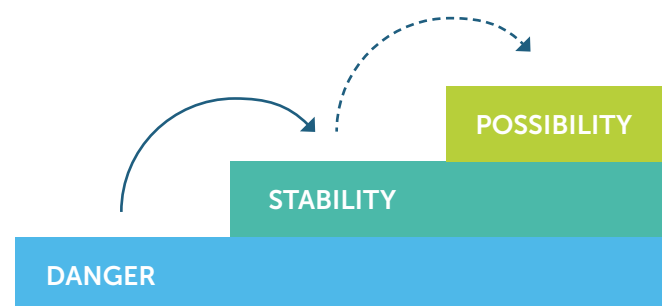


COMMUNITY FOCUS

Treating for stability: an ethnographic study of aspirations and limitations in haemophilia treatment in Europe

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Background: Recent improvements in approaches to treatment have opened a window of opportunity to redefine and expand the goals of treatment in haemophilia. This article explores treatment culture in light of these improvements and its potential impact on the range of possibilities in the lived experience of haemophilia. **Aims:** The aim of this article is to further investigate findings related to how health care professionals (HCPs) approach haemophilia treatment and care, one of the main themes identified in an ethnographic study of the everyday life of people with haemophilia (PwH). This large-scale study investigated PwH's beliefs and experiences related to their condition,



Since the introduction of prophylaxis as a treatment for haemophilia, treatment goals have aimed at moving people with haemophilia (PwH) out of a 'danger zone' and into an experience of stability around their condition. As even more effective treatments become available, it may be time to move beyond this focus on treating for stability, toward more ambitious outcomes and possibilities for PwH.

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their treatment, and their personal ways of managing the condition. **Methods:** The study used ethnographic research methods. Five haemophilia experts helped frame the research design by providing historical and disease area context prior to the initiation of field research. In the field, study researchers collected data through 8–12 hours of participant observation, semi-structured interviews, written exercises, facilitated group dialogues, and on-site observations of the interactions of PwH with friends, family, and HCPs. Study researchers also conducted on-site observation at haemophilia treatment centres (HTCs) and interviewed HCPs. The study employed a multi-tiered grounded theory approach and combined data were analysed using techniques such as inductive and deductive analysis, cross-case analysis, challenge mapping, and

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clustering exercises. This article explores findings related specifically to how HCPs approach haemophilia treatment and care, and is thus focused on a subset of the data from the study. **Results:** Fifty-one PwH in Italy, Germany, Spain, UK, and Ireland were interviewed and followed in their daily lives. Eighteen HCPs from seven HTC were interviewed, and on-site observation was undertaken at six of the HTCs. Most haematologists in the study 'treated for stability', rather than to guide PwH to overcome limitations. 'Treating for stability' here refers to an approach to haemophilia care that focuses on measuring success in terms of annual bleed rate, instilling a focus on mitigating risk, rather than an approach that allows PwH to overcome the limitations they face due to their condition. However, some haematologists had moved beyond treating for stability to instead treat for possibilities, enabling a better quality of life for PwH. **Conclusions:** These results suggest that a culture of 'treating for stability' could be limiting progress in expanding the goals of treatment in haemophilia. Expanded metrics of success, more flexible approaches to treatment, and higher ambitions on behalf of PwH may be needed in treatment and care, in order for PwH to fully benefit from treatment advances and to increase their quality of life.

Keywords: *treatment goals, haemophilia, metrics of success, flexible treatment, annual bleed rate, ethnography*

In Europe and the United States, the relatively recent advent of life-saving treatment has created a greater sense of protection from bleeds for people living with haemophilia. Treatment has advanced from the 1960s onwards and what was once considered a fatal diagnosis can today be safely treated, with the life expectancy of a person with haemophilia almost matching that of a person without haemophilia ^[1]. People with haemophilia (PwH) and their carers can now largely feel safe on a day-to-day basis due to the care offered by current treatment options. Advances in treatment, including the introduction of prophylaxis, have also helped improve quality of life (QoL) among PwH ^[1]. However, studies suggest that there is still room for moving towards possibilities beyond this basic protection ^[2-5]. Recent improvements in approaches to treatment have opened a window of opportunity to redefine and expand the goals of treatment in haemophilia; however, a culture of 'treating for stability,' i.e. a treatment approach focusing on mitigating risk, could be restricting possibilities for PwH.

