



Self-concept and coping strategy among adolescent persons with haemophilia (PwH) and its correlation with clinical and socio-demographic parameters

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Abstract—

Background- Haemophilia is associated with long term chronic complications especially chronic arthropathy which affects physical and psycho-social aspects of patient's life especially in adolescent age group.

Aim -To identify the self-concept and coping strategy of adolescent person with haemophilia (PwH), correlation between the two and to determine their association with selected socio-demographic and clinical variables.

Method- In this study adolescent (12-16 years) haemophilia A and B patients across all severity attending day care center were included. Patients with acute bleeding were excluded from the study. The self-concept was assessed using Rosenberg's Self-Esteem Scale and structured interview schedule based on CSI-SF tool was used for coping strategies. Clinical data were collected by record analysis and demographic proforma was used for personal data. The tools were validated using Inter-rater reliability method.

Result – A total of 65 PwH were included in the study; 49.1% and 50.9% had severe and moderate disease respectively. The study revealed that 86.15% had moderate self-concept and 83.1% adopted engagement coping (emotional and problem focused) strategies. No relation was found between self-concept and coping strategies, a positive association found between coping strategies and age of adolescents with haemophilia.

Conclusion—Majority of adolescent haemophilia patients have moderate self-concept and adopt engagement coping strategy to deal with psychosocial and emotional stress associated with chronic debilitating disease. Thus, there is a need of holistic approach to management of haemophilia focusing on both physical and psycho-social wellbeing.

Key words: hemophilia, chronic debilitating disease, self-concept, coping strategy, psycho-social health

Introduction-

Haemophilia is an X linked recessive bleeding disorder diagnosed mostly in early childhood which demands immediate management of acute bleeding episodes and long-term treatment to prevent chronic complications mainly joint damage .[1] There has been advancement in the field of diagnosis and management of bleeding disorders leading to increased longevity which demands a holistic approach to management of this chronic disease including physical and psycho-social support to

these patients to handle increasing demand of family and society with age. Despite the available adequate treatment, patients with bleeding disorders in developing countries like ours still endure difficulties in daily life, such as hospital visits and frequent injections [2]. The situation is more challenging with the transition to the adolescent phase, when they undergo physical, psychological, and social changes. [3] The disease affects the patient's growth and development, lifestyle at school, career planning, work, family, finances, and leisure. [4]. Anger and denial might cause increases in risk-taking and attempts to avoid appropriate treatment. [5] The psychological and social aspects of adolescent haemophilia patient can be analysed in terms of self-concept and coping strategies. Self-concept, is perception of illness and refers to the way adolescents with haemophilia think about themselves, evaluate their appearance, thoughts, and behaviours, and perceive themselves both as individuals and in comparison, to others.[6] Coping strategies refers to the specific efforts, both behavioural and psychological, that adolescents with haemophilia employ to minimize stressful events.[7]

Thus, we understand that haemophilic adolescents, who are suffering since infancy, require bio-psychosocial direction to provide them most convenience life such as disease control, identifying the difficulties and gaining the maximum empowerment. For that, the study will emphasize the coping strategies on complication management, pain management and perception or self-concept about haemophilia in relation to some selected socio demographic and clinical data. The present study aimed to identify the self-concept and coping strategy of adolescent people with haemophilia (PwH), correlation between the two and to determine their association with selected socio-demographic variables. The primary objective was to assess self-concept and coping strategies of PwH. The secondary objective was to find out the relation between self-concept and coping strategies and to determine the association of them with selected demographic variables.

Method This descriptive study was done at N.R.S. Medical College & Hospital Kolkata from January 1 to June 30 in the year 2023. Ethical permission was obtained from institutional ethics committee (IEC). The study included all PwH between the age of 12 to 16 years attending haemophilia clinic and day care centre and were willing to participate in the study. All PwH who were acutely ill were excluded from the study since it was a day care based study. The conceptual framework adopted for the study was based on Betty Neuman's Model.[8] Nonprobability purposive sampling technique was followed to select 65 samples. Data were collected by record analysis for clinical profile, 'r' demographic proforma for personal data, and self-concept was assessed by Rosenberg's self esteem scale and structured interview schedule based on Coping Strategies Inventory Short-Form tool (CSI-SF tool) for coping strategies. Demographic proforma was developed for collecting the personal data, consisting of 8 items such as age, gender, patient's education, education of father, education of mother, types of family, habitat (rural/urban), and monthly family income. Disease related data were collected by record analysis of clinical profile. It described the disease condition of samples consisting of 3 items such as type of haemophilia, duration of illness, type of treatment. Rosenberg self-esteem scale [9] had sixteen items and was calculated by assessing scores of '4' for response category of "strongly agree", '3' for "agree", '2' for "disagree" and '1' for "strongly disagree". The total score for sixteen items ranges from 16- 64. Score 16-31 was noted as 'low self-concept'; 32-48 as 'moderate self-concept' and 49-64 as 'high self-concept'. Evaluation of coping strategies was done by interview schedule based on CSI-SF tool developed by Addison CC et al in 2007. [10] It is a five Point Rating Scale with sixteen items and eight subscales each having two questions. Each item is scored on a scale of 'never' (=1), 'seldom' (=2), 'sometimes' (=3), 'often' (=4), 'almost always' (=5). This tool is designed to collect data regarding engagement & disengagement by calculating highest score.

