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## Understanding adherence to treatment and physical activity in children with hemophilia: The role of psychosocial factors

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### ABSTRACT

The objective of this study was to identify psychosocial factors to explain intentions of children and adolescents with hemophilia to adhere to recommendations for self-care. Twenty-four patients with hemophilia A and B, aged 6–18 years old, and their parents, completed a survey. Measures assessed factors from the theory of planned behavior, physical activity, and medical treatment adherence. The results indicate that past behaviors, attitudes, and subjective norms explained a large proportion of the intention to engage in future nonrecommended physical activity. This study supports the need to investigate motivational factors underlying behaviors in larger scale studies and identifies targets for future interventions.

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### KEYWORDS

Adherence; hemophilia; intention; physical activity; theory of planned behavior

## Introduction

Hemophilia is a rare genetic disorder that primarily affects men and that is caused by a clotting factor deficiency, leading to bleeding episodes in muscles and joints. Bleeds not immediately treated may eventually lead to impairment such as arthritis or disability [1]. Hemophilia can be medically managed with a lifelong prophylactic or on-demand treatment, which involves the infusion of the deficient clotting factor to help prevent bleeding and arthropathy. Regular exercise is recommended to patients in order to promote strong muscles, develop balance, and improve fitness, which in return can contribute to healthier joints and diminish the occurrence of bleeds. However, physical activity likely to cause trauma, such as high contact and collision sports, should be avoided [2].

Like in other chronic illnesses, difficulties in following recommendations for self-care have been reported in children and adolescents with hemophilia for both physical activity and treatment adherence. While suboptimal adherence is not uncommon in parents of young people living with hemophilia, adherence tends to be higher when parents take responsibility of their children's infusion [3]. When young people with hemophilia gain in autonomy and start regularly self-infusing, adherence to treatment reduces remarkably. In a recent survey, 90% of children aged 0–12 years old had a “high” or “very high” adherence to prophylaxis, compared with 54% for those aged 13–18 years old [4]. In addition, despite recommendations, a survey revealed that 60% of young people with hemophilia manage their condition

by limiting any type of physical activity [1]. Other studies suggest that a high proportion of children and youth with hemophilia engage in high-risk sports just like their friends [5,6].

Studies on adherence of people with hemophilia have identified over 20 reasons, mostly provided by health care professionals or patients, to explain nonadherence (ie, “lack of time,” “forgetfulness,” “fluctuation or disappearance of symptoms,” etc.), although these reasons were not systematically correlated with the actual level of adherence [1,4] [7–9]. However, no theoretical model has been used until now to understand motivation for treatment, thus precluding the development of intervention strategies.

The theory of planned behavior is one of the most frequently used theories to understand motivation for treatment and posits that intention to adopt a behavior is a central predictor of future behavior [10]. In pediatrics, parents are often responsible for their child’s treatment, thus making it difficult to assess the patient’s engagement. To counter this problem, it is possible to assess a child’s intention to follow future recommendations. Studying intention also allows us to study a child’s level of motivation to their treatment plan and its determinants. The theory of planned behavior posits three core factors leading to the formation of intention: attitudes, subjective norm, and perceived behavioral control. Finally, past behavior can also influence intentions and behaviors [11].

According to this theory, *attitudes* are based on the patient’s beliefs about the consequences of performing the behavior (ie, costs and benefits) and can be assessed broadly by its components (eg, the anticipated benefits of the recommendations in preventing joint bleeds) [12]. *Subjective norm* represents the social influence on behavior and is based on the patient’s beliefs about others’ opinions of the behavior, and the patient’s motivation to comply with these opinions. Finally, *perceived behavioral control* refers to the confidence about one’s abilities, knowledge, and skills to perform the behavior. The theory of planned behavior has been widely used to predict many health-related behaviors in people of all ages, and successful interventions have been developed using this model [13]. To date, self-care in children and adolescents with hemophilia has not been studied through this framework. The identification of predictive factors of self-care could help identify important motivational targets for future interventions.

## Objective

The objective of this study was to identify individual psychosocial factors to explain intentions of young people with hemophilia to adhere to the following recommendations: 1) infuse prophylactic treatment, 2) infuse on-demand treatment, 3) practice recommended physical activity, and 4) avoid practice of physical activity that was discouraged by their health care providers. To do so, we assessed associations between intentions to follow recommendations and potential psychosocial factors from the theory of planned behavior: attitudes toward recommendations, subjective norm (related to parents and friends), perceived behavioral control, and past behavior (past adherence to medication, recommended and discouraged physical activity).

