

ORIGINAL ARTICLE

Clinical haemophilia

A qualitative study on the experiences of haemophilia carriers before, during and after pregnancy

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Abstract

Introduction: Haemophilia carriers (HCs) face considerable haemostatic and psychological challenges during reproduction.

Aim: To explore the perspectives of HC's on healthcare in the current standard of haemophilia treatment during all reproductive phases: preconception, pregnancy, childbirth and the postpartum period. In addition, we examined the psychological impact of haemophilia during these phases.

Material and methods: Focus group discussions (FGDs) and semi-structured interviews were conducted with HC's in January/February 2020 until data saturation was reached. All sessions were recorded, transