0.05)
Own disorder/virus infections										
Own disorder	(0.10)	0.15	0.17	0.12	(−0.11)	(−0.02)	(−0.01)	(0.04)	(−0.02)	(−0.02)
Tested for HIV	0.22	0.13	0.12	0.22	0.23	(0.01)	(−0.10)	−0.14	(−0.06)	(0.06)
Perceived AIDS risk	0.16	0.14	(0.07)	0.19	0.21	(0.03)	(0.10)	(−0.00)	(0.09)	(−0.05)
Perceived hepatitis risk	0.24	0.17	(0.10)	0.23	(−0.09)	(−0.02)	(0.10)	(0.02)	(0.07)	(−0.04)
Sociodemographic										
Age	−0.18	(0.01)	0.21	(−0.08)	−0.18	(−0.06)	(0.04)	(0.03)	0.11	(−0.09)
Relationship duration	−0.18	(0.01)	0.13	(−0.06)	−0.17	−0.11	(0.08)	(0.04)	0.16	−0.12
No. children	(−0.05)	(−0.02)	(0.06)	(−0.06)	−0.08	(0.08)	(0.01)	(−0.02)	(0.08)	(−0.10)
Education level	0.16	0.12	(−0.09)	0.12	(0.11)	0.15	(−0.08)	(−0.03)	−0.18	0.13
Social participation	0.11	(0.03)	(−0.07)	(0.04)	0.20	0.13	(−0.02)	(−0.02)	(−0.07)	(0.10)

[a] Correlations in parentheses are not significant ($p > 0.05$).

Table 2 Pearson correlations[a] between characteristics of 348 couples and outcomes in partners

	Partner outcomes				
	Burden	*Impact*	*Satisfaction*	*Self-esteem*	*Health*
Patient characteristics					
Physical					
Severity classification	0.26	0.42	(−0.00)	(−0.05)	(−0.07)
Perceived severity	0.42	0.44	−0.13	(−0.11)	−0.12
ADL-score	0.31	0.49	−0.16	(−0.02)	−0.13
Pain	0.49	0.49	−0.11	(−0.07)	−0.15
HIV status	(−0.00)	(0.05)	(−0.01)	(0.03)	0.15
Health	−0.21	−0.25	0.27	(0.08)	0.20
Psychological					
Anxiety	0.14	0.25	−0.25	−0.20	(−0.10)
Anger	(0.02)	(0.07)	−0.14	−0.13	(−0.09)
Depression	0.16	0.20	−0.24	−0.14	−0.11
Optimism	−0.13	−0.22	0.21	0.17	(0.10)
Partner characteristics					
Caregiving					
No. caring tasks	0.40	0.49	−0.14	−0.12	(−0.04)
No. ADL tasks	0.35	0.45	−0.14	(−0.07)	−0.11
Frequency	0.42	0.53	−0.18	−0.14	(−0.09)
Perception as a caregiver	0.33	0.43	−0.11	−0.15	(−0.04)
Psychological					
Anxiety	0.30	0.23	−0.57	−0.67	−0.35
Anger	(0.10)	(0.00)	−0.27	0.27	−0.12
Depression	0.24	0.21	−0.53	−0.61	−0.36
Optimism	−0.32	−0.28	0.47	0.63	0.27
Social support					
No. talking categories	0.20	0.24	(0.04)	(0.06)	(0.04)
No. Support categories	0.24	0.30	(−0.04)	(−0.01)	(−0.10)
Positive interactions	(−0.08)	(−0.01)	0.30	0.19	0.12
Negative interactions	0.28	0.22	−0.22	−0.17	−0.17
Relationship problems	0.21	0.26	−0.51	−0.43	−0.19
Information/knowledge					
Hemophilia	0.24	0.35	(−0.03)	0.13	(−0.01)
AIDS	(−0.10)	(0.05)	0.13	0.15	0.11
Hepatitis	(−0.06)	0.15	(0.10)	0.13	(0.04)
Heredity	(0.08)	0.19	(0.06)	0.16	(−0.02)
Own disorder/virus infections					
Own disorder	0.16	0.13	−0.17	(−0.10)	−0.57
Tested for HIV	0.14	0.17	(0.00)	(0.03)	(0.10)
Perceived AIDS risk	0.30	0.24	(0.00)	(0.00)	(0.09)
Perceived hepatitis risk	0.26	0.27	(0.05)	(0.05)	(0.03)
Sociodemographic					
Age	(−0.01)	(0.08)	(−0.07)	(0.07)	−0.15
Relationship duration	(0.01)	(0.08)	(−0.03)	(0.06)	(−0.07)
No. children	(−0.07)	(0.01)	(−0.04)	(−0.01)	(0.10)
Education level	(−0.04)	(−0.03)	0.13	(−0.01)	(0.07)
Social participation	(−0.01)	(0.00)	(0.09)	(0.04)	0.20

[a] Correlations in parentheses are not significant ($p > 0.05$).

