Author's response to reviews

Title: Motivational techniques to improve self-care in hemophilia: The need to support autonomy in children

Authors:

Sarah Bérubé (sarah.berube.2@umontreal.ca)
Florine Mouillard (florine.mouillard@gmail.com)
Claudine Amesse (claudine_amesse@ssss.gouv.qc.ca)
Serge Sultan (serge.sultan@umontreal.ca)

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Author's response to reviews: see over
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Dear Editor,

The present letter refers to your reply dated July 14, 2015, concerning our manuscript submitted to BMC Pediatrics, “Motivational techniques to improve self-care in hemophilia: The need to support autonomy in children” (MS:7842706691481612). I thank you very much for your detailed comments and suggestions. Please thank both reviewers for their input.

You will find attached a revised version of this manuscript.

In the following lines, I will address each of the issues raised by the reviewers, explaining how and where a recommendation was implemented. The whole manuscript was re-examined and minor errors were corrected. We also sent the manuscript for a thorough English grammar check.

I hope this revised version will be suitable for your journal.

Very sincerely,

Serge Sultan, Ph.D
**Preliminary remark**

As recommended by referee 1, the first section of the discussion was placed in the background section. Thus, the line numbers changed in the current version as compared to the initial manuscript.

**Referee 1**

**Major Compulsory Revisions**

[1] Paper is written as a review paper, but the search strategy employed is not clear. Similarly, the decision to focus only on "Self-Determination" is not well-justified. It is unclear if the authors are aware of other approaches and the paper would benefit from a more systematic review of behavioural strategies/motivational training. The self-determination model is only one of many approaches and it is not clear why this is the focus of the paper.

Following your comment, we added a clearer explanation for using a motivational approach on lines 56-75. Specific arguments were added to support the choice of self-determination theory on lines 94-98 “Compared to other frameworks, such as motivational interviewing or empowerment, techniques from SDT can be incorporated into family dynamics and healthcare practices as a way to communicate to children about their illness and its management, and thus are specifically applicable to the developing child. It is also possible to use these communication tools with children of all ages.”

Importantly, the manuscript is designed as a feature paper, not a systematic review paper. We wish to attract the reader’s attention on alternative approaches to address adherence. Data on adherence to treatment in pediatric hemophilia are scarce. Most existing studies involve adults and focus on reasons provided by health-care professionals or patients to explain non-adherence. These studies have identified over 20 factors including “lack of time” or “not having infusions”, but these factors were not systematically correlated to the actual level of adherence and are difficult to interpret and use in practice, as they were not related to any theoretical framework. Our manuscript argues that important motivational factors underlie these issues, especially during adolescence (cf. background section). The main focus of the paper is to explain why it would be beneficial to adapt motivational understanding and motivational techniques to influence adherence in hemophilia, as is the case with other chronic illnesses requiring complex self-care behaviours.

[2] Reference 1 is a survey of health care providers. The initial part of the discussion should clarify this point (as it stands now, it seems to suggest that it reports patient-reported adherence).

Following this comment, we added “In a recent survey of healthcare providers in 147 treatment centers around the world…” on lines 10-11.

[3] The paragraph entitled "From treatment adherence to self-care" focuses on physical activity in hemophilia and is not linked well to the paper's thesis. This should be clarified further and the title should be adjusted.
This section was moved to the background section (as suggested by referee 1) and the heading was removed given that this paragraph and the preceding one both focus on adherence issues.

**Minor essential revisions**

[1] The paper needs a full review for English grammar. There are many errors. For example, "adolescent" is utilized in multiple places when "adolescence" is correct. In the abstract summary the term "confronted to" should be replaced by "confronted by".

The paper was entirely reviewed for English grammar.

[2] The first section in the Discussion re:adherence would be better placed in the background section.

This section was placed at the end of the background section.

[3] Key references should be added that are relevant to the hemophilia adherence literature to clarify why this is so important (examples: adherence and QOL Garcia-Dasí et al. Haemophilia 2015, adherence and improved pain McLaughlin et al Haemophilia 2014). Also, recent published data about self-management and educational strategies that have shown to have benefit may enrich the paper.

Thank you for these suggestions. As recommended, key references on the reported barriers related to adherence were added (line 14: Schrijvers et al., 2013; Nazzaro et al., 2006; Geraghty et al., 2006; Hacker et al., 2001; De Moerloose et al., 2008) as well as references on the effects of low adherence (line 6: McLaughlin et al., 2014; García Dasí et al., 2015). We did not add references on self-management or other educational strategies as the paper focuses on motivational strategies and how this could be implemented in hemophilia. Although previous work on education is fundamental these references may be confusing to the reader.

**Referee 2**

**General comments**

[1] The paper is a discussion of the potential application of self-determination theory (SDT) on enhancing pediatric hemophilia patients’ adherence to self-care. The topic is important and interesting in the field. I believe SDT will show great promises in this context. However, problems of logical presentation and clarity, and minor language errors present throughout the manuscript. The theoretical background of SDT is not descripted precisely and correctly. Some special terms used in the manuscript, such as non-autonomous motivation, major needs, self-motivation, do not appear to be in accordance to SDT or its related research. The proposed strategies based on SDT are not properly cited or they do not appear to be derived from the tenet of SDT. The authors should strengthen the theoretical background of SDT, and logically present the concept of SDTs and its constructs before making elaboration of how they could be applied in
the pediatric hemophilia.

Following your suggestion, the paper was reviewed so that the SDT approach would be well justified and more clearly presented to the reader. Following your comments, we decided to use the original terms from SDT when needed and add an explanation of these terms. The reference to non-autonomous motivation was removed as this sentence was changed, the term “major needs” was changed into “innate psychological needs” (line 99), and “self-motivation” was changed to “integrated regulation” (lines 84-). References were added to further support the proposed communication strategies (Ryan and Deci, 2000; line 126: Grolnick et al., 1997b; Joussemet et al., 2008b; Deci and Ryan, 2002; Reeve and Jang, 2006).

Specific comments

[1] Abstract Par1 Line 1-2: Who’s “adherence and selfcare”? We added “…in children and adolescents with hemophilia”.


[3] Abstract Par 1 Line 3: “Adherence rates” of who and of what? This unclear phrase appears throughout the manuscript. “Adherence rates” was changed to “Medication adherence rates of hemophilia patients”.

[4] Abstract Par 3 Line 4-5: “adherence in the developing child”, what does it mean? This was changed to “optimal adherence in adolescents as they move from parent-care to self-care”.

[5] P4 Line 2: adherence rate of what? Adherence is not meaningful to all behaviours, and its rate may vary a lot from one behavior to another.

We reformulated into: “Research has consistently found that adherence to treatment tends to decrease during late childhood and adolescence in patients with other chronic health conditions” (lines 7-9).

[6] P4 line 15-16: “as a child starts self administering factor infusion during early adolescence”, this phrase is difficult to be understood, and sound problematic.

It was changed to “during early adolescence, when self-administrating usually starts” (line 9-10)

[7] P4 line 15: “Adherence to prophylaxis regimens” reads quite confusing to me as a reader who
had no prior knowledge about how prophylaxis regimen are related to the topic of interest.

Following the comment, we decided to define prophylaxis and episodic treatment in the first few sentences of the background section.

[8] P4 Line 16: “90% of children” who are patients of what?? I think same issue appears throughout the manuscript. It should be clarified.

As recommended, we changed this to “90% of children with hemophilia” and made sure to correct this omission in the manuscript. (line 11-12)

[9] P4 Line 20: “when confronted to a bleed” sound confusing to me again. Readers have no clue about what it means.

In the first sentences of the background section, we added a description of hemophilia and of bleeding episodes. We also changed “when confronted to a bleed” for “when bleeding occurred”. (line 14-15)

[10] P4 Line 25: “factor prophylaxis”, what does it mean?