## Methods

### Participants

Patients aged 6–18 years old with severe hemophilia A or B (factor VIII/IX < 1%), treated at our institution and accompanied by one of their parents, were approached for this study.

We chose to recruit people with severe form of hemophilia because of the intensity of symptoms these patients experience and the use of prophylaxis as a treatment. All eligible patients spoke French or English. Exclusion criteria included: diagnosis < 1 year and the presence of a severe psychiatric disorder. The survey was a one-time assessment administered to participants during their bi-annual appointment at the hemophilia clinic. Questions were read to patients, and a research assistant (SB) collected responses, while one of their parents answered a questionnaire in a different room. This questionnaire included sociodemographic questions as well as questions on past behavior for younger children (see *Measures*). This project was approved by the Sainte-Justine UCH's Research Ethic Board.

### **Measures**

To comply with theory of planned behavior norms and guidelines, we developed items and questions as recommended by Azjen and Fishbein [12,14].

#### **Intention**

Intention to adopt health behaviors was measured using one item for each behavior. The participant responded on a 7-level scale: *In the future, I intend to [the behaviour] as recommended, 1-strongly disagree to 7-strongly agree*. For interpretative purposes, we reversed items on the intention to adopt discouraged physical activity, higher scores translating as the intention to avoid discouraged physical activity.

#### **Attitudes**

We measured general attitude toward health behaviors using a generic item: *I see [the behavior] as: 1-negative to 7-positive*. We also evaluated the component of attitude related to the perceived benefits of the behavior in bleeding prevention by multiplying a measure of perceived benefit (*[the behavior] will prevent bleeding in my joints, 1-strongly disagree to 7-strongly agree*) by a measure of perceived importance (*Preventing bleeding in my joints is, 1-not important to 7-important*).

#### **Subjective norms**

Two forms of perceived social norms were measured: (a) perceived pressure from parents, and (b) from peers. Each social norm was measured using two items. The first assessed parents' or peers' perception of the behavior: *My friends appreciate that I [the behaviour] as recommended, 1-strongly disagree to 7-strongly agree*; while the second assessed the degree of influence of the parents-peers: *Regarding my [behaviour], I want to listen to my friends or do what they want me to do, 1-strongly disagree to 7-strongly agree*.

#### **Perceived behavioral control**

Items of perceived behavioral control were also developed. They were phrased as: *How confident are you or do you feel able to always [the behaviour] as recommended? 1-not at all confident to 7-very confident*.

Adherence to treatment was measured using the Validated Hemophilia Regimen Treatment Adherence Scale (VERITAS-Pro and VERITAS-PRN for adherence to prophylactic and on-demand treatment regimens, respectively) [15–17]. The questionnaires included 24 items and topics related to timing, dosing, planning, remembering, communication, and treating/skipping. Recent research indicated good psychometric properties—high reliability—with  $\alpha$  values of 0.85 and 0.92 for total scale and a test–retest  $r > 0.77$ . The children's scores

**Table 1** . Sample description.

Characteristics	Mean (SD), range	n (%) (n = 24)
Age (years)	11.8 (3.3), 6–18	
Country of birth		
Canada		20 (83%)
Other		4 (17%)
Type of hemophilia		
A (severe)		21 (88%)
B (severe)		3 (13%)
Type of treatment		
Prophylaxis		20 (83%)
Immune tolerance		4 (17%)
Bleeding episodes (past year)	4.3 (6.3), 0–24	

on the VERITAS questionnaires were used when the participants were autonomous in their treatment management. Otherwise, parent measures were used. In our sample, internal consistency for total scale remained high for VERITAS-Pro and VERITAS-PRN in both children-report ( $\alpha$  values  $> 0.87$ ) and parents-report ( $\alpha$  values  $> 0.73$ ).

To assess physical activity, we asked participants and parents to estimate physical activity in a typical week for both recommended and at-risk activities. As activities may vary greatly according to the season in Canada, we inquired for both winter and summer activities and averaged both sets of results. We used the same wording as in the Godin Leisure-Time questionnaire: [18] *How many days per week do you engage in recommended physical activity for at least 15 minutes?* Because estimation of physical activity is not reliable for children under 10 years old [19], parent estimations were used with these participants.

