Measuring adherence in pediatric hemophilia patients: French-language adaptation of the VERITAS-Pro and VERITAS-PRN and psychometric properties

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Abstract: Veritas-Pro and Veritas-PRN are instruments to assess adherence to treatment of people with hemophilia. Low adherence can limit treatment effectiveness and result in increased bleeding episodes and chronic pain. The purpose of this study was to adapt both questionnaires into French, report preliminary psychometric data in a group of pediatric patients and their parents, and compare ratings between young patients and their parents. The translation process followed these steps: (1) forward translation (2) backward translation (3) final version, and (4) testing. Translators were familiar with the pediatric community. Thirteen pediatric patients and twenty parents participated in the study. The translation process was conducted without major difficulties, and children easily understood the French versions. Preliminary psychometric analyses showed good to excellent consistency for the total scale and most subscales. However, the consistency of the Dose (VERITAS-Pro) and the Plan (VERITAS-PRN) subscales were low. When pediatric patients were primary infusers, they reported more skipping of prophylaxis than in parents’ estimation of their behavior. For episodic treatment, when parents were primary infusers, they reported better adherence for the Time subscale vs. children’s estimation of their behavior. Parents also rated higher adherence on prophylaxis.

ABOUT THE AUTHOR
The Psycho-Oncology Centre is a research unit specializing in the study of the psychosocial aspects associated with pediatric diseases. Created in 2012, the laboratory is located at CHU Sainte-Justine and is affiliated with the Université de Montréal (Quebec, Canada). The laboratory is directed by Serge Sultan and comprises researchers, clinician-researchers, Ph. D. students, and trainees. Our mission is to better understand the vulnerability and resilience factors in individuals and families facing chronic pediatric illness, and to develop interventions to improve the quality of life of patients, their family and their caregivers. The aim of this study is to support empirical research on the determinants and outcomes of adherence to treatment in hemophilia, as well as the development of interventions in this field.

PUBLIC INTEREST STATEMENT
Hemophilia is a rare bleeding disorder in which the blood does not clot normally. Over time, repeated bleeding can lead to permanent joint damage and chronic pain. Bleeding episodes can be prevented by appropriate self-care. Infusions of the missing factor protein on a regular basis or when needed are usually at the core of the treatment plan. The questionnaires we translated in this study measures the extent to which patients follow the recommendations for their infusions, that is if they administer them correctly and in a timely manner. This allows researchers in this field to further study the impact of the treatment on the physical health and the quality of life of patients. This type of questionnaire is also necessary to study the personal and social factors that can influence patients to follow their treatment properly, and to measure the impact of interventions developed to support them.
when they were themselves in charge of performing the infusions than when young patients were autonomous. Preliminary psychometric analyses support the use of the French versions for the total scale and most subscales. However, the results did not support the use of the subscales Dose (VERITAS-Pro) and Plan (VERITAS-PRN) in the adapted versions. Further studies should be performed on larger samples.

**Subjects:** Health Psychology; Pediatric Nursing; Coagulation & Hemostasis; Pediatrics & Child Health

**Keywords:** Hemophilia; pediatrics; adherence; hematology; translation

1. **Introduction**

Hemophilia is a rare and inherited bleeding disorder caused by a deficiency in a coagulation factor, most often factor VIII (hemophilia A) or factor IX (hemophilia B). The severe form is characterized by spontaneous bleeding, predominantly in joints and muscles. Over time, repeated bleeding can lead to arthropathy, chronic pain, and impaired health-related quality of life (Gringeri, Ewenstein, & Reininger, 2014; Valentino, 2010). Joint bleeding can be greatly prevented with replacement therapy, which consists of intravenous infusions of the missing factor on a regular schedule (prophylaxis) or in case of injury or high risk for bleeding (episodic treatment) (Manco-Johnson et al., 2007).

In a recent literature review on adherence in hemophilia, non-adherence to prescribed prophylactic regimens was linked to a wide range of patient, condition and treatment-related factors, as well as healthcare system and socioeconomic factors (Thornburg & Duncan, 2017). Non-adherence was also shown to limit treatment effectiveness and resulted in poorer outcomes such as increased bleeding episodes and chronic pain (Thornburg & Duncan, 2017). In pediatric patients, poorer adherence can also result in a greater number of missed days of school (Krishnan, Vietri, Furlan, & Duncan, 2015). To prevent long-term damages in the joints, it is recommended that one should start prophylaxis early in life (Coppola, Tagliaferri, Di Capua, & Franchini, 2012).

