Journal of Chitwan Medical College 2024;14(47):67-71
Available online at: www.jcmc.com.np

ORIGINAL RESEARCH ARTICLE

PSYCHOSOCIAL BURDEN OF PARENTS HAVING CHILDREN WITH HAEMOPHILIA IN NEPAL

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Received: 23 Nov, 2023
Accepted: 14 Mar, 2024
Published: 30 Mar, 2024

Key words: Children; Haemophilia; Parents; Psychosocial burden.

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*DOI:*https://doi.org/10.54530/jcmc.1444

Citation

Shakya M, Banstola RS, Shrestha R. Psychosocial burden of parents having children with haemophilia in Nepal. Journal of Chitwan Medical College.2024;14(47):67-71.

Peer Reviewed

ABSTRACT

Background: Parenting a child with haemophilia, a hereditary bleeding disorder, causes more stress than parenting a normal child. The main aim of this study was to find out the psychosocial burden faced by parents having children with haemophilia in Nepal.

Methods: In this cross-sectional study, 76 parents having children with haemophilia were recruited from Nepal Haemophilia Society using a non-probability purposive sampling technique. The Zarit Burden Interview scale was used. Chi-square test and odds ratio at 95% confidence interval were used to assess an association between parents' psychosocial burden with the parents and child-related variables using SPSS version 16.

Results: A moderate level of psychosocial burden was prevalent among 60.5% of the parents. There was a significant association between parents' psychosocial burden with parents' education (p = 0.037), income (p = 0.03), type of Haemophilia among children (p = 0.03) and type of treatment received by children (p = 0.01). Further, the odds ratio predicted that psychosocial burden was 3.0 times higher (95% CI = 1.1 – 8.3, p = 0.04) among those parents with insufficient income and 4.2 times higher (95% CI = 1.3 – 13.4, p = 0.02) for the parents whose children were under factor VIII and associated treatment.

Conclusions: Psychosocial burden, particularly of moderate intensity was higher in parents having children with haemophilia. Those parents with low education level, with less income and whose children are diagnosed with Haemophilia A and under factor VIII and associated treatment should be more considered in reducing their psychosocial burden.

INTRODUCTION

Haemophilia is a hereditary bleeding disorder, which results in excessive bleeding due to a partial or total lack of essential blood clotting factors.[1] Haemophilia A results from a deficiency in clotting factor VIII and deficiency of factor IX causes haemophilia B.[1,2] An estimated prevalence of Haemophilia A is 1 in 5000 live births, while haemophilia B is prevalent for 1 in 30,000 live births.[1,2] Moreover, the mortality rate for people with congenital haemophilia A was reported between 0.9 and 2.2/100 person-years.[3] Haemophilia shows a sex-linked recessive inheritance,[2,4] which is caused by a mutation in *F8* and *F9* genes located on the X chromosome that provide instructions for making the clotting factors. This mutation prevents clotting factors work properly.[4]

The main aim of the management of haemophilia is either treatment of bleeding or prophylactically prevention of bleeding with replacement of deficient factors. Though regular prophylaxis is a standard recommended care for bleeding prevention and related complications, its widespread adoption is hindered by a very high cost.[7] Furthermore, haemophilia causes a huge physical, psychological, social and economic impact on affected individuals and their families which demands life-long health and personal care.[2,8,] Parents, particularly might have to sacrifice or change their employment, career, finance, social connections and self-care.[9,10,11] This entire process would be overwhelming and may cause psychological trauma to the parents. Given the lack of understanding among Nepalese parents, this study aims to determine the psychosocial burden experienced by parents having children with haemophilia and associated factors in Nepal.

METHODS

A descriptive cross-sectional research design was used and the study setting was the Haemophilia Care Unit (HCU), Nepal Haemophilia Society (NHS), Kathmandu. Only one caretaker, either the father or the mother of the children having haemophilia was included in the study. There were in total 313 children with haemophilia, from birth to 19 years, registered in NHS, so the required sample size was 76 using Solvin's formula.