## Analysis

Associations between intention and psychosocial predictors were explored using partial correlation coefficients after controlling for age and past year bleeding episodes. Nonparametric tests were conducted to compare levels of associations across behaviors. Finally, all variables were entered in a regression analysis, and the model R-squared value was used to estimate the total variance of intentions explained by the studied psychosocial factors. An alpha level of 0.05 was used to detect significant associations. All analyses were performed with the *Statistical Package for the Social Sciences* SPSS v19.

## Results

All 31 patients who were in the list of the local hemostasis center and who met the criteria for inclusion and exclusion were approached for this study. Twenty-six patients (84%) and their parents accepted to participate (Table 1). Reasons for refusing to participate included: not having an appointment scheduled at the clinic ( $N = 3$ ) and lack of time at the moment of the assessment ( $N = 2$ ). Two participants were excluded from our study: one patient had an intellectual disability, and the other had an attention-deficit disorder, making a final sample size of 24. The mean age of participants was  $11.8 \pm 3.3$  years old. All participants were on prophylaxis and received supplementary injections in case of bleeding (on-demand treatment).

First, we observed that intention to follow recommendations was high for recommended physical activity, prophylaxis, and on-demand treatment ( $>6/7$  on a strongly disagree-strongly agree scale). However, intention to avoid practicing discouraged physical activity was significantly lower than that for other behaviors (Wilcoxon  $z = 2.31-2.96$ ,  $p < 0.05$ ) (Table 2).

**Table 2** . Description of means and standard deviations of children's intention, past behaviour, and psychosocial factors for four health behaviors.

		Physical activity		Treatment <sup>a</sup>	
		Recommended	Discouraged	Prophylaxis	On-demand
Intention	(1–7 scale) <sup>b</sup>	6.13(1.33)	4.75(2.25)	6.60(0.75)	6.70(0.66)
Past behavior					
Practice <sup>c</sup> (Nb days)		4.04(1.93)	1.54(1.94)		
Non-adherence <sup>d</sup>				38.45(13.71)	35.95(10.92)
Attitude					
General	(1–7 scale)	5.92(0.93)	5.00(1.62)	6.05(1.54)	6.15(1.53)
Bleeding prevention					
	Benefits (1–7 scale)	4.46(2.06)	5.42(1.69)	6.50(0.69)	6.25(1.16)
	Importance (1–7 scale)	6.79(0.42)	6.79(0.42)	6.79(0.42)	6.79(0.42)
	Benefits × importance <sup>e</sup>	30.50(14.56)	37.04(12.22)	43.90(5.73)	42.05(7.82)
Normative pressure					
Parents					
	Pressure (1–7 scale)	6.58(0.72)	5.54(1.79)	6.70(0.73)	6.70(0.73)
	Influence (1–7 scale)	4.75(1.75)	4.75(1.75)	5.95(1.32)	6.60(0.68)
	Pressure × influence <sup>e</sup>	31.79(12.58)	26.96(14.94)	40.35(11.19)	44.50(7.58)
Peers					
	Pressure (1–7 scale)	5.21(1.74)	4.58(2.10)	5.78(1.22)	6.06(1.24)
	Influence (1–7 scale)	2.50(1.35)	2.50(1.35)	2.72(2.19)	3.44(2.00)
	Pressure × influence <sup>e</sup>	12.58(7.73)	10.42(6.97)	16.11(15.06)	21.19(13.97)
Perceived control	(1–7 scale)	6.17(1.13)	4.75(2.05)	6.50(0.89)	6.45(0.89)

Note. *Intention*: Intention to follow the recommendations; *Attitude: general*: General positive or negative attitude towards the recommendations; *Attitudes: bleeding prevention*: Attitude regarding the efficacy of the recommendations in preventing joint bleed.

<sup>a</sup>N = 20 for this analysis, patients with inhibitors excluded.

<sup>b</sup>Likert scale 1 (lowest intention) to 7 (highest intention to follow recommendations).

<sup>c</sup>Number of days of practice in a typical week.

<sup>d</sup>Score at Veritas-Pro/PRN from 24 (most adherent) to 120 (least adherent).

<sup>e</sup>Items multiplied to reflect the importance given by the person in this aspect.