Maintaining a rigorous treatment schedule can be challenging for patients, especially during adolescence, when patients learn to self-administer their treatment instead of their parents or caretakers doing so (Schrijvers et al., 2016). During the teenage years, adherence to treatment tends to decline drastically in hemophilia, as in other chronic illnesses (Geraghty et al., 2006a; Taddeo, Egedy, & Frappier, 2008a). The development of a reliable tool to assess adherence in hemophilia is crucial to the study of adherence, its determinant and its impact, especially in the more vulnerable populations such as adolescents. Accurately measuring adherence may usefully open on interventions designed to optimize adherence levels in order to improve bleeding prevention among this population.

The Validated Hemophilia Regimen Treatment Adherence Scale was developed to address this need (Duncan, Kronenberger, Roberson, & Shapiro, 2010a, 2010b). This standardized measure of adherence has two different versions to assess both adherence to prophylactic (VERITAS-Pro) and episodic treatment (VERITAS-PRN). The 24-item questionnaires include six subscales (VERITAS-Pro: Time, Dose, Plan, Remember, Skip, Communicate; VERITAS-PRN: Treat, Time, Dose, Plan, Remember, Communicate). Time refers to taking the treatment as schedule, Dose refers to infusing the prescribed dose, Plan refers to the organization around the supplies, Remember refers to the missed infusions, Skip deals with the doses purposely not administered, Communicate is for calling the treatment center when needed, and Treat makes reference to administering infusions when symptoms of bleeding occur. Total scores range from 24 to 120, and subscale scores range from 4 to 20 with lower scores indicating higher adherence. Both questionnaires have shown adequate psychometric properties in US samples (Duncan et al., 2010a, 2010b). Scores on the VERITAS-Pro and VERITAS-PRN have correlated moderately-to-strongly with global adherence rating by primary infusers and medical staff (Duncan et al., 2010a, 2010b). Scores on the VERITAS-Pro correlated as well as with the percentage of recommended infusions administered from a web-based self-report log system and validated against pharmacy dispensation (Duncan et al., 2010a).
Adaptations are now available in Spanish, Dutch and Brazilian Portuguese (Cuesta-Barriuso et al., 2017; Ferreira, Leite, & Duncan, 2018; Lock et al., 2014).

To date, no French-language version is available for the French-language patients and families. The objectives of the current study were to translate and adapt the VERITAS-Pro and VERITAS-PRN from English into French, to report preliminary psychometric data of the adapted versions, and to compare ratings between children and their parents.

2. Methods

2.1. Participants
Participants were recruited at the Sainte-Justine UHC Hemostasis Center. All children with severe hemophilia were approached if they had an appointment during the recruitment period and met the inclusion criteria: 6–18 years of age, French speaking, on prophylaxis and/or episodic treatment. The exclusion criteria were: diagnosis for less than one year or presence of psychiatric disorder as documented in medical records. Parents were also recruited as long as they spoke French. Written informed consent was obtained from all participants. The Sainte-Justine UHC Research Ethics Board approved this project.

2.2. Translation and adaptation process
Three bilingual translators familiar with the pediatric community participated in the study: two whose mother tongue was French (JSL and SB, see acknowledgments) and one whose mother tongue was English (LG, see acknowledgments). The methodology used to translate and adapt the questionnaires was based on the World Health Organization (WHO) Process of translation and adaptation of instruments (Organization WH, 2015).

2.3. Forward translation
The first step was a forward translation by two independent translators to produce two conceptually equivalent French versions of each the VERITAS-Pro and VERITAS-PRN. The focus was on cross-cultural and conceptual, rather than on linguistic/literal equivalence. The instruments needed to be easily understood by pediatric patients. Technical terms were avoided. The forward translations were then compared and discrepancies were discussed between translators until a consensus emerged to produce the intermediate French versions.

2.4. Back-translation
Using the same approach as that outlined in the forward translation, the intermediate French versions were then back translated into English by an independent translator. The translated questionnaires were then compared to the original English version. Differences were discussed to explore if important cross-cultural or conceptual aspects were involved. Modifications were made to achieve a pre-final French-version of each questionnaire.