Ethical approval was received from the Institutional Review Committee – Nepalese Army Institute of Health Sciences (Reference No.: 245). Formal permission was also received from the study site. Participation in the study was voluntary and written consent was obtained from all the participants. A revised version of the Zarit Burden Interview (ZBI)[12] scale was used to measure the burden of caregivers/parents. There were 22 questions on the 5-point scale with a minimum score of zero and a maximum score of four in each. The level of psychosocial burden obtained through ZBI was classified as negligible, mild, moderate, and severe based on the score of 0 – 20, 21 – 40, 41 – 60 and 61 – 88, respectively.[13] The ZBI was translated to the Nepali language after consultation with bilingual experts. The findings of this study in the Nepali version were reliable as indicated by Cronbach's alpha (0.88). Data analysis was done in SPSS version 16. Descriptive statistics (frequency, percentage, mean and standard deviation) were used to explain the characteristics of different variables measured in the study. Inferential statistics (chi-square test) was used to find out an association between the socio-demographic variables and parental burden. Odds ratio was calculated and the p-value equal to or less than 0.05 was considered significant at 95% of confidence interval.

RESULTS

Table 1. depicts the socio- and bio-demographic information of parents and their children having haemophilia. Majority (71.1%) of parents were young adults aged between 20 years to 40 years. Mean age was 36.4 ± 7.7 years. Regarding sex of parents, 51.3% of parents were mothers. Regarding education status, 52.6% of parents had primary to secondary levels of education, and 61.8% of them had income insufficient to meet their livelihood needs.

Most of the children with haemophilia were aged below 10 years (53.9%) with a mean age of 9.7 ± 4.6 years. Regarding the type of haemophilia, 80.3% of children had haemophilia A and 76.3% of them had mild to moderate levels of disease severity. Duration of treatment of haemophilia was noted above 5 years in 44 children (57.9%). The majority (78.9%) of children received factor VIII and associated treatment. The average cost of haemophilia treatment was estimated at US$ 44.5 ± 35.7 per child and a family history of haemophilia was noted in 32.9% of children with Haemophilia A being the more common (76.0%) type of haemophilia in the family.

Table 1: Socio- and bio-demographic information of parents and their children having haemophilia n=76

Socio-demographic information of parents		No (%)
Age (years)	20 – 40 years	54 (71.1)
	41 – 55 years	22 (28.9)
	Mean ± SD 36.4 ± 7.7years	
Sex	Male	37 (48.7)
	Female	39 (51.3)
Ethnicity	Brahmin/Chhetri	21 (27.6)
	Janajati	38 (50.0)
	Madhesi	10 (13.2)
	Dalit	7 (9.2)
Education	Illiterate	11 (14.5)
	Primary to secondary level	40 (52.6)
	higher-secondary or above	25 (32.9)
Occupation	Agriculture	22 (28.9)
	Business and services	24 (31.6)
	Homemakers	14 (18.4)
	Labours	16 (21.1)
Income	Insufficient to meet the needs	47 (61.8)
	Sufficient to meet the needs	29 (38.2)
Bio-demographic information of children		
Age (years)	≤ 10	41 (53.9)
	>10	35 (46.1)
	Mean ± SD 9.7 ± 4.6 years	
Type of Haemophilia (HP)	Haemophilia A	61 (80.3)
	Haemophilia B	15 (19.7)
Disease severity	Mild to moderate†	58 (76.3)
	Severe	18 (23.7)
Duration of HP diagnosis (years)	≤ 5	32 (42.1)
	> 5	44 (57.9)
Type of treatment	*Factor VIII and associated treatment	60 (78.9)
	#Factor IX and associated treatment	16 (21.1)
Cost of treatment (US$)	mean ± SD	44.5 ± 35.7
Haemophilia in family	Yes	25 (32.9)
	No	51 (67.1)
Type of haemophilia in family (n = 25)	Haemophilia A	19 (76.0)
	Haemophilia B	6 (24.0)

≤ less than and equal to; > greater than, SD = standard deviation
**factor VIII (n = 6) + Plasma and factor VIII (n = 36) + Blood, and factor VIII (n = 1) + blood, plasma and factor VIII (n = 6)*
#factor IX (n = 4) + plasma and factor IX (n = 11) + Blood and factor IX (n = 1)
† Only five cases (6.6%) had mild severity in this group

Table 2 reveals the level of psychosocial burden of parents having children with haemophilia. The moderate level of psychosocial burden was highly prevalent in the majority (60.5%) of the parents. The severe type of psychosocial burden was noted in only seven parents (9.2%) with a mean score of 46.4 ± 12.7.

