

Abstract

The World Federation of Hemophilia recommend comprehensive care that prioritises prevention and prompt management of bleeding, joint damage and complications, as well as regular exercise and physical activity to stimulate normal psychomotor development, develop strong muscles, balance and coordination, maintain healthy body weight and improve overall fitness. However, fear still persists around the risks of participating in physical, especially vigorous, activities, despite the availability of prophylactic treatment that makes it possible for children and adolescents to have fewer spontaneous bleeds and secondary complications. Reasons for poor exercise adherence include overprotection by parents and carers and the belief that joint disease and bleeding cannot be prevented. Especially in the early years, parents are anxious regarding their child's participation in physical activities and sports. Having knowledge of haemophilia and the appropriate physical activities for young people would help parents, carers and teachers gain confidence.

We propose an educational resource for parents, carers and teachers that is designed to improve their knowledge and confidence in discussing, encouraging and managing exercise in young people with haemophilia. We will evaluate the effectiveness of this learning tool in bringing about change or intention to change.

Lead organization: In Vivo Academy Limited

Main collaborators:

- Haemophilia Foundation Australia
- Haemophilia Foundation of New Zealand
- ProPatient

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LOI Reviewer comments

Overall, the panel felt this to be an extremely well-constructed and well-written proposal, and it was unanimous that this LOI should go forwards to full proposal stage. No specific weaknesses were highlighted, although a panel member felt that the proposal could be strengthened even further with additional assessment methods aimed at collecting data on QoL and actual changes to exercise routines, and it was felt that the full proposal should contain details of the experts involved in content development.

How we addressed the comments

After discussion with the Haemophilia Foundation Australia (HFA) and the Haemophilia Foundation of New Zealand (HFNZ) regarding the LOI feedback, both groups recommended asking the target audience to evaluate the effectiveness of the learning tool in instigating change or intention to change behaviours as an additional, strengthened assessment method. However, they advised caution with questions to the patients assessing quality of life changes as this would be construed as human research and would require a research workup and ethics processes beyond the scope and budget of this project.

Details of the experts involved in content development are listed in [Organizational Details](#).

Proposal

1. Overall goal and objectives

The goal of our proposed activity is to improve the knowledge and confidence of teachers and parents/carers in discussing and managing exercise in younger haemophilia patients.

Upon completion of our proposed activity, users of the educational resource will be able to: (1) list the benefits of regular exercise for people with haemophilia (PWH); (2) describe particular exercises and sports that are suitable for PWH based on their individual body make-up and existing joint conditions; (3) be motivated to facilitate and encourage regular and appropriate exercise for PWH; and (4) describe steps to take in the event of a bleeding episode while a PWH is participating in exercise/sport.

2. Current assessment of need in target area

Haemophilia affects approximately 2850 people in Australia and 440 people in New Zealand^{1,2,3,4,5} but advancements and new drugs in recent years allow PWH to live longer, with improved quality of life.⁶ The World Federation of Hemophilia recommend comprehensive care that prioritises prevention and prompt management of bleeding, joint damage and complications, as well as regular exercise and physical activity to stimulate normal psychomotor development, develop strong muscles, balance and coordination, maintain healthy body weight and improve overall fitness.¹ In Australia, only 43% of children with haemophilia aged between 4 and 18 years old meet the physical activity guidelines recommended by the government. Less than 8 hours per week, including 3.8 hours of vigorous physical activity, is the median level of physical activity for these children.⁷ Australian children with haemophilia also have lower disease-specific quality of life related to school and sports compared with children from several European countries.⁸

While being involved in physical activity at any age has both general and haemophilia-specific health benefits and may reduce the use of prophylactic clotting factor products due to increase in endogenous factor VIII activity, it is important for PWH to start engaging in physical activities at a young age to prevent the physical signs and biomechanical changes of sedentary behavior later in life.^{9,10} Limited physical activity in children with haemophilia has been found to result in skeletal and muscular impairments, problems with weight control and increased anxiety.^{7,11} However, while the availability of prophylactic treatment makes it possible for children to have fewer spontaneous bleeds and secondary complications,¹² the fear of the risks of participating in physical, especially vigorous, activities still persist.⁹

Some of the reasons for poor exercise adherence include overprotection by parents and the belief that joint disease and bleeding cannot be prevented.¹⁰ While a child may feel capable of participating in school activities, he/she may have apprehensions about interacting with peers. Williams et al. speculate that the parents may have influenced their child into this thinking. Parents have been found to be more anxious than the children themselves about the latter's participation in school and sports. As with any other chronic diseases, parents often overestimate the physical and social discomfort of their child with haemophilia.¹³ The early years of the child's life when parents lack the knowledge and confidence in optimally

managing haemophilia are often described as a time when they are “feeling out of control”, their “emotional well-being [is] vulnerable” and when quality of life is poor.¹⁴