2.5. Testing
The first 5 children were asked to give their general impressions of the questionnaires. They were asked if the items, instructions, and response scale were clear, easy to understand, and if there were any words they found difficult to comprehend, in line with the practice of cognitive interview (Hak, van der Veer, & Jansen, 2008). All recruited children and parents were asked to complete the questionnaires and to think of the specific treatment regimen assessed by the questionnaires when filling them. Participants were assigned to two different groups depending on whether the child or the parent was the primary infuser. Children and parents who were not the primary infusers did also complete the questionnaires for comparison purpose.

2.6. Psychometric and statistical analysis
Descriptive statistics (mean scores, standard deviations) were calculated for each subscale. Lower scores represent higher adherence. Comparison between raters’ scores we made using Wilcoxon
rank tests. For this, we paired the child and the parent’s estimate of adherence, i.e. their score on the Veritas-PRO or -PRN questionnaires. We used Mann-Whitney U tests to compare parents’ scores for each of the two categories (children as primary infusers vs. parents as primary infusers), and children’s scores for each of the two categories. Mean scores in the present study were also compared to the validation study using Cohen’s d where the difference is characterized as small (d = 0.20), medium (d = 0.50), or large (d = 0.80) (Cohen, 1992). Because of the small number of items in each subscale which might bias the alpha consistency coefficient, consistency was documented using inter-item correlations with values between .15 and .50 being considered acceptable (Clark & Watson, 1995; Streiner, 2003). For information purposes, Cronbach’s alpha levels (α) are given as supplementary material. Item-total and item-subscale correlations were calculated, with a value above 0.30 to be considered as acceptable (Nunnally & Bernstein, 1994).

As recommended, correlations were calculated by taking into account totals without the test item. For information purposes, we calculated the Pearson’s correlation (r) between adherence to treatment and the number of bleeding episodes in the last year as informed by the nurse. All statistics were performed using SPSS software (SPSS Inc., Chicago, IL).

3. Results
Twenty pediatric patients, aged 6–18 years old (M = 12 ± 4) with hemophilia A or B were recruited for this study. Of those, thirteen were able to complete the questionnaires (nine primary infusers and four whose parents were in charge of their treatment). The other ones were too young to be aware of their parent’s behaviors regarding infusion management and adherence. All participants were on prophylaxis in addition to episodic treatment in case of bleeding. The parents (thirteen mothers, seven fathers) completed the questionnaires. Demographic characteristics for the sample are displayed on Table 1.

Translators panel findings

3.1. Forward translation
The forward translation was conducted without major difficulties. The two versions of each questionnaire were very similar. Two significant issues were raised. The word supplies (items 10, 11, and 12, VERITAS-Pro) was translated to équipement [equipment] by translator 1 and to fournitures [supply] by translator 2. The concern was based on the fact that fournitures [supply] would better preserve the meaning of the English word but was thought to be harder to understand for children. Translators agreed to choose fournitures [supply] and to check for the understanding of the word during testing. Second, the word prescribed (item 15, VERITAS-Pro) was translated to prescrit [prescribed] by

| Table 1. Demographic sample characteristics |
|--------------------------------------------|
|                                            | Total | Child report n = 13 | Parent report n = 20 |
|                                            |       |                     |                     |
| Age in years, mean (SD)                    |       |                     |                     |
| Hemophilia type n (%)                      |       |                     |                     |
| A                                          |       |                     |                     |
| 11 (85)                                    |       |                     |                     |
| B                                          |       |                     |                     |
| 2 (15)                                     |       |                     |                     |
| Country of birth n (%)                     |       |                     |                     |
| Canada                                     |       |                     |                     |
| 12 (92)                                    |       |                     |                     |
| Other                                      |       |                     |                     |
| 1 (8)                                      |       |                     |                     |

Note: Mean age of children who were primary infusers was 14 ± 3 yrs for prophylaxis and 16 ± 3 yrs for episodic treatment.
Mean age of children whose parents were primary infusers was 10 ± 2 yrs for prophylaxis and 10 ± 3 yrs for episodic treatment.
translator 1 and to fournir [provided] by translator 2. Prescrit [prescribed] was chosen because fournir [provided] would refer more to the idea of a written schedule.