The aim of this article is to explore perspectives and practices around approaches to haemophilia treatment, based on findings from a large-scale ethnographic study exploring the everyday life of people with haemophilia (PwH), including their beliefs and experiences related to their condition, their treatment, and their personal ways of managing their condition. The overall results of the study are previously published ^[6].

METHODS

The methods of the ethnographic study have been previously described ^[6]. Historical and disease area context was provided prior to the initiation of interviews by five haemophilia experts to help frame the research design. The experts included a specialist nurse at a paediatric haemophilia treatment centre (co-author NM), a practicing psychologist for people with haemophilia (co-author ATO), a physiotherapist, an anthropologist, and a medical psychologist working within the area of haemophilia. This study employed a multi-tiered grounded theory approach and gathered data through semi-structured interviews with PwH and health care professionals (HCPs), as well as on-site observations of the daily lives of PwH, PwH/HCP interactions, and HCP/HCP interactions. Study researchers used audio recording, video, photography and extensive field notes to capture the data, which was analysed in combination using various approaches (e.g. inductive and deductive analysis, challenge mapping, and clustering exercises). Researchers spent one to two days with individual PwH and spent an average of four hours at the haemophilia treatment centres (HTCs) visited. The in-depth nature of the interviews and observations allowed the study researchers to uncover the underlying needs and challenges faced by PwH, and the drivers behind current treatment approaches, as well as unearthing 'softer' experiential metrics, such as the aspirations, fears, doubts, and attitudinal shifts currently dominating the discourse within the European haemophilia community. All statistics in this article are based on analysis of self-reported participant information.

Recruitment

PwH were recruited for this study in Italy, Germany, Spain, UK, and Ireland, through patient organisations in each country. The recruitment criteria aimed for a representative sample of PwH, screening candidates by haemophilia type, disease severity, treatment regimen, presence of inhibitors, and age range (under 12, 13–18, 19–49, 50+). HCPs were recruited for a mix of experience levels as well as representation of larger and smaller clinics.

Ethical Considerations

PwH and HCPs participating in the study signed a GDPR-compliant consent form, which informed them of the terms of participation and the way their personal data would be managed. The study was conducted following the ethical standards outlined by the ICC/ESOMAR International Code on Market and Social Research [7], which sets out global standards for self-regulation for researchers and data analysts, as well as relevant national standards for participating countries [8-11].

Given the highly personal nature of the data collected in this study, participant privacy and anonymity were of high concern. As guaranteed in the GDPR-compliant consent form, personal data was handled with the utmost care. In order to identify the different participants while preserving confidentiality, each participant in the study was assigned a unique number. Quotes and cases are labelled with the participant's age range (e.g. teenager). All potentially identifying information about participants has been omitted.

RESULTS

The study included 18 HCPs in multidisciplinary functions (haematologists, nurses, physiotherapists, dentists) as well as administrative staff from seven HTC of a range of sizes (from ~30 to ~500 patients) and patient groups (adults, paediatrics, both) across the five countries. Additionally, the research rests on the described and observed experiences with HCPs and the healthcare system of 51 PwH and their families and friends.

One of the main findings of the study concerned approaches to treatment, here grouped into three sub-themes: treating for stability, the PwH-HCP relationship, and the potential of treating for possibility.

1. Treating for stability

The study researchers observed that the majority of HCPs generally act conservatively in their treatment practices. For most HCPs in the study, the most important metrics of success involve lowering annual bleed rates and avoiding inhibitors, informing their attitudes towards treatment options, changes in treatments, and their communication around protection levels and physical activity. From this perspective of success, newer medicines were generally described as presenting risks. Most HCPs in the study were also reluctant to push for new treatment regimens as they considered all factor-based products to be equally effective in terms of the main goals of lowering annual bleed rates and avoiding inhibitors. In the words of one haematologist:

"I don't see a big difference between plasma, recombinant, and EHL treatment options."

Similarly, most HCPs in the study acted cautiously towards non-factor-based treatments, emphasising that there are important unknowns around side-effects and drug-to-drug interactions. When PwH experienced problems on their current treatment, many HCPs described checking compliance in terms of following the treatment regimen, and reviewing the types of activities PwH engaged in, then testing a new frequency and dosage plan, before considering trying another product.

PwH-reported experiences also indicated that HCPs are generally reluctant to try new products; however, many HCPs were making clinical adjustments to existing treatment plans. Where 42% (n=14/32) of PwH without inhibitors (excluding the Irish PwH, who had changed treatment due to the national tender system) had changed dosage or frequency in the last three years, very few PwH (6%; n=3/51) in the total sample had exchanged their treatment product. Many PwH who described experiencing bleeds despite treatment adherence were primarily asked to be more careful. An example of this pattern seen across clinical settings was observed during a regular examination of a young PwH who was experiencing bleeds. In this case, the haematologists in the clinic explained to the family that the first action should be to work with a traumatologist. Although the parents explicitly asked about other medications, the doctors avoided this in their conversation, putting the focus on other personal and physical changes the PwH could make.

HCPs generally geared their approaches to treatment towards preventing bleeds. Rather than enabling the pursuit of more activities, the treatment regimen of most PwH in the study aimed to achieve a level of protection at which no spontaneous bleeds occur, but also required that the PwH limit themselves to avoid bleeds. One HCP described his clinic's approach as follows:

"We generally aim for three per cent trough levels because above that they will have no spontaneous bleeds."

In the PwH-reported data, the authors found that 74% (n=14/19) of all PwH who had not experienced any bleeds within the last year refrained from physical activities to avoid bleeds.

2. PwH and HCP relationship

The majority of PwH-HCP relationships observed in the study were characterised by a closeness and trust, well-cultivated over many years. The vast majority of PwH put a lot of weight on the words of their HCPs and rely on them as their primary source of information. In the interviews with PwH, 78% (n=39/50) described a deep trust for their HCP. While the nature of this relationship allowed PwH to feel supported and cared for, it also seemed to result in PwH having a relatively uncritical perspective on their HCP's advice and adopting the HCP's perspective on approaches to treatment, which, as discussed above, is focused on prevention and activity restriction.

The PwH's trust in their primary HCPs, whether haematologists or nurses, came across in many ways, with some even describing them as 'extended family'. For example, when a teenage participant was homesick during summer camp, the nurse at the clinic comforted him and was a reassuring presence that convinced him to stay. Additionally, this nurse was the only person who could help him relax enough for his injection. Many PwH in the study were willing to travel great distances (up to 14 hours' travel) in order to preserve trusted relationships with the staff in their preferred HTC. One boy's (child) parents spent 350 euros per month traveling to see their preferred HCP. It took one man (40s) 2.5 hours to get to his preferred clinic, and he makes the trip every three months on top of his 70-hour working week. Another man (30s) also travelled a long distance to attend a better clinic:

"The improvement in care is worth the extra miles."

The majority of HCPs also strived to foster these close relationships, even in the midst of a hectic day, showing deep care and commitment to their patients' wellbeing, with one haematologist stating:

"Some patients I've known since they were babies, so it's inevitable that we develop a relationship."

PwH emphasised two factors when explaining what made them trust HCPs. Firstly, medical haemophilia expertise is essential. Almost all PwH in the study had alarming stories to tell about encounters with general practitioners who did not have specialised knowledge about their rare condition, which emphasised the importance of being able to see and consult

with someone who has a deep understanding of haemophilia. Secondly, PwH and carers emphasised the importance of a caring bedside manner, where HCPs take an interest in the details of the patients' lives beyond their condition and go the extra mile to provide good care, e.g. by providing 24-hour phone support and texting with immediate responses. Many PwH stated that they prefer not to be treated as a case, but rather to be asked questions that demonstrate an interest in them as people. For one young man (30s), a haematologist's lack of ability to show interest and listen made him switch to a different HTC, even though this was less conveniently located for him.

All PwH in the study considered HCPs the primary source of learning about haemophilia. Once this trusted relationship was established, PwH rarely questioned their HCPs' advice, as expressed by one young participant (20s):

"The doctors tell me what to do. I trust them."

Furthermore, participants often blamed their own actions or the failure of their own bodies when they experienced a bleed, rather than asking for improved protection. One young man (20s) who is plagued by ankle and back bleeds blamed his own activity levels for the damage, saying that he overstepped the boundaries:

"My ankle is like an elderly person's ankle. I pushed it too much. But there was less awareness when I was younger."

Another young man (20s), who has issues with injecting, tended to blame his own body rather than the equipment or his training:

"My body reacts differently every time I inject. The veins move and change in size. Sometimes it feels like [the treatment] burns and other days it doesn't."

Others, including one older participant (50+), held back from activities and personal goals like travel in order to avoid bleeds, but never thought to ask for a treatment regimen that would allow these activities.

3. The potential of treating for possibility

Our research indicated that treatment practices in Ireland differ from the other countries in the study, with researchers observing there a treatment culture more

Table 1: Comparison of treatment regimen and outcomes reported by Irish and non-Irish participants

	IRISH RESPONDENTS	NON-IRISH RESPONDENTS
PwH on prophylaxis treatment and fully adherent	12 out of 12 100%	28 out of 39 75%
PwH who adapt their treatment regimen to physical activities	8 out of 12 66.6%	12 out of 39 30.8%
Non-inhibitor PwH who had a bleed in the last year*	3 out of 10 30%	23 out of 33 69.7%
PwH (or carers) who are generally adherent to prophylaxis treatment who worry about not being adequately protected**	3 out of 12 25%	14 out of 33 42.4%

* 2 Irish PwH and 6 non-Irish PwH had current inhibitors

** 6 PwH (or carers) in the non-Irish population reported not being generally adherent

aimed at enabling possibility. Based on the insights obtained from the Irish HCP as well interviews with PwH, it appears the majority of HCPs in Ireland take a more flexible approach to treatment. Examples from Ireland of this more flexible approach to protection included a more 'situational' use of factor that expands the number of activities PwH can take part in. One nurse told researchers:

"We tell them to tailor their prophylaxis to their lifestyle. If it's gonna be a rough Saturday night, take an extra dose. [...] Troughs are at least five per cent, so at sports practice, if they fall and bang themselves, they should be treated exactly like any other boy who falls and bangs themselves."

In another example of this flexibility to patients' lives, doses of treatment are greatly increased by some HCPs (paediatric and adult) during hurling season (a popular Irish sport) in order to allow PwH to partake in the contact-heavy sport.

The HCPs in Ireland reported aiming for higher trough levels across the board (3% for adults and 5% for children in Ireland, compared to the reported 1–3% in other European countries), with the newest treatments made accessible by a national tender process, with the goal of reducing spontaneous bleeds and promoting better long-term joint health. Based on the sample in this study, it appears that PwH are generally provided with more information and given clearer guidance in Ireland. The Irish Haemophilia Society (IHS) supplies pamphlets, over-the-phone support, and home visits upon request. Nurses from one of the Irish clinics visit schools to give teachers information sessions on haemophilia and to ensure that they are confident having a student with the condition.

In this study, PwH in Ireland lived more active lifestyles relative to PwH in similar age groups in the other countries included. Some of the adult and teenage participants had at some point received tailored treatment regimens to allow them to participate in high impact activities, such as Gaelic football and/or hurling, which were described as important parts of Irish national identity. One nurse explained the why a more pragmatic and flexible approach to patients' lives has been adopted by her HTC:

"We used to tell people not to play GAA [Gaelic Athletic Association sports, e.g. hurling] or football, but they will play anyway."

All adult Irish PwH in the data mentioned hurling and Gaelic football, and most had participated in them at some point in their lives. Additionally, PwH in Ireland expressed less uncertainty about dealing with haemophilia than PwH in other countries and fewer PwH in Ireland expressed worry about not being sufficiently protected.

The respondent-reported data from the study, albeit from small numbers, also supports that PwH in Ireland receive more effective care than in other countries in the study, suggesting that they may be more adherent, receive more adaptive treatment regimens, and experience fewer annual bleeds (Table 1).

DISCUSSION

For decades, approaches to haemophilia treatment have been about ensuring survival and stability, by moving PwH out of the untreated 'danger zone' towards day-to-day stability. The insights from this study suggest that the current standard of treatment is still focused on establishing basic protection rather than enabling personal goals. Most HCPs in the study used metrics of success that revolve around preventing

bleeds and were wary of or did not see advantages in changing treatment. While this 'baseline treatment' allows PwH to live longer than in the past, it does not fully enable them to pursue ordinary personal goals, for example playing football when they choose to, taking part in the same activities as their friends at school, or staying mobile in daily activities without chronic pain.

Analysis of the treatment practices of HCPs operating in the different health care systems covered by the study indicates that they generally make treatment choices and give advice to patients with the aim of moving PwH out of the danger zone and towards an experience of stability around their condition. The implication of this approach is that if HCPs are treating primarily to avoid spontaneous bleeds – so, in theory, all bleeds that occur happen due to accidents or activities – then patients are, to some extent, considered responsible for their bleeds. When PwH experienced issues, they were generally encouraged to decrease their activity levels to stay safe and avoid bleeds, rather than changing treatment regimen ^[12-15]. In this way the overarching goal of treating for stability limits the possibilities that treatment can offer today.