As parents become resilient, they seek new knowledge, which gives them the confidence to further socialize and look at haemophilia in a more positive light. They also act as “advocate[s] for their child[ren] and [ensure] that appropriate safety procedures [are] in place in order to avoid bleeds and injuries”.¹⁴ As much as they tend to be very protective due to the child’s need for treatment, parents aspire to a normal life for their child. Because of this, they actively seek knowledge about haemophilia, especially its management and coping with it.¹⁵ Being knowledgeable about haemophilia also lets them regain control over their lives, better manage uncertainty, and impose fewer restrictions on the child.¹⁴

The studies that explored the different strategies to support parents in raising children with haemophilia showed that imposing restrictions on the child’s activities worsens the parents’ concerns and stress. Also, parents have to take the lead and be assertive in their communications with health professionals to “get what they believed [is] the best for their child”. According to Khair and Chaplin “the ongoing impact of having a child with haemophilia [is] characterized by awareness of the importance of resilience to external (dealing with other people) and internal (perceived threat to the child and family) challenges”. Support from family and community, of specialist health care staff, and involvement with haemophilia society are all deemed crucial.¹⁶

Transitioning through life stages can be challenging, more so for people with chronic conditions like haemophilia. Moving away from parental- to self-care and creating new friendships, young adolescents need to arm themselves with the right information about their condition to be more self-sufficient in terms of dealing with other people and being compliant with prophylaxis injections, exercise and rehabilitation regimes.^{17,18}

Physiotherapists in the HFA needs assessment report state “it [is] important that young people receive[d] “good advice in regards to bleed management, ‘at risk’ activities and likely prognosis of musculoskeletal conditions with particular sports/activities” so that they could make informed decisions about activities such as contact sports, running, skateboarding etc.” They also need to prepare for their eventual independence from their parents and “before they start to disengage from their Haemophilia Centre during their early teenage years”.¹⁸

Resources to educate parents, schools (especially teachers) and the wider community on bleeding disorders are recommended to better understand the situation, debunk myths, and ensure that safety and psychosocial issues are addressed appropriately.¹⁸ Haemophilia treatment centres can provide education and reinforce healthy lifestyle advice, and physiotherapists initiate or coordinate exercise programmes. However, because both Australia and New Zealand have vast rural and remote populations, access to treatment centres is recognised as a barrier. To overcome the distance, telehealth or the delivery of health care or education through communication technology is currently used in some treatment centres to connect to local clinics.¹⁹

3. Target audience

The target audience for our proposal includes patients, parents/carers and teachers, especially physical education (PE) teachers of PWH in the 7–16-age group. The haemophilia foundations (on the state and territory levels) would promote the resource to parents. Parents, with the involvement of haemophilia treatment centres, almost always initiate the communication with their child's school regarding the availability of educational resources on haemophilia for teachers.

Sharon Caris, Executive Director of HFA believes that an educational activity targeting children in early–late primary age and adolescents is most appropriate “before kids start doing too much sports and exercise and getting into bad habits” (S. Caris, pers. comm., 26 May 2017). Suzanne O’Callaghan, Policy Research and Education Manager of HFA agrees, “So joint damage can be prevented and good practice developed early.” (S. O’Callaghan, pers. comm., 7 September 2017).

Ms Alison Morris, a very experienced physiotherapist at the Princess Margaret Hospital Haemophilia Centre in Perth, Australia, added that children in the age range of 12–16 years may be experiencing challenges in self-administering prophylactic clotting factors and therefore should be supported and advised with regard to the activities and sports that are not suitable for them (A. Morris, pers. comm. 15 September 2017). Their parents and teachers would benefit from this educational resource.

4. Project design and methods

Format and content

The planned activity is a series of ~40 short-burst (ranging from 30 seconds to no more than 2 minutes each), inter-linked Q&A videos and simple animations that are learner-directed, allowing the users to understand the issues surrounding exercise for young PWH.