3.2. Back-translation

After the back-translation process, minor discrepancies were encountered (e.g. timing vs. calendar; inconvenient vs. bothers me; occur vs. happen; convenient vs. practical moment). All of those were discussed until a consensus emerged. Translators were unsure of how to translate I keep close track (item 10, VERITAS-Pro). Two expressions were proposed to the first five participants: 1) je suis de près [I follow closely] and 2) je fais un suivi rigoureux [I follow rigorously]. Translators expressed some doubts on how to translate the word convenient (item 6, VERITAS-PRN). They agreed to use convenable [suitable] and to check the understanding of the word during testing.

3.3. Testing

The first five participants (M = 14 ± 3 yrs) were interviewed. All participants mentioned that the questionnaires were clear, easy to understand, and easy to answer. For items 10 to 12 (VERITAS-Pro), all children reported understanding the word fournitures [supply], and therefore, the wording was maintained. For item 10, patients had the choice between 1) je suis de près [I follow closely] and 2) je fais un suivi rigoureux [I follow rigorously]. None of the children were sure of the meaning of rigoureux [rigorously]. Thus, the second option was chosen. For item 6 (VERITAS-PRN), none of the children were certain what convenable [suitable] meant. Some children suggested using pratique [practical] instead of convenable [suitable]. Thus, pratique was chosen. Finally, the word colonoscopie [colonoscopy] raised some questions from the children.

Preliminary psychometric data and Scores

3.4. Internal consistency

When examining inter-item correlations, internal consistency for VERITAS-Pro total scale and for most subscales was acceptable, except for the subscale Dose (total sample) and Plan (parent report only) for which correlations were low (see Table 2). For the Remember and Skip subscales, inter-item correlations were high (> .50) in the total sample reflecting items were very similar to each other and that there might be some redundancy. For VERITAS-PRN, inter-item correlations were acceptable for total scale and for most subscales (see Table 3). The Plan subscale was problematic with a very low inter-item correlation. The inter-item correlation in total sample was high for the Remember subscale pointing at some redundancy.

These results were consistent with the alpha analysis except for Dose and Communicate domains of the VERITAS-PRN for which the alpha coefficient for the parent report was low (.41 and .37), but had an acceptable inter-item correlation (.30 and .15). For problematic scales, discarding individual items did not manage to increase alphas to an acceptable level. Alphas indicated an excellent internal consistency for total scale.

3.5. Item analysis

All items were analyzed for item-total and item-subscale correlations (see supplementary file).

For VERITAS-Pro, item 14 from the Remember subscale (remember to infuse) was the one that contributed most to the total score with an item-total correlation of .78 for the total sample. For the total sample, none of the items of the Dose subscale correlated substantially with either their own subscale or to the total scale. It was also the case for item 9 of the Plan subscale (parent report only) and item 24 of the Communicate subscale.

In VERITAS-PRN, the most correlated item to the total score was item 20 also from the Remember subscale (remembering to follow the recommendations in case of bleeding) with an item-total correlation of .74 for the total sample. None of the items of the Plan subscale were correlated substantially to either their own subscale or to the total scale. It was also the case of
item 1 (child report only) and item 3 of the Treat subscale, item 12 of the Dose subscale (parent report only), item 17 of the Remember subscale (parent report only), and item 22 of the Communicate subscale.

3.6. Veritas-pro and VERITAS-PRN scores
Mean scores are presented in Tables 4 and 5. Total scores for the total sample ranged from 24 to 67 for the VERITAS-Pro and from 24 to 70 for the VERITAS-PRN. In general, the participants in our study had similar adherence scores to prophylaxis (M = 39.30 ± 12.10) as in the validation study (M = 41.2 ± 14.7, d = .14) (Duncan et al., 2010a). However, the parents of our sample, when they were the primary infusers, reported lower scores for prophylaxis (M = 31.00 ± 5.20, d = .50) than those in the validation study (35.3 ± 11.1), indicating higher adherence in our sample. Participants in our study also reported higher adherence for episodic treatment (M = 36.88 ± 10.56) than in the validation study (M = 46.5 ± 11.1, d = 0.89) (Duncan et al., 2010b). It is important to note that the participants of our study were not strictly on an episodic regimen as in the validation study (they also received prophylaxis), and that the validation study only included patients over the age of 16.

When they were primary infusers, children reported more skipping behaviors for prophylaxis (VERITAS-Pro, Skip subscale) than in parent estimates of children behaviors (Wilcoxon z = 2.032, p < 0.05). They also tended to report lower adherence to episodic treatment (VERITAS-PRN, total score) than their parent estimates (z = 1.926, p < 0.10). Also, whether the child (z = 1.826, p < 0.10) or
| Mean (SD)                  | Total               | Children as primary infusers | Parents as primary infusers |
|---------------------------|---------------------|------------------------------|-----------------------------|
|                           | All n = 33          | Child report n = 13          | Child report n = 9          |
| Total scale               | 39.30 (12.10)       | 44.62 (14.07)                | 47.56 (15.59)               |
| Time                      | 6.27 (2.39)         | 7.00 (2.45)                  | 7.67 (2.60)                 |
| Dose                      | 4.97 (1.38)         | 4.85 (0.99)                  | 5.00 (1.00)                 |
| Plan                      | 6.61 (2.62)         | 7.38 (3.15)                  | 7.89 (3.62)                 |
| Remember                  | 7.21 (3.09)         | 8.62 (3.31)                  | 9.11 (3.72)                 |
| Skip                      | 6.06 (3.05)         | 7.54 (3.48)                  | 7.78 (4.02)                 |
| Communicate               | 8.18 (3.55)         | 9.23 (4.09)                  | 10.11 (4.40)                |

|                           | Parent report n = 20 | Parent report n = 9          | Parent report n = 4          |
| Total scale               | 35.85 (9.48)         | 41.78 (10.38)                | 38.00 (7.75)                 |
| Time                      | 5.80 (2.29)          | 7.22 (2.68)                  | 5.50 (1.29)                  |
| Dose                      | 5.05 (1.61)          | 4.78 (0.83)                  | 4.50 (1.00)                  |
| Plan                      | 6.10 (2.15)          | 6.89 (2.71)                  | 6.25 (1.50)                  |
| Remember                  | 6.30 (2.64)          | 8.11 (2.37)                  | 7.50 (2.08)                  |
| Skip                      | 5.10 (2.36)          | 5.67 (3.24)                  | 7.00 (2.16)                  |
| Communicate               | 7.50 (3.07)          | 9.11 (2.89)                  | 7.25 (2.75)                  |

|                           | Parent report n = 11 | Parent report n = 11         |                           |
| Total scale               | 31.00 (5.20)         |                                |                            |
| Time                      | 4.64 (0.92)          |                                |                            |
| Dose                      | 5.27 (2.05)          |                                |                            |
| Plan                      | 5.45 (1.37)          |                                |                            |
| Remember                  | 4.82 (1.83)          |                                |                            |
| Skip                      | 4.64 (1.29)          |                                |                            |
| Communicate               | 6.18 (2.64)          |                                |                            |
| Table 5. Mean and standard deviation for VERITAS-PRN scores |
|----------------------------------------------------------|

| Mean (SD)       | Total                  | Children as primary infusers | Parents as primary infusers |
|-----------------|------------------------|-----------------------------|-----------------------------|
|                 | All n = 33             | Child report n = 13         | Parent report n = 20        | Child report n = 4 | Parent report n = 4 | Child report n = 9 | Parent report n = 16 |
| Total scale     | 36.88 (10.56)          | 42.69 (12.53)               | 33.10 (7.10)               | 48.25 (14.67)     | 34.00 (5.47)       | 40.22 (11.52)       | 32.88 (7.59)         |
| Treat           | 4.94 (1.39)            | 5.31 (1.49)                 | 4.70 (1.30)                | 5.25 (0.50)       | 4.75 (0.96)        | 5.33 (1.80)         | 4.69 (1.40)          |
| Time            | 6.52 (2.67)            | 8.15 (2.82)                 | 5.45 (1.99)                | 9.00 (2.31)       | 5.25 (1.26)        | 7.78 (3.07)         | 5.50 (2.16)          |
| Dose            | 5.91 (2.61)            | 6.23 (3.44)                 | 5.70 (1.98)                | 8.50 (5.80)       | 6.25 (1.71)        | 5.22 (1.20)         | 5.56 (2.06)          |
| Plan            | 5.33 (1.83)            | 5.77 (2.09)                 | 5.05 (1.64)                | 6.00 (1.83)       | 4.00 (0.00)        | 5.67 (2.29)         | 5.31 (1.74)          |
| Remember        | 6.00 (2.69)            | 7.15 (3.48)                 | 5.25 (1.74)                | 8.25 (4.35)       | 5.50 (2.38)        | 6.67 (3.20)         | 5.19 (1.64)          |
| Communicate     | 8.18 (3.51)            | 10.08 (4.01)                | 6.95 (2.56)                | 11.25 (2.99)      | 8.25 (2.87)        | 9.56 (4.25)         | 6.63 (2.47)          |
the parent \( z = 2.132, p < 0.05 \) was the primary infuser, children tended to report lower adherence for the Time subscale vs. parents’ report for episodic treatment (VERITAS-PRN, Time subscale).

