sciendo

COMMENTARY

Uncertainty to normality using ethnographic and qualitative research: a personal view

David Page

It is fascinating for a person born with haemophilia almost 70 years ago to read the very insightful paper 'What more can we ask for?: an ethnographic study of challenges and possibilities for people living with haemophilia' by Hughes et al. in this issue of the *Journal of Haemophilia Practice*. While medical treatments have advanced miraculously over the last seven decades, from fresh frozen plasma to cryoprecipitate and a whole range of factor concentrates, to non-factor therapies and now to the cusp of gene therapy, the disease itself has remained unchanged as has human nature. The key challenges uncovered in the research – 1) the quest for 'normalcy'; 2); the clash between outcomes desired by clinicians and those pursued by patients; 3) struggles in understanding health care providers and applying their recommendations to an individual's everyday life in the context of chronic disease; and 4) life-stage-specific challenges – are as acutely experienced in people with haemophilia (PwH) today as they were in the 1950s.

©Shutterstock

Trying to appear 'normal' as a PwH in the 1950s and 1960s was admittedly more difficult. Even in those parts of the world with the most advanced care, long hospital stays, frequent surgeries, early joint damage and barriers to regular schooling were routine. Not wanting to have to miss out on life, PwH developed numerous tricks to hide their bleeds from parents to avoid hospitalisation. Between episodes of bleeding, however, PwH tried very hard to be 'normal'. They ditched their crutches

This commentary relates to the following papers by Thomas Hughes, Mikkel Brok-Kristensen, Yosha Gargeya, Anne Mette Worsøe Lottrup, Ask Bo Larsen, Ana Torres-Ortuño, Nicki Mackett and John Stevens, published in the *Journal of Haemophilia Practice* 2020; 7(1):

- "What more can we ask for?": an ethnographic study of challenges and possibilities for people with haemophilia. *https://doi.org/10.17225/jhp00151*
- "He's a normal kid now": an ethnographic study of challenges and possibilities in a new era of haemophilia care. *https://doi.org/10.17225/jhp00167*
- Navigating uncertainty: an examination of how people with haemophilia understand and cope with uncertainty and protection in an ethnographic study. *https://doi.org/10.17225/jhp00168*
- Treating for stability: an ethnographic study of aspirations and limitations in haemophilia treatment in Europe. *https://doi.org/10.17225/jhp00169*

DAVID PAGE
National Director of Health Policy, Canadian Hemophilia Society

and played sports with their buddies, invited girls to school dances, and usually managed to graduate from high school despite missing months of classes. Yet this paper uncovers many similarities with the past. Many of today's PwH still experience regular bleeds, require mobility aids, have chronic pain, and experience barriers to schooling and employment. They develop their own strategies to appear 'normal' to themselves and to the world: not adhering to recommended prophylaxis, putting the disease 'in the background'. They are encouraged in this perception of 'normalcy' by frequent comparisons to the past: their uncles and grandfathers lived through the days of plasma and cryo, and the horror of blood-borne infections. They, on the other hand, are living in the Golden Age of haemophilia; treatment has vastly improved. Moreover, they classically exhibit the *disability paradox* in which people with serious and persistent health challenges report that they have a good or excellent quality of life when most external observers, walking in their shoes for a day, would have an entirely different experience.

Parents attending patient association meetings in the middle of the last century were obsessed with two questions: "How can I protect my little boy from all the risky activities he wants to do?" and, "What is the balance between letting a boy be a boy and wrapping him in cotton batten?" Encouraged by clinicians, many parents went to enormous lengths to protect their children from bleeds, usually without much success. Others took a different approach, denying the dangers, and being overly permissive. The research in this paper reveals that this dilemma – the search for the perfect balance between activity and risk – is still present. Clinicians and PwH still have different perspectives. Clinicians may focus on measures to reduce risk: adherence to prophylaxis, minimising bleed rates, raising trough levels; PwH focus on their lives: the pain they experience, how that pain limits participation in desired activities and how bleeds, regardless of their frequency, have a negative impact on their enjoyment of life. These differing goals do not lend themselves to physician/patient alignment in achieving optimal health outcomes. They underline the need for personalisation of treatment in which the answer to the question "What matters most?" leads to personalised solutions.

The paper uncovers that, as in the past, a large percentage of PwH entrust their treatment-related decisions to their health care providers, despite recent research on shared decision-making that shows this practice leads to greater understanding of a disease and better adherence to mutually agreed treatments. In the absence of such engagement, it is not surprising to see that a majority of PwH observed in this study have difficulty in translating the information provided to them into effective health protection strategies. Factor concentrate prophylaxis, with its high peak at the moment of infusion and low trough several days later, is not an easy concept to understand or to apply to real life. Inevitably, PwH develop their own models to adapt their care to their lives. At different moments, these models are misaligned, sometimes too cautious and sometimes too risky.

The paper is notable in that it describes some of the challenges of hemophilia – treatment burden, activity restriction and joint problems across four life stages: childhood when the PwH are largely being cared for, the teenage years of testing boundaries, the years of starting a family and building a career, and finally, the later years. It is worth noting that these last two stages were hardly considered in the mid-twentieth century when life expectancy was short. The different experiences across these last stages add more evidence calling for greater personalisation of care. Happily, options exist today where few did in the past.

My parents had little doubt when I was growing up that haemophilia was a serious and often fatal disease. Today, with the many advances, there is a growing perception among some that the problem is solved. Life expectancy is close to normal. As severe haemophilia has been modified through prophylaxis to more closely resemble mild or moderate disease, annual bleeding rates have become low. They are so low, in fact, that many health technology assessments, focused on bleed rates, recommend not funding innovations, citing "lack of evidence of efficacy and cost-effectiveness." As administration of treatment has moved away from clinics into homes, haemophilia treatment centres in many jurisdictions have seen cuts to staffing. Yet, this paper supports emerging data from other studies of patient-reported outcomes, claiming that all is not solved: despite the incredible advances in care, regular bleeding persists, the burden of treatment is important, and this has profound implications at all stages of people's lives.

David Page
St-Michel-de-Bellechasse, Quebec, Canada
September 2020

HOW TO CITE THIS ARTICLE:
Page D. Uncertainty to normality using ethnographic and qualitative research: a personal view. *J Haem Pract* 2020; 7(1): 173-174 *http://doi.org/10.17225/jhp00170*