Table 2: Level of the psychosocial burden of parents having children with haemophilia

Level of Psychosocial Burden	No. (%)
Negligible (0-20 score)	3 (3.9)
Mild (21-40 score)	20(26.4)
Moderate (41-60 score)	46(60.5)
Severe (61-88 score)	**7**(9.2)
Total	76(100)

Mean±SD of psychosocial burden score= 46.4 ±12.729 (possible range of score from 0 to 88)

Table 3: Association between parent's psychosocial burden and socio- and bio-demographic variables **n=76**

Socio-demographic Variables of Parents		Level of psychological burden		*P*-value
		Negligible to mild burden No (%)	Moderate to severe burden No (%)	
Age (years)	20 – 40 years	15 (27.8)	39 (72.2)	0.46
	41 – 55 years	8 (36.4)	14 (63.6)	
Sex	Male	14 (37.8)	23 (62.2)	0.16
	Female	9 (23.1)	30 (76.9)	
Education	Illiterate	1 (9.1)	10 (90.9)	0.037‡
	Primary to secondary level	10 (25.0)	30 (75.0)	
	higher-secondary or above	12 (48.0)	13 (52.0)	
Occupation	Agriculture	5 (22.7)	17 (77.3)	0.28
	Business and services	2 (14.3)	12 (85.7)	
	Homemakers	6 (46.2)	7 (53.8)	
	Is itLabours	5 (31.2)	11 (68.8)	
Income	Insufficient to get the needs met	10 (21.3)	37 (78.7)	0.03
	Sufficient to get the needs met	13 (44.8)	16 (55.2)	
Bio-demographic information of children				
Age (years)	≤ 10	13 (31.7)	28 (68.3)	0.09†
	>10	10 (28.6)	25 (71.4)	
Type of Haemophilia (HP)	Haemophilia A	15 (24.6)	46 (75.4)	0.03†
	Haemophilia B	8 (53.3)	7 (46.7)	
Disease severity	Mild to moderate	17 (29.3)	41 (70.7)	0.75
	Severe	6 (33.3)	12 (66.7)	
Duration of HP diagnosis (years)	≤ 5	7 (21.9)	25 (78.1)	0.17
	> 5	16 (36.4)	28 (63.6)	
Type of treatment	*Factor VIII and associated treatment	14 (23.3)	46 (76.7)	0.01†
	#Factor IX and associated treatment	9 (56.2)	7 (43.8)	
Cost of treatment (US$)	mean ± SD	39.3 ± 6.7	46.8 ± 37.3	0.41
Haemophilia in family	Yes	5 (20.0)	20 (80.0)	0.17
	No	18 (35.3)	33 (64.7)	

≤ less than and equal to; > greater than, SD = standard deviation, negligible to mild psychosocial burden = score of 0 – 40, moderate to severe psychosocial burden = score of 41 – 88

**factor VIII (n = 6) + Plasma and factor VIII (n = 36) + Blood, and factor VIII (n = 1) + blood, plasma and factor VIII (n = 6); #factor IX (n = 4) + plasma and factor IX (n = 11) + Blood and factor IX (n = 1)*

† Chi-Square test, ‡ Fisher's exact test

Table 4: Bivariate analysis of factors significantly associated with the psychosocial burden in parents having children with haemophilia **n=76**

Factors	Variables		OR (95% CI)	p-value
Parental factors	Education	Illiterate†	6.7 (0.8 – 55.8)	0.08
		Primary to secondary level‡	2.8 (0.96 – 8.0)	0.066
	Income	**Insufficient to get the needs met**	**3.0 (1.1 – 8.3)**	**0.04**
Child-related factors	Type of haemophilia	Haemophilia A	3.5 (1.1 – 11.3)	0.056
	Type of treatment	**Factor VIII and associated treatment**	**4.2 (1.3 – 13.4)**	**0.017**

OR = odds ratio, † comparison between illiterate and literate, ‡ comparison between primary to secondary level and higher-secondary and above educations

Table 3 shows an association between parents' psychosocial burden and socio- and bio-demographic variables. There was a significant association between parents' psychosocial burden with parents' education (p = 0.037), their income (p = 0.03), type of Haemophilia among children (p = 0.03) and type of treatment received by children (p = 0.01).