Moreover, parents rated higher adherence on prophylaxis when they were themselves in charge of performing the infusion than when children were autonomous with infusions \( \text{Mann-Whitney } U = 11.50, z = -2.895, p < 0.01 \). In children’s report, this difference was not significant.

When comparing across domains of adherence within each instrument, the highest adherence was on the Dose subscale for prophylaxis (total sample, Friedman’s ANOVA, \( p < 0.01 \); Wilcoxon, \( p < 0.05 \)). For episodic treatment, the lowest adherence was on the Communicate subscale (total sample, Friedman’s ANOVA, \( p < 0.01 \); Wilcoxon, \( p < 0.05 \)).

Last year bleeding episodes and adherence

A significant correlation was found between adherence as reported by children and the number of bleeding episodes in the last year \( r = -.73, p < .01 \) with VERITAS-Pro scores, \( r = -.63, p < .05 \) with VERITAS-PRN scores). Counterintuitively, children experiencing more bleeding episodes in the last year reported higher adherence ratings. We will discuss this result and the possible role of behavior change in the next section. Interestingly, there were no significant associations when parent’s estimates were considered \( r = -.22, p = .36 \) with VERITAS-Pro scores, \( r = -.09, p = .72 \) with VERITAS-PRN scores).

To obtain the French-language versions, contact the author of the original questionnaires, Natalie A. Duncan (email: nduncan@ihtc.org).

4. Discussion

In hemophilia, the lack of a validated measure of adherence to treatment has been a barrier to adherence research in the French-speaking population. We translated into French the VERITAS-Pro and VERITAS-PRN using a standard procedure based on understandability by the pediatric population. We also reported internal consistency of both instruments in an independent pediatric sample. The questionnaires were adapted to French without major issues. Results of this preliminary study support the reliability of the French versions of the tools, especially when using total scores. However, the results suggest there may be difficulties with the internal consistency of the Dose subscale of the VERITAS-Pro and the Plan subscale of the VERITAS-PRN in our population.

During the translation process, we achieved a French version that was easily understood by both pediatric patients and their parents. Consistency was particularly low for the Dose subscale of the VERITAS-Pro. It is possible to think that all items might not have been interpreted the same way by parents or might not reflect the practice at our healthcare center. For example, taking a close look at specific items, we found that although all patients and parents reported always using the right dose, as many as 36% of our total sample answered that they increased or decreased the dose without calling the hemophilia center. In our treatment center and under certain circumstances, healthcare professionals may advise some patients to add an infusion or to slightly increase the dose prior to some specific physical activity. For those parents, positive answers may not represent a lack of adherence. Furthermore, none of the items of the Dose subscale were associated with either the total scale or the subscale score. It is possible that the low variability in scores impacted the size of the correlations as most participants judged their adherence to be high on Dose issues (with all of the participants answering being fully adherent to item 5).

The Plan subscale also had low internal consistency for both VERITAS-Pro (parent report) and VERITAS-PRN (parent and child report). It should be noted that the child report for this subscale might be biased because the management of supplies may well be under the parent’s responsibility without them necessarily being aware of their parent’s behaviors regarding those aspects.
Furthermore, low variability in scores may have impacted the size of the correlations, as adherence was generally good regarding planning aspects.