Table 3 Pearson correlations[a] of burden and impact domains and total scores ($n_{\max} = 340$)

	Impact (items)									
	Relationship	*Family life*	*Social contacts*	*Daily activities*	*Leisure*	*Activities together*	*Own health*	*Attention for own problems*	*Sexual relation*	*Total score*
Burden (subscales)										
Consequences of hemophilia	0.49	0.52	0.25	0.51	0.49	0.51	0.26	0.33	0.31	0.57
Social response	0.25	0.27	0.40	0.25	0.27	0.28	0.29	0.33	0.19	0.38
Virus infections	0.30	0.30	0.24	0.21	0.23	0.23	0.21	0.17	0.30	0.33
Dependency	0.25	0.35	0.32	0.29	0.25	0.28	0.18	0.27	0.19	0.35
Coping of patient	0.27	0.24	0.20	0.30	0.24	0.25	0.22	0.19	0.16	0.31
Heredity	0.15	0.20	(0.07)	0.15	0.16	0.15	(0.07)	0.14	(−0.00)	0.16
Total score	0.47	0.49	0.38	0.46	0.46	0.47	0.32	0.37	0.34	0.58

[a] Correlations in parentheses are not significant ($p > 0.05$).

Table 4 Predictors of burden, impact and outcome measures of well-being in partners of patients with hemophilia. Results[a] of hierarchical multiple regression analyses (β weights and adjusted R squares)

	Partner outcomes				
	Burden ($n_{max}=305$)	*Impact* ($n_{max}=327$)	*Satisfaction* ($n_{max}=328$)	*Self-esteem* ($n_{max}=326$)	*Health* ($n_{max}=344$)
Patient characteristics					
Physical					
Severity classification	−0.16				
Perceived severity	0.15	(0.02)			
ADL-score		(0.14)			
Pain	0.40	(0.03)			(0.11)
HIV status			0.13		0.15
Health					
Psychological					
Anxiety			(−0.05)	(−0.04)	
Optimism		(−0.08)			
Partner characteristics					
Caregiving					
No. caring tasks	0.22				
Frequency		0.17			
Perception as a caregiver		0.14		(−0.09)	
Psychological					
Anxiety			−0.37	−0.46	
Depression					−0.22
Optimism	−0.15	(−0.09)		0.23	
Social support					
Positive interactions			0.16		
Negative interactions	0.14				
Relationship problems			−0.25		
Information/knowledge					
Hemophilia		(0.10)		0.13	
Hepatitis	−0.15				
AIDS					
Own disorder/virus infections					
Own disorder					−0.50
Perceived AIDS risk	0.18				
Perceived hepatitis risk		0.11			
Sociodemographic					
Social participation					0.11
Burden (total score)	—	0.30			
Adjusted R square	0.44	0.50	0.42	0.47	0.42

[a] Only variables selected by stepwise regression, within blocks, are shown. Standardized regression coefficients in parentheses are not significant ($p > 0.05$).

more caregiving tasks ($\beta=0.22$) and more negative social interactions of partners ($\beta=0.18$), more pain in patients ($\beta=0.18$) and less information received by partners about AIDS ($\beta=-0.16$). Coping of patients was perceived as more problematic when patients suffered from more pain ($\beta=0.24$) and partners were less optimistic ($\beta=-0.17$). Seropositivity for HIV seemed to reduce burden regarding 'consequences of hemophilia' ($\beta=-0.14$) and 'coping of the patient with hemophilia' ($\beta=-0.22$).

Impact

Of all life areas, the relationship was most likely to be positively influenced by hemophilia (in 16% of the partners). The four most (negatively) affected areas of the partners' lives were: activities together, leisure, family life and the relationship with the patient. If the answer categories 'positive and negative influence' and 'negative influence' were taken together, 41% of the partners seemed to experience a negative influence on the activities together. Likewise, 35% of the partners seemed to experience a negative effect on leisure, 27% on family life and about one quarter of all partners experienced a negative (as well as positive) influence on their relationship. A majority of partners reported no influence of hemophilia on their health (75%) or social contacts (72%). On average, the answering category 'no influence at all' was most frequently mentioned (for 5.5 out of 9 life areas), followed by the answer that indicated a positive and negative influence of hemophilia (1.7 times).

The total impact of hemophilia on the partners' lives was predominantly predicted by the burden experienced ($\beta = 0.30$), followed by the number of caregiving tasks performed by the partner ($\beta = 0.17$), the partners' perception as a caregiver ($\beta = 0.14$) and their perceived risk of hepatitis ($\beta = 0.11$) (Table 4).

Hemophilia-related burden particularly seemed to influence family life, the relationship and activities with the patient, daily activities and leisure (see Table 3). The consequences of hemophilia particularly seemed to have an impact on family life ($r = 0.52$) and activities ($r = 0.51$). Burden regarding social response could influence the partners' social contacts ($r = 0.40$). Virus infections could have an effect on family life and the sexual relationship ($r = 0.30$). The dependency and coping of patients may influence family life, the relationship, social contacts and activities (r between 0.35 and 0.24). Furthermore, heredity of hemophilia could have an impact on family life ($r = 0.20$).

Well-being and Perceived Severity

Predictors of partners' well-being (i.e. satisfaction ratings, self-esteem and health) were also assessed by hierarchical multiple regression analysis (Table 4). Satisfaction ratings in partners were predominantly influenced by their general level of anxiety ($\beta = -0.37$), followed by relationship problems ($\beta = -0.25$), positive social interactions ($\beta = 0.16$) and the subjective health of the patient ($\beta = 0.13$). Self-esteem in partners depended on their level of anxiety ($\beta = 0.46$), their optimism ($\beta = 0.23$) and their knowledge of hemophilia ($\beta = 0.13$). Strongest predictors of the partners' self-reported health were the presence of a disorder ($\beta = -0.50$) and depression in partners ($\beta = -0.22$). Also, the patients' health and the partners' social participation were positively associated with the partners' health ratings. After characteristics of patients and partners were added to the regression model, burden and impact scores seemed to have no significant contribution to the prediction of satisfaction, self-esteem and health in partners.

The subjective evaluation of hemophilia by partners was most strongly predicted by their perceived burden ($\beta = 0.22$), followed by the patients' perceived severity ($\beta = 0.19$), the frequency of caregiving ($\beta = 0.15$) and the partners' knowledge of hemophilia ($\beta = 0.14$). In general, partners evaluated hemophilia as more severe than did patients (40% of the partners and 19% of the patients rated hemophilia as a serious or very serious disorder).

DISCUSSION

We defined 'burden' as the amount and intensity of problems experienced by partners in response to hemophilia, and distinguished six domains of burden. Our results indicated that different patient and partner characteristics related to different burden domains. In addition we found that each area of partners' lives was influenced to a different extent these characteristics, with specific hemophilia-related burden adding to the prediction of impact on their lives, and that hemophilia appeared to have no direct influence on the partners' well-being.

Pain in patients was the most prominent problem in partners, and, of all patient characteristics, this variable showed the strongest association with total burden and impact in partners (Table 2). Of all partner characteristics, caregiving variables were most strongly related to the total burden and impact scores (Tables 2 and 4). Obviously, the more mental and practical support was provided, the greater the burden partners experienced. Also, the more frequent care tasks were conducted and the more partners perceived themselves as caregivers, the stronger the impact on the partners' lives. Reducing pain or helping patients to cope with pain would probably decrease burden in partners. Enhancing communication skills in couples who have to deal with pain might be useful, as partners may have an important role in how patients cope with their pain (see Kerns, Haythornthwaite, Southwick and Giller, 1990).

In general, psychological characteristics of partners appeared to play a prominent role in predicting burden and outcomes of well-being in partners, but psychological characteristics of patients appeared to be of little of no importance (Table 4). The partners' anxiety was most apparently related to dealing with the consequences of hemophilia and virus infection, and their optimism seemed to determine whether a patients' coping behaviour was viewed as problematic. Partners who perceived themselves at higher risk of hepatitis (a variable closely related to their perceived AIDS risk, $r=0.59$) experienced higher burden regarding virus infections. Reducing their risk perception, for example by providing sufficient information, would probably reduce this burden. As negative social interactions were associated with high burden regarding 'social response' and 'dependency on external resources', these adverse social reactions are likely to contribute to feelings of stigmatization or isolation in partners. If partners and people around them (e.g. family, friends, neighbours, colleagues), however, receive sufficient information about hemophilia, partners are less likely to feel stigmatized.