Following your comment, we decided to define prophylaxis and episodic treatment in the first few sentences of the background section.

[11] P5 Line 27-32: The statements do not read very logical and derived based on evidence. Also, the authors should explain how they are linked to the topic of discussion in the paper.

The purpose of this section is to explain how motivation for treatment might be an issue for adolescents. Following your comment, we added a reference (Taddeo et al., 2008) to support our statements and added the following description to explain how the statements are linked to the topic: “Motivational factors appear central to adherence and self-care behaviors so that patients do not see their treatment plan as being imposed on them by their caregivers.” (line 28-29)


Thank you for this reference. It was added to the manuscript: “It has been shown that fostering motivation in patients can lead to better adherence to the recommended exercise [11]” (line 33-34).

[13] P6 Line 51: Does “while parental supervision progressively decreases.”? Perhaps a citation would help strengthen this argument.
We added reference (Lindvall et al., 2006) to support this specific argument. In this article, the median age to start self-infusing was 11.6 years and children became autonomous at a median age of 14.1 years (line 62).

[14] P6 Line 51-57: The terms from SDT (e.g., motivation, autonomous motivation) just arise without explanation. Perhaps their definitions need to come first before they are mentioned.

As mentioned earlier, we decided to define each term from SDT.

[15] P6 Line 58-59: Autonomous motivation is a concept from SDT, so this sentence sounds quite awkward.

We changed “autonomous motivation” for “motivation” (line 78).

[16] P6 Line 59-60: “they are at the center of their actions” do not appear to be the AIM of SDT.

Following this comment, we decided to remove this sentence and replaced it with: “According to this approach, it is possible to set limits without precluding children from becoming inherently motivated for their behavior and thus motivated to participate in activities that are not necessarily pleasant, such as infusions (Koestner et al., 1984)” (line 79-81).

[17] P6 Line 62-63: What is “non-autonomous motivation”? As far as I know, SDT does not have this term or construct. I think the authors refer to controlled motivation.

Indeed, we were referring to controlled motivation while trying to avoid jargon for unfamiliar readers. Following your comment, we decided to change this sentence: “A child would be motivated by external factors if he/she engages in the recommended activities in order to obtain parental approval, rewards or to avoid guilt” (line 85-86).

[18] P7 Line 81: What is autonomy support? How it is defined under SDT, and how it is related to autonomous motivation?

We defined and used “integrated regulation” instead of using the term “autonomous motivation”. To explain how autonomy support is related to integrated regulation, we added a description on lines 98-103: “The sets of communication techniques derived from SDT aim at fulfilling an individual’s innate psychological needs for autonomy, competence and relatedness, which in turn can facilitate and foster a more integrated form of regulation [31]. Among these needs, autonomy would be essential in achieving a more integrated form of regulation and for this reason, many interventions are referred to as “autonomy-supportive” even though they also respond to needs for competence and relatedness [31].”

[19] P7 line 85: “major needs” a term from SDT? I believe the author refer to basic psychological needs. Again, we need more solid theoretical explanation before it can be discussed and be applied in this manuscript.

Following this comment, we decided to use “innate psychological needs”, which is the term
used by Ryan and Deci (2000) (line 99 and after).

[20] P7 Line 85-97: Some of the autonomy supportive strategies proposed do not appear to be in line with SDT or research based on SDT, please provide citations or more elaborations.

Line 126: We added references from which these techniques were adapted (Grolnick et al., 1997b; Joussemet et al., 2008b; Deci and Ryan, 2002; Reeve and Jang, 2006).

[21] P7 Line 87: “When the child is ill” sounds weird. Who is THE child?

This sentence was changed to: “However, this can represent a challenge with a child with hemophilia…” line 126-228.

[22] P8 line 108: “Multiple, complex behaviors are necessary for appropriate self-care.”, where does this statement derive from?

This sentence was removed as it was not essential in the summary.

[23] Table 1 line 2: Use “children” or “his/her feelings”

As recommended, this was changed to “his/her feelings”.