The VERITAS-Pro and VERITAS-PRN provided a detailed measurement of adherence. It thus may help identify areas where adherence could be improved. Communicating with the treatment center in case of questions or concerns received the lowest estimate of adherence for episodic treatment and showed some of the lowest adherence scores for prophylaxis. Working on establishing a good relationship with patients and their parents in order to have them willingly call their hemophilia center might be a key to increase adherence in pediatric patients. Time spent with the healthcare providers and good relationships with the nurse and the hematologist are factors associated with increased adherence in hemophilia (De Moerloose, Urbancik, Van Den Berg, & Richards, 2008; Tran et al., 2017). On the other hand, the Dose subscale received the highest adherence score for prophylaxis, just as it was the case in the validation study of the VERITAS-Pro (Duncan et al., 2010c). For the communication subscale, the standard deviation was the largest of all subscales in both questionnaires, which can be explained by large individual differences on those aspects. However, we also noticed that item 24 could lend itself to different interpretations. The wording “I call the treatment center when I have questions …” might have led some participants to answer about the occurrence of their questions rather than their reliance on their health care team when they have questions. We noticed the same issue in the original version.

In a worldwide survey, infrequent bleeding was ranked as the most common reason for non adherence (Geraghty et al., 2006a). In our study, we found that children's total scores positively correlated with past year bleeding episodes indicating that having serious bleeding episodes might act as a motivator for future adherence. Similar results were found in a Spanish validation study of the VERITAS-Pro on 13–62 year-old patients in which those who have had bleeding episodes in the last month had higher adherence (Cuesta-Barriuso et al., 2017). According to our results, this increased adherence might persist in time, even after the full recovery of hematrhosis. However, in our sample, parent report of adherence was not correlated with the number of bleeding episodes in the last year. It is possible that the way children answered the questionnaire was more influenced by their recent bleeding episodes or that parents were not always aware of their child’s behavior regarding adherence, especially when children were more autonomous with their injection.

Pediatric patients who were autonomous with their treatment also had a lower adherence (as reported by parents) compared to children whose parents were in charge of infusing. These results are consistent with studies finding higher adherence to treatment at a younger age when parents are the primary infusers (Duncan, Shapiro, Ye, Epstein, & Luo, 2012; Geraghty et al., 2006b). Decreased adherence at adolescence is common in many chronic illnesses with explanations ranging from emphasis on the present, inability to see the long-term consequences, testing limits, and so on. (Taddeo, Egedy, & Frappier, 2008b). In our study, there was also a pattern for children to report lower adherence than in parents’ estimates, but the differences were not significant across all scales. This could mean that parents are not always aware of all instances of non adherence from their older children, or that children underestimated their degree of adherence, basing their opinion on recent events of non adherence. It was also reported that parents of children with hemophilia experienced significant levels of guilt as compared to parents of healthy children, e.g., guilt over genetic transmission (Browne, Mally, & Kane, 1960; Kim, Kang, Cho, Song, & Ji, 2008). It is possible that this guilt makes them less inclined to report events of non adherence. This might have been particularly the case for the Time subscale of the VERITAS-PRN, which refers to infusing immediately when there are signs of bleeding instead of waiting for a more convenient moment. For this subscale, parents’ estimates tended to reflect systematically higher adherence than in the children’s reports, no matter who were the primary infusers. This is very consistent with the common observation in child psychology, that it is necessary to explore children’s behaviors through the views of both, children and parents.

We should recognize the limitations of this research. Firstly, the external validity of our results is limited due to the prophylaxis treatments being available and covered by the healthcare system in Canada, which may not be the case in other French-language areas, such as Africa or the
Caribbean. Secondly, the small sample size limited the scope of psychometric analyses and also prevented us from studying dimensionality using standard procedures like factorial analysis. Further studies should explore the consistency and validity of these adaptations of VERITAS-Pro and VERITAS-PRN in larger samples.

To conclude, we developed a French-language adaptation of questionnaires to assess adherence in hemophilia VERITAS-Pro and VERITAS-PRN. We confirmed feasibility and understandability in a pediatric population using patient report and parent report. Preliminary psychometric analyses support the use of the scales with good to excellent consistency for total scores in both questionnaires. However, concerns were raised about some subscales that may lack consistency. Perhaps individual items should be adapted to increase consistency. Future studies should address the reliability and validity of the scales in a French population.

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