The education resource will feature a haematologist and two physiotherapists (from Australia and New Zealand) covering the following topics:

- Practical guidance and explanations on recommended sports and physical activities for PWH who want and can do them based on body make-up and existing joint problems, and their associated risks (e.g. video showing a problematic gait or injury)
- Typical bleeding problems and injuries/complications in the target age group, how to monitor bleeding issues and modify activities in the school environment so that the student with haemophilia can participate (rather than be sent to the library, for example)
- What to do when PWH have had a bleed and are in rehabilitation phase (e.g. guidance for parents when sending their child to school)
- Practical guidance on joints, joint bleeds, joint destruction, factor replacement therapy and trough levels using animations

Supplementary educational materials will be provided as links from the videos for users who may be interested to expand their knowledge and understanding of the topic.

Evaluation

The activity will be evaluated by asking participants to evaluate the effectivity and impact of the educational resource in bringing about behavioural change. For example:

- After completing this activity, how likely are you to encourage a PWH (e.g. a family member, a student with haemophilia) if he/she wants to or can participate in appropriate physical activity?
 - To a great extent
 - Somewhat
 - Very little
 - Not at all
- Would you be willing to be contacted in 4–6 weeks' time to evaluate the impact of this educational activity on the PWH's exercise habit

We will also assess how the educational resource affected the learners' perceived gains in independence, interpersonal relationships, and overall well-being.

The planning committee would include a paediatric haematology specialist, two physiotherapists with expertise in haemophilia from Australia and New Zealand, a health behavioural change strategist and representatives of HFA, HFNZ, ProPatient and In Vivo Academy. The plan would focus on addressing the need to inform young PWH, parents carers and the relevant school personnel and/or coaches about the benefits of exercise for PWH, and the types of exercise that are most appropriate and beneficial. The proposed project would also help dispel misconceptions about exercise for PWH, to motivate PWH to exercise and motivate carers and teachers to facilitate this, and to inform young PWH, carers and teachers/mentors and coaches how to address bleeding episodes should they occur.

The series of videos will be accessible from the websites of the HFA and HFNZ, through a link to the ProPatient website (<http://propatient.com/>). The supplementary educational materials would be available as a PDF, downloadable from both foundation websites. The activity would also be promoted via the HFA and HFNZ social networking sites (Facebook/Twitter), their mailing lists and advertising in their respective newsletters, directly promoting to primary schools and haemophilia treatment centres in Australia and New Zealand. Because treatment centres liaise directly with parents and with schools and teachers where the PWH attend, this proposed educational resource will strengthen their recommendations for appropriate exercise and physical activities.

Innovation

With an eye to a learning environment that is supportive and productive, promotes self-motivation, and provides individualised feedback, the proposed programme would afford learners the opportunity to acquire knowledge and confidence in a consequence-free, personalised environment. The short videos link through an advanced branching logic system (similar to "choose your own adventure" discovery narrative approach), each designed to empower learners with clinically relevant content and coaching. These videos are meant to be consumed at learners' own pace and in accordance with their own interest. (See <http://app.propatient.com/visits/living-with-pain/scene/575eb61f2550861000ca7018>.)

Throughout the activity quizzes and surveys will be made available to track knowledge changes, confidence and confidence gaps and relevant demographics.

5. Evaluation and Outcomes

This activity will track all learner choices anonymously and will aggregate all insights and outcomes. Supporters and partners will receive a detailed outcomes analysis of all choices made within the simulation twice annually, and monthly user metrics will be reported. Outcome metrics may include: (1) user metrics, including number of users and levels of engagement; (2) users' areas of interest and most commonly asked questions; (3) self reported psychosocial issues or queries; and (4) a variety of survey questions related to the therapeutic area and learner demographics.

Follow-up surveys will be sent via two processes: (1) to those who opt-in to participate as a result of receiving the invitation to access the education resource; and (2) widely (and rolling) via the foundations' mailing lists and social media platforms, with the condition that the respondents access the resource first and then return to complete the survey after a specific timeframe.

We plan to publish the outcomes of the project as an abstract in a local or international haemophilia conference (to be determined) or at the World Federation of Hemophilia International Musculoskeletal Congress in 2020 in Kuala Lumpur, Malaysia; and as a report in *HFA National Haemophilia Journal*.

6. Detailed work plan and deliverables schedule

Time frame	Workplan
November–December 2017	Planning Planning with HFA, HFNZ, the haematologists and physiotherapists from Australia and New Zealand and the clinical psychologist with expertise in behaviour change
December 2017–March 2018	Development of content and materials <ul style="list-style-type: none">• Writing of outlines and scripts to be reviewed and approved by the planning team• Video shoot with the haematologist and physiotherapists
February 2018	Start of marketing initiatives
April–November 2018	Educational resource available online

May–December 2018	6-week rolling follow-up survey
November–December 2018	Data analysis
November 2018– January 2019	Publication plan and submission