Contradictory results in the prediction of burden were found for the classified severity of hemophilia and HIV status. The negative beta weight for severity of hemophilia was due to a curvilinear association of this variable with burden (in particular, virus-related burden). Partners of men with moderately severe hemophilia showed the highest total burden score, although the amount of problems reported was equal to partners of patients with severe hemophilia. If we understand adjustment to reflect 'little distress or burden in response to problems experienced', partners of patients with moderately severe hemophilia appeared to be less well adjusted to hemophilia. Coping with similar problems might be less efficient in these partners than in partners of patients with severe hemophilia because they face stressful encounters less frequently and might perceive their situation as less predictable or controllable. Another explanation is perhaps that as these partners, in general, have less health care contacts than the wives of patients with severe hemophilia, they are less well-informed about relevant issues, which in turn could result in unrealistic appraisal or fear and ineffective coping. Other contradictory findings concerned the negative associations between the patient's HIV status and 'consequences of hemophilia' and 'coping of patient'. These effects might find their explanation in HIV overruling other problems related to hemophilia. In the presence of HIV, partners as well as patients might view the 'consequences of hemophilia' as less severe, or 'coping with hemophilia' as less important, resulting in lower burden scores for these domains.

The impact of hemophilia on the partners' lives was most strongly associated with the burden experienced (Table 4). In particular family life, the relationship and activities with the patient, daily activities and leisure were affected by the hemophilia-related burden (Table 3). Impact on the relationship was often positive, or positive and negative, which echoed a finding reported for 111 parents of children with hemophilia stating that 'if subjects thought that the quality of their marriage was influenced by their child's hemophilia, they more often thought that their marriage had changed for the better than for the worse' (Varekamp, Suurmeijer, Rosendaal, Van Dijck, Bröcker-Vriends and Briët, 1990). Preventing burden, in particular regarding the consequences of hemophilia and social response, would reduce the impact of hemophilia on the partners' lives. More specifically, preventing pain, disability and virus infections (i.e. HIV) in patients, and minimizing caregiving demands (e.g. diminishing the frequency of caregiving) would reduce the impact of hemophilia.

Hemophilia and its related burden or impact appeared to have no direct effect on the well-being of partners (Table 4). Most prominent predictors of partners' satisfaction, self-esteem and health were psychological characteristics (low anxiety and depression, high optimism), social support (high quality of relationship, and positive interactions), and the absence of a chronic illness in partners. Nevertheless, we might suppose hemophilia to indirectly influence the partners' well-being by affecting the patients' health, which in turn showed a direct positive association with satisfaction and health in partners. This suggestion is consistent with results reported by Rothman, Hedrick, Bulcroft, Hickam and Rubenstein (1991) which indicated that patients who reported more impairment in psychosocial health status tended to have proxies (e.g. partners) who reported more impairment in their own health status.

The burden experienced, the frequency of caregiving and knowledge of hemophilia all added to the partners' subjective evaluation of the disease. This might explain why partners, on average, tended to perceive hemophilia as a more severe disorder than did patients. The finding is in line with the general notion that significant others tend to underestimate the patients' quality of life (see review by Sprangers and Aaronson, 1992), and consistent with studies which revealed that partners or family caregivers tend to over represent the level of impairment or illness experience relative to the patient's own judgement (Clipp and George, 1992; Epstein, Hall, Tognetti, Son and Conant, 1989; Magaziner, Simonsick, Kashner and Hebel, 1988; Rothman *et al.*, 1991; Rubenstein, Schaire, Wieland and Kane, 1984) and also the notion that the accuracy of proxy-generated scores on patients' functioning could be negatively influenced by the perceived burden of caregiving (Rothman *et al.*, 1991).

Finally, the present study demonstrated results on burden and impact of hemophilia on partners, but can we extrapolate our findings to other chronic disorders if we account for specific characteristics and courses of a disease (i.e. acute, progressive, episodic)? We presented burden as a multidimensional concept and, in general, we suggest a core set of four domains to be relevant in studying burden related to chronic disorders: 'physical demands' (e.g. consequences of the disorder), 'psychological demands' (e.g. coping of the patient), 'social demands' (e.g. social response), and 'financial or material demands' (e.g. dependency on external resources). Each domain should consist of a list of both generic and disease-specific potential problems to enable comparisons between disorders and within partners of specific patient groups. A panel of experts can define disease-related problems for any type of disorder to develop relevant and meaningful domains of burden. Additionally, including clinical measures and variables expressing the patients' self-reported disease-related physical health in regression analyses, and controlling for sex, age and type of relationship, will enable the study of burden and impact in any population of caregivers.

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