Practice of physical activity was higher for recommended activities ( $4.0 \pm 1.9$  days) as compared to that of discouraged activities ( $1.5 \pm 1.9$  days) ( $z = 3.429, p < 0.05$ ). Importantly, practice of discouraged physical activity also significantly differed from 0 ( $p < 0.05$ ). Adherence scores indicated relatively good adherence to prophylaxis and on-demand treatment from our participants in comparison to scores published in previous studies.

Attitudes were globally positive ( $z = 2.65$ – $4.25, p < 0.05$ ). However, attitude for avoiding risky physical activity was significantly lower than that for other behaviors ( $z = 1.80$ – $2.40, p < 0.05$ , except for on-demand treatment,  $p = 0.07$ ) as was also the case for perceived behavioral control ( $z = 2.75$ – $3.26, p < 0.01$ ). Median perceived behavioral control for discouraged physical activity was neutral, indicating that participants as a group were not confident regarding their ability to avoid discouraged physical activity in the future. Concerning recommended physical activity, a neutral median score was found for the item pertaining to the utility of recommended physical activity in preventing bleeding episodes, meaning that participants did not necessarily see the usefulness of recommended physical activity as a preventative measure. To identify factors of behavioral intention, we focused on statistically significant partial correlations (Table 3). After controlling for age and bleeding episodes, higher intention regarding recommended physical activity tended to be associated with larger perceived encouragements from peers. Intention to avoid discouraged physical activity was associated with a lower practice of risky physical activity in the past, a positive general attitude toward avoidance, a positive attitude regarding the efficacy of avoidance in preventing bleeding episodes, as well as the perception of a more encouraging social environment, from both parents and peers. Intention to perform on-demand treatment as recommended was associated with a stronger normative parental pressure and a higher level of perceived behavioral control.

**Table 3.** Partial correlations of behavioral intentions with psychosocial factors controlled for age and past year bleeding.

		Intention to follow recommendations			
		Physical activity		Treatment <sup>a</sup>	
		Recommended	Discouraged	Prophylaxis	On-demand
Past behavior <sup>b</sup>	Physical activity	0.341	0.490*		
	Adherence <sup>c</sup>			0.240	− 0.199
Attitude	General	0.048	0.709**	0.161	0.080
	Bleeding prevention	− 0.008	0.612**	0.429	0.399
Normative pressure	Parents	0.341	0.543**	0.278	0.681**
	Peers	0.414 <sup>†</sup>	0.478*	− 0.040	0.129
Perceived control		0.000	0.166	− 0.163	0.496*

<sup>a</sup>N = 20 for this analysis, patients with inhibitors were excluded.

<sup>b</sup>Degree at which patients followed the recommendations in the past (some item score have been reversed).

<sup>c</sup>Adherence: We used reversed scores at VERITAS-Pro-PRN.

\* $p < 0.05$ , \*\* $p < 0.01$ .

Finally, in additional analysis, we performed a multiple regression analysis for each of the studied health behaviors where all the predictors were entered in a single model to estimate the R-squared values. A comparison of the variance of intentions accounted for by psychosocial factors (excluding past behavior) showed 54% for recommended physical activity intention, as compared to 68% for discouraged physical activity, 22% for prophylaxis, and 54% for on-demand treatment, indicating that psychosocial factors were more powerful in explaining intention to adopt discouraged physical activity than other behaviors.

## Discussion

Considering the important and irreversible consequences of nonadherence to treatment in hemophilia, it is important to understand what leads children and adolescents to follow recommendations. This is the first study that systematically explored factors of health behaviors in hemophilia using a behavioral theory approach. We found that different psychosocial factors accounted for an important part of the variance of intentions for each behavior, suggesting that different approaches should be used to influence adherence for each domain.

The main finding of this study is the high variance in the intention to avoid discouraged physical activity due to psychosocial factors (68%). Considering that this behavior may be one of the hardest to follow for young patients (lowest intention of all four behaviors), it should represent a valuable target for psychosocial interventions. Variance in intention for discouraged physical activity was also higher than that for other behaviors, suggesting that young people with hemophilia differ greatly in their commitment to avoid risky physical activity. On average, patients spent 1.5 days per week engaged in discouraged physical activity for 15 minutes or more. As two or more bleeds in a joint may cause irreversible damage, assessing all social factors involved in the desire to practice discouraged physical activity would be beneficial, including attitudes and perceived pressure from parents and peers [20].