The tool was given to five experts for the validation based on criteria checklist. Among them one was professor of haematology department, one expert from medical surgical nursing, two experts from child health (paediatric) nursing and one expert from psychiatry nursing. Tool was translated into regional language (Bengali) and national language (Hindi), and retranslation was done with the help of respective language experts. Linguistic validation was done by the experts from respective languages. Reliability of semi-structured demographic proforma was established by inter-rater method. The computed value of 'r' was found to be 0.9. The tool was found reliable with 100% inter-rater's agreement. The reliability of the Four-point rating scale of Semi Structured Interview schedule for self-concept and five -point coping strategies rating scale was established by Cronbach's alpha method. [11] The result of r was 0.94 and 0.78 respectively. The results indicate that the tools were reliable for using data collection. Ethical permission was obtained from institutional ethics committee. Informed written consent was taken from the parents of respondents. Both descriptive and inferential statistics were used to analyse, classify, and tabulate the data. Findings related to the demographic variables, self-concept and coping strategies by frequency and percentage. Findings related to co-relation between self-concept and coping strategies by correlation coefficient. Findings related to the association between self-concept and the selected demographic variables by chi-square. Findings related to the association between coping strategies and the selected demographic variables by chi-square.

Result:

A total of 65 patients were included in the study. Percentage respondents belonging to age 12-14 years were 64.6% (42/65) and 15-16 years were 35.3% (23/65). All were male patients and maximum (76.9%) belonged to rural area. 63.07% were educated till class VII and 36.92% were educated till class X. In terms of parent's education 21.53%, 55.38% and 21.53% fathers were illiterate, educated till class VIII and class XII respectively.

Among all the mothers 21.53%, 73.84% and 4.61% were illiterate, educated till class VIII and class XII respectively. They had revealed a family income of INR 2000-7000, INR 7001-12000 and >INR 12,000 in 81.53%, 16.92% and 1.53% families respectively. Out of all PwH studied, 87.69% (57/65) and 12.3% (8/65) had hemophilia A and B respectively. Out of all, Hemophilia A patients 46.1% and 41.5% belonged to the categories of severe and moderate respectively. Among hemophilia B patients, 9.2% and 3% were moderate and severe hemophiliacs respectively. Out of all PwH, 84.6% and 15.3% were on 'prophylaxis' and 'on demand' therapy respectively. The duration of illness was 10-12 years in 53.8 % and 13-15 years in 46.1 % PwH. Moderate and high self-concept was seen in 86.15% (56/65) and 13.8% (9/65) respectively (Table 1). 'Low self-concept' was absent in the study population. Engagement and disengagement coping strategies was present in 83.1% (54/65) and 16.9% (11/65) of respondents respectively (Table 1). Sub scaling of coping strategy was done based on CSI-SF tool (Table 2), out of four subscale the adolescents adopted Emotion Focused Engagement (EFE) for coping mostly with mean of 70% i.e. they communicate their feelings to others and also increase their social involvement followed by Problem Focused Disengagement (PFD) with mean of 65% where they used to avoid the situation, Problem Focused Engagement (PFE) with mean of 55% and lastly Emotion Focused Disengagement (EFD) with the mean of 40% (Table 3).

Table 1: Distribution of adolescent people with haemophilia according to self-concept and coping strategies (n=65)

	Frequency (n)	Percentage (%)	Mean +/-SD	Median
Self-Concept				
Low (16-31)	0	0	43.615+/- 3.5075	44 (IQR)
Moderate (32-48)	56	86.15		
High (49-64)	9	13.8		
Coping strategies				
Engagement	54	83.1	NA	NA
Non-Engagement	11	16.9	NA	NA

Abbreviation: IQR= interquartile range; NA= not applicable

Table 2: Sub scaling of coping strategy by CSI-SF based tool

Subscale	Range	Median	Mean	SD
PFE	4-20	11.1	10	2.89
EFE	4-20	13.58	13	2.49
PFD	4-20	13.3	13	2.92
EFD	4-20	8.29	9	3.07
E	3-40	21.6	24	4.15
D	3-40	21.6	22	4.48

Abbreviations: SD= standard deviation, PFE = problem-focused engagement; E=engagement, EFE= emotion focused engagement, PFD=problem-focused disengagement, D=disengagement, EFD=emotion focused disengagement.

