# Understanding health and treatment decision-making among youth with hemophilia: a qualitative approach

**Shannon Lane[1], Irwin Walker[2], Anthony Chan[3], Nancy Heddle[1], Emmy Arnold[1], Man-Chui Poon[4], Leonard Minuk[5], Lawrence Jardine[6], Ian Chin-Yee[7], Kathryn Webert[8]**

*[1]Medicine, McMaster University, Hamilton, ON, Canada; [2]Division of Hematology and Thromboembolism, Medicine, McMaster University, Hamilton, ON, Canada; [3]Pediatrics, McMaster University, Hamilton, ON, Canada; [4]University of Calgary, Foothills Hospital, Calgary, AB; [5]Division of Hematology, Medicine, London Health Sciences Centre, University of Western Ontario, London, ON, Canada; [6]Paediatric Hematology/Oncology, Children's Hospital, London, ON, Canada; [7] Division of Hematology, Medicine, University of Western Ontario, Medicine, London Health Sciences Centre, London, ON, Canada; [8]Molecular Medicine and Pathology, McMaster University, Hamilton, ON, Canada; Medical Director, Utilization Management, Canadian Blood Services, Hamilton, ON, Canada*

## Introduction

The first generation of young people with hemophilia to have grown up using prophylaxis are making the transition from childhood to adolescence and early adulthood; a time of physiological and psychosocial change. Though the benefits of primary prophylaxis for preventing joint damage and arthropathy are now well established[1-3] a variety of unresolved issues related to prophylactic treatment persist, including: the duration of prophylaxis; can and should it be stopped, and at what age[4-7]; and can and should prophylactic treatment be tailored or individualized.[8,9] The reasons why some individuals do not follow treatment recommendations and/or discontinue prophylaxis into early adulthood are also not well understood. To date research in these areas has relied predominantly on quantitative methods when the issues and variables are relatively simple and known, which are appropriate for measuring and analyzing causal relationships between variables.[10] However, when knowledge in an area is lacking and the issues are complex (such as with hemophilia treatment decision-making), qualitative techniques are better suited for exploring and uncovering the breadth, depth and range of an issue. It is towards understanding these non-medical determinants of health and treatment decision-making that this research is directed.

## Objectives

**General**: This qualitative study was undertaken to explore attitudes about health and living with hemophilia, and develop a conceptual understanding of treatment decision-making from the perspectives of young men (15-29 years) with severe hemophilia A or B in Canada.

**Specific:** **1)** To identify factors that affect young peoples hemophilia management and treatment decisions and explore inter-relationships between factors;
**2)** To develop an understanding of treatment decision-making to assist healthcare providers with communication strategies to ensure optimal, individualized client care.

## Methods

**Data Collection:** Data were collected using semi-structured interviews and managed using QSR International's NVivo 9 software.

**Data Analysis:** This study employed the Framework method (NatCen, UK) of qualitative analysis; which includes the processes of: familiarization, the identification of a thematic framework, indexing, charting, mapping and interpretation.

## Results

A total of fifteen interviews, lasting between 45-90 minutes, were conducted (in-person or by phone) with participants aged 15-29 years, recruited from three sites across Canada. The major factors involved in treatment decision-making identified by this study were: disease knowledge and information (acquired from: the hemophilia treatment centre, parents, involvement in hemophilia community, and lived experience including: bleeding history, presence or absence of trouble or target joints and/or inhibitors); childhood disease experiences and type of treatment; activity level; and physical and situational cues to treat. The Framework method facilitated analysis of the inter-relationships between factors and resulted in a typology of four different 'types' or approaches to treatment: routine prophylaxis – rigid, routine prophylaxis – lifestyle cues, situational prophylaxis, and no prophylaxis. The typology is presented in detail in the table below.