For the significant variables, we calculated the odds ratio at a 95% confidence interval. Table 4 depicts the bivariate analysis of factors significantly associated with the psychosocial burden in parents having children with haemophilia. Among parents-related factors, the parents with insufficient income to get their needs met had a three times higher burden (OR = 3.0; 95% CI = 1.1 – 8.3, p = 0.04). In child-related factors, those

parents whose child was receiving factor VIII and associated treatment had a 4.2 times higher burden (OR = 4.2; 95% CI = 1.3 – 13.4, p = 0.017).

DISCUSSION

This study comprehensively examined the psychosocial burden of parents having children with haemophilia. Socio-demographic data showed majority (51.3%) of caregivers were mothers, whereas the European multi-centre study showed 81.3% of the caregivers were mothers. [22] This difference might be attributed to the difference in setting and population of the study. Agreement with the studies from European countries, UK and India, haemophilia A was more common (80.3%) than haemophilia B (19.7%) in this study.[1,2,23] In previous studies, the prevalence of Haemophilia A was 1 in 5000 live births whereas, haemophilia B was prevalent for 1 in 30,000 live births.[1,2] Though the stress of having a child with haemophilia was well recognised, particularly among mothers in the family regarding medical care,[24] overall burden was reported comparable between parents.[25] Agreement to the literature, this study also did not find a significant difference in the psychosocial burden between mothers and fathers.[24, 25] Based on this finding, it can be said that both the father and mother perceive equal level of psychosocial burden in having to care a child with haemophilia.

Here, 60.5% of the parents having children with haemophilia had a moderate level of psychosocial burden and this finding closely agrees with the study conducted in UK which reported that 65% of the parents "worried about what will happen to their child in the future".[23] Similarly, in a multicentre study in Europe the majority of parents (68.6%) reported that haemophilia had an impact on their life in general.[26]

Consistent with the literature, this study showed a trend towards a higher level of psychosocial burden among illiterate than literate parents and among parents with primary to secondary level education than higher-secondary and above education. A previous review study showed that caregivers with no formal qualification reported significantly higher burden in the domains of emotional stress, financial burden, interaction with others, perception of a child dealing with haemophilia, impact and frequency.[22]

Caregiving for a child with haemophilia is burdensome and impacts on caregiver's financial status.[24,] In this study, the risk of psychosocial burden associated with haemophilia increased 3 times in parents having insufficient income than those having sufficient income. This could be reasonably supported as parents could have lost their days for work and employment opportunities due to the time required to dedicate to the care of those children. In support of this finding, a study in Europe reported that 13.6% of parents changed their occupation and more than 40% of parents worked part-time due to their child's haemophilia.[22] In another study from the UK, 55% of caregivers reported an economic impact related to haemophilia.[23] It was also indicated that 63% of parents having a child with haemophilia were affected negatively by employment, causing economic loss.[27] In this study economic burden might arise from loss of income because the cost of treatment was not significantly associated with the severity of parent's burden.

In this study, factor VIII and associated treatment were significantly related with an increased burden in parents that is 4.2 times higher than that for factor IX and associated treatment receiving group. However, this study did not elucidate an association based on treatment with and without inhibitors. Also, this finding could be confounded by the number of cases and severity of haemophilia A than the severity of haemophilia B. A further study is warranted to investigate the parent's psychosocial burden relating to employment status and treatment with inhibitors.

CONCLUSION

A significant number of parents experienced the psychosocial burden of having a child with haemophilia. Parents' educational level, income that is insufficient to meet livelihood needs, having children diagnosed with Haemophilia A and children receiving factors VIII and associated treatment were associated with the psychosocial burden among parents. Therefore, nurses, social workers, health care providers and all who are involved in the care to the child with haemophilia and their families can have important practical implications. The intervention in better supporting and maintaining the psychosocial health of parents and in reducing their experience of burden can be planned and implemented focusing on those with less education, less income and those parents whose children are diagnosed with Haemophilia A and whose children are receiving factor VIII and associated treatment.

CONFLICT OF INTEREST: None

FINANCIAL DISCLOSURE: None

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