Table 3: Ranking of coping strategy with the help of CSI-SF based tool

scale	mean	mean percentage	rank
PFE	11.12	55	3
EFE	13.58	70	1
PFD	13.3	65	2
EFD	8.29	40	4
Abbreviations: PFE = problem-focused engagement; EFE= emotion focused engagement; PFD=problem-focused disengagement; EFD=emotion focused disengagement.			

There was no correlation between self-concept and coping strategies as the calculated value of r (0.03) is less than table value of r (0.244) ($n=65$; r df(63) = 0.244 $p > 0.05$). There was no association between self-concept and age, education of father, education of mother, types of treatment and duration of illness as calculated value of chi square is less than table value. There was statistically no significance between coping strategies and education of father, education of mother, types of treatment and duration of illness as calculated value of chi square test is less than table value. A statistically significant association was found between coping strategies and age as calculated value of chi square is more than table value (Table 4).

Table 4: coping strategies and age of adolescents with hemophilia

Variables	Coping strategies		Total	χ^2	df	Remarks
	≤median	>median				
≤14yrs	28	14	42	4.59	1	Significant
>14 yrs	9	14	23			
χ^2 df(1) = 3.84,p<0.05						

Discussion-

The study was conducted with the main purpose of assessing the self-concept and coping strategies among the adolescents with haemophilia and its correlation with clinical profile of disease and demographic data. In the present study it was revealed that maximum (86.15%) of the respondents were having moderate self-concept. The study conducted by Varadarajan M et al [12] with the aim of analysing clinical and psychological profile of children with bleeding disorders showed that 21.4% had low self-esteem; in the present study it was revealed that maximum (63.07%) of the respondents adopted to engagement coping strategies and mostly utilized EFE followed by PFE. However, no association was found between self-concept and coping strategy and, they were not related to clinical and other demographic variables. Except a significant association found between coping strategies and age of the adolescents. However, in a study done by Torres-Ortuno A et al [13] assessing the adherence to prophylaxis therapy it was found that adherent patients had appropriate coping strategy both cognitive and behavioural and non-adherent had inadequate coping strategy, high problem avoidance, social support was not sought, and they do not modify the negative meaning of the situation. There have been multiple

studies showing higher depression, anxiety scores and increased anxiety disorder diagnosis among children and adolescents with hemophilia having strong impact on quality of life. [14,15] Jiménez-Cebrián Ana María et al [16] found greater depression scores and range status were observed in patients with hemophilia compared to control. However, in a study, there was no differences between the hemophilic subjects and healthy controls in depression scores, and they concluded that although a higher distress and psychological problems could occur in the patients, hemophilic patients could show a good adjustment to this disease, like many other chronic diseases, and it can be accepted as a “life condition” that they can live with. [17] Von Macksen et al. [18], in a multicenter, cross-sectional study, described the impact of sport on health-related quality of life, patients participating in more sport reported significantly better than those participating in less sport. A statistically significant association was found between coping strategies and age of the adolescents. A similar study was conducted by [Ana Torres-Ortuño](#) et al [19] with the aim of assessing different coping strategies of patients with hemophilia showed that patients on prophylactic therapy had maladaptive coping strategy and hypochondrial behaviour. However, age variable was non-significant. Joint damage and age correlated ($p < 0.05$) negatively with perception of disease, perceived quality of life and coping strategies. [20]

Limitations –

There were few limitations in the present study e.g.- small sample size and that self-concept and coping strategies were not compared with treatment adherence, disease outcome and quality of life. The reliability or accuracy of the answers could have been affected, as patient were wearing mask and maintaining social distancing due to the ongoing pandemic at the time of interview.

Conclusion-

This study indicates that the adolescents with hemophilia have moderate to high self-concept and engaging coping strategy. Evidence suggests that there is no relation between self-concept and coping strategies, but the coping strategies is having positive association with the age of adolescents with hemophilia. In spite of the available adequate treatment, patients with bleeding disorders still endure difficulties and impairments in daily life which needs to be addressed at all stages of life. There should be awareness program regarding the importance of coping strategies. In hospital there should be counselling area regarding adolescence development, coping mechanism of the adolescents as well as their family members towards the disease process.

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