## Treatment Typology

| Name and Description of Type | What individuals in this type have in common… | Why might they do what they do… |
|---|---|---|
| **Routine Prophylaxis – Rigid [n = 4]**<br>Treats routinely, rigidly follows a schedule, does NOT use cues | • Lots of bleeds as children<br>◦ (2 with inhibitors, 2 not on routine prophylaxis as children)<br>• Have trouble/target joints<br>• Are NOT very physically active | • These individuals have had difficult experiences of hemophilia as children (due to inhibitors, lack of routine prophylaxis), which led to reduced quality of life<br>• Currently all doing much better, (infrequent bleeds)<br>• They "buy-in" to the benefits of treatment and know what they want to avoid |
| **Routine Prophylaxis – Lifestyle Cues [n = 6]**<br>Treats routinely, follows a schedule AND uses lifestyle cues (sport or activity) to assist with decision-making, (i.e.: the 2nd day, or the 3rd day) | • Majority have had a lot of childhood bleeds<br>• Majority have not had inhibitors<br>• Majority have target joints (except 1 individual who is 15 years)<br>• All relatively active in sport<br>• All responded 'yes' or 'sometimes' when asked whether they think about the long-term implications of treatment decision-making | • These individuals are quite physically active<br>• They are thinking about the long term consequences of their treatment decisions because they already have trouble or target joints and want to continue to maintain their current physical state and quality of life so they can continue to be active<br>• Aware of the risks involved in sport/activity they infuse beforehand to reduce likelihood of a bleed and/or damage |
| **Situational Prophylaxis [n = 4]**<br>Treats rarely, (once/week to once/month), NOT according to a routine schedule, BUT uses situational cues (work, sport, etc.), and physical cues (target joints/ bleed risk) to assist with decision-making. | • Majority never developed a prophylaxis routine during childhood<br>• None have ever had inhibitors<br>• All have trouble or target joints<br>• None are very active<br>• All 26 years or older at time of interviews | • These individuals are older, have reached an age when the effects of multiple bleeds have "set in" resulting in trouble or target joints<br>• They are not very active, but treat preventatively before engaging in sport/activity/work, to prevent further damage<br>• Having never developed a routine prophylaxis habit during childhood, they are unlikely to do so now |
| **No Prophylaxis [n = 1]**<br>Does not treat preventatively, sometimes chooses NOT to treat small tissue bleeds. Sometimes, eventually, pain is the cue to treat. | • Only one individual in this 'type' – an outlier or are there others?<br>• Trial period of routine prophylaxis during childhood, (for < than 6 month), primarily treated bleeds 'on demand'<br>• Deals with bleeds in his, "own way," according to his own beliefs, "not everything needs an infusion" | • This individual does not want to be on routine prophylaxis<br>• This individual does not feel he has all the knowledge about hemophilia that he needs; acknowledges he could learn a lot more<br>• Describes himself as 'stubborn' and raised the concept of self-image, expressed discomfort with the world knowing he has hemophilia<br>• Does not think about the long term consequences of treatment decisions, "Unfortunately I don't, when I … I should" |

## Conclusions

Treatment decision-making among young men is complex, affected by numerous different factors. Analysis of the inter-relationships between factors revealed important differences in how individuals with hemophilia approach and make decisions about treatment using factor concentrate. Until now, this variation in approaches to treatment had not been systematically explored and documented in the field. Future research should be directed towards validating the typology using a larger sample. Knowledge of the typology and four unique types can be used to enhance client-provider communication, treatment planning and care. These findings can also be used to individualize hemophilia treatment, care and education and enhance the development of educational tools and intervention.

## References

1. Manco-Johnson MJ, Abshire, TC, Shapiro AD, Riske B, et al. (2007). Prophylaxis versus episodic treatment to prevent joint disease in boys with severe hemophilia. *N. Engl. J. Med.,* 357(6):535-44
2. Feldman BM, Pai M, Rivard GE, Israels S. (2006). Tailored prophylaxis in severe hemophilia A: interim results from the first 5 years of the Canadian Hemophilia Primary Prophylaxis Study. *Journal of Thrombosis and Haemostasis,* 4(6):1228-36
3. Petrini P, Lindvall N, Egberg N, Blomback M. (1991). Prophylaxis with factor concentrates in preventing hemophilic arthropathy. *Am. J. Pediatr. Hematol. Onco*l., 13(3):280-7
4. Hay CR. (2007). Prophylaxis in adults with haemophillia. *Haemophilia.*, 13(Suppl. 2),10-5
5. Richards M, Altisent C, Batorova A, Chambost H, et al. (2007). Should prophylaxis be used in adolescent and adult patients with severe haemophilia? An European survey of practice and outcome data. *Haemophilia , 13*(5):473-9.
6. Van DK, Fischer K, van der Bom JG, Scheibel E, et al. (2005) Can long-term prophylaxis for severe haemophilia be stopped in adulthood? Results from Denmark and the Netherlands. *Br. J. Haematol., 130*(1):107-112.
7. Astermark J. (2003). When to start and when to stop primary prophylaxis in patients with severe haemophilia. *Haemophilia 9*(Suppl. 1):32-36
8. Fischer K. (2012). Prophylaxis for adults with haemophilia: one size does not fit all. *Blood Transfusion.,10(2): 169–173*
9. Franchini M and Mannucci PM. (2012). Prophylaxis for adults with haemophilia: towards a personalised approach? *Blood Transfusion*,10(2):123-4
10. Morse JM. (1994) Designing funded qualitative research. In: Denzin NK, Lincoln YS, (Eds.) *Handbook of Qualitative Research*. (pp. 220-235). Thousand Oaks: Sage Publications, Inc.

## Acknowledgements

This project was funded through the *Care Until Cure* research program which is sponsored by the Canadian Hemophilia Society and Pfizer.

Poster printing services supported by Baxter Bioscience.

WFH2012 274--We
Youth Issues
Shannon Lane
DOI: 10.3252/pso.eu.WFH2012.2012
Poster presented at: WFH 2012 World Congress

PosterSessionOnline.com