Subjective norms related to parents was significantly related to intention to avoid risky physical activity ( $r = 0.543$ ,  $p < 0.01$ ), and children perceived less pressure from parents to avoid risky physical activity than that for other favorable behaviors. It must certainly be difficult for parents to restrict their child from activities practiced by other children. Parents may also want to compensate for the consequences of their child's physical condition, or for the

guilt they may experience related to the genetic transmission of the disease [21]. These observations promote the need for interventions that help parents deal with their feelings about restrictions [22].

Unsurprisingly, as reflected by subjective norm results, the role of parents appeared to be important, explaining individual intentions of two of four behaviors in our study. Research has shown that information that parents communicate to their child can greatly differ from the information they get from professionals due to a variety of biases [23]. Our results suggest that parental erroneous beliefs or opinions could easily transfer to children and are probably a relevant target to enhance children's commitment to their treatment plan.

Consistent with previous research, we found that peers may have a greater effect on physical activity than on treatment adherence [24]. Thus, if disclosure to peers appears appropriate, emphasis could be on special needs for physical activity rather than on providing information about medical treatment. Professionals should routinely ask patients to what extent the disease interferes with friendships or peer relationships, especially if restrictions in physical activity may lead to a sense of social isolation [24,25].

Another important result was that general attitude was associated with the intention to adopt risky physical activity. As attitudes are global affective evaluations, this stresses the need to address motivational issues beyond the necessary step of informing and educating. Information interventions alone are known to have only a negligible effect in improving adherence in pediatrics [26]. A wide range of efficient interventions have been developed to address values and concerns and modify behaviors in pediatrics [27]. Following our results, interventions for children with hemophilia could help them feel more comfortable with restrictions about physical activity as well as develop more positive views of appropriate activities.

Although the general attitude appeared to be positive for recommended physical activity, the children of our sample did not seem to fully realize its benefits, as shown by a median neutral score on the item assessing treatment efficacy in preventing joint bleed. This may reflect the fact that communication with patients is more focused on avoiding risky physical activity than on the benefits of recommended physical activity. As some children may not have had bleeding episodes in years, making it hard for them to realize the long-term consequences of their actions, it would be very useful to increase patients' knowledge on treatment efficacy in order to promote adherence [7,28]. In that sense, strategies addressing communication on such "hot topics" within the family should be developed [29].

Finally, our limited results on perceived control should be interpreted with caution. Given that most of our participants still relied on their parents regarding behaviors relating to their illness, perceived control may not have been validly measured in the present study. Further research should therefore address this topic in samples composed of older children and adolescents who are more autonomous with their treatment (eg, age range 14–18 years old).

### **Limitations**

The results found in this study should be carefully interpreted. First, the sample size prevented us from relying on basic inferential statistics. Consequently, this limits the external validity of our findings. Second, the correlational design of this study prevents us from drawing causal conclusions between factors and intentions. Third, the limited size of some of the correlations may also be due to a lack of variability or ceiling effects. Therefore, the present research should be considered as a preliminary step to identify promising factors of self-care behaviors to be further tested in larger samples. The results support the need to systematically investigate these psychosocial factors to identify targets for future interventions.

## Conclusion

To conclude, this study found that several psychosocial factors were associated with intentions of young people with hemophilia to behave in accordance with recommendations of the health care team. We found that past behavior, general attitude and attitude related to bleeding prevention, and subjective norm related to parents and peers explained a large proportion of the intention to avoid risky physical activity. Further studies should therefore target physical activity and how choices are made in young people with hemophilia. Given the differential associative patterns across behavior domains, future studies should address the different dimensions of health behaviors in hemophilia, without limiting their research to medical treatment adherence.

## Implication for practice

Hemophilia is a rare bleeding disorder for which psychosocial studies are needed considering its important impact on the daily life of patients and their family. Identification of predictive factors of self-care can help identify targets for future interventions. Several psychosocial factors were associated with intentions of young people with hemophilia to behave in accordance with recommendations of the health care team. We found that past behavior, general attitude and attitude related to bleeding prevention, and subjective norm related to parents and peers explained a large proportion of the intention to avoid risky physical activity. These results stress the importance of assessing those psychosocial factors when promoting appropriate self-care in children with hemophilia.

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## Declaration of interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

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