Existing research suggests that the closeness of the PwH-HCP relationship, which was also found in this study, has an overall positive impact on patient outcomes. A close relationship with HCPs seems to be a key predictor of high adherence levels ^[16,17] and other positive effects, such as improved treatment outcomes ^[23]. However, a potential downside based on the findings of this study could be that PwH are reluctant to ask for more possibilities because they have instilled so much trust in their HCPs and largely adopt the HCPs' stability-focused treatment goals. While shifting the burden of finding credible information and decision-making to a trusted practitioner can free PwH and carers from emotional stress, it also has the possible implication that they rarely ask for more protection. PwH often appeared satisfied with their current treatment and with continuing on it, preferring not to challenge the status quo, despite having to hold back from certain activities and personal goals. In fact, when bleeds happened, many PwH did not question their treatment regimen, but rather drew attention to their own bodily failings or rash behaviour. By adopting this stability focus, rather than asking for changes in their treatment regimens that would allow them to expand their own possibilities in life, PwH's lives, in most of the countries in this study, seemed more constrained than advances in treatment options would otherwise allow. One key exception was observed in

Ireland, where the stability and security approach to treatment has shifted to an approach to treatment that could expand possibilities in the lives of PwH.

Ireland presents a case study in treating more for possibility than stability. Here, haemophilia management has moved further beyond treating to 'stability' than the other countries included in the study by focusing more on personalised care and outcomes. This may in part have been enabled by a different health care system. In this study, PwH in Ireland generally received treatment regimens providing more protection and a higher degree of flexibility in their care compared to in other countries. As a result, they appeared to be more active and dealing with less uncertainty around protection. PwH in Ireland were even able to play Gaelic Athletic Association (GAA) sports, and past research suggests that not being able to take part in sports with a strong association to the national identity could potentially lead to social challenges ^[15]. The approach to treatment in Ireland also seems to allow a more active life at an older age for PwH. Existing research indicates that higher trough levels, better adherence, and increased individualisation leads to superior outcomes for PwH in terms of both bleeds and QoL ^[2], and respondent data from our study supports these claims.

This study supports previous evidence ^[2] that PwH should be encouraged to reach for the new possibilities that are enabled by advances in treatment. In order to fully benefit from recent treatment advances and enable less restricted lives, more treatment flexibility and higher ambitions on behalf of PwH are needed ^[3]. Ultimately, this means an expansion of the metrics of success to better account for the lived experience of haemophilia beyond annual bleed rates, which several modern treatments options can deliver. However, more research is needed in this area, as potential additional relevant outcome measures such as health-related quality of life (HRQoL), pain scores, and psychosocial issues are currently lacking agreed upon methodologies, indicators and research questions ^[19-22].

Limitations of the study

The findings described above are representative of patterns observed across several European countries and conclusions about differences between countries are somewhat limited by the relatively low number of patients and HCPs per country. Furthermore, the impact of different health care systems and type of treatment centre (i.e. HTC vs CCCs) on the treatment approaches adopted by HCPs should be further investigated. It is also important to note the potential of self-selection bias in the volunteer-based recruitment approach.

CONCLUSION

Our study suggests that PwH develop a close relationship with and instil a lot of trust in their main HCPs, who often go to great lengths to nurture this relationship and help their patients. While the close relationship between PwH and HCPs has a range of very positive effects, findings of this study indicate that it may also contribute to PwH adopting a stability discourse, holding them back from asking for more protection through treatment. However, the study also suggests that improved treatment outcomes, allowing for greater possibilities for PwH, would be possible if more ambitious metrics of success were adopted. While the treatment paradigm in most European countries is centred around protecting the PwH from bleeds and giving them a sense of stability, there are examples in our study where some PwH are more encouraged to live a life less restricted by their haemophilia. With improved treatment options, care, and guidance, it seems that this aspiration is possible for PwH.

DISCLOSURES

This study was carried out by ReD Associates with funding from Sobi. Sobi and Sanofi reviewed the article. The authors had full editorial control of the article and provided their final approval of all content. TH, MBK, YG, AML, and ABL are employees of ReD Associates. ATO is a researcher at the University of Murcia. NM is a researcher and nurse at Alder Hey Children's Hospital. JS is an employee of Sobi.

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Informed consent has been obtained from the participants in the study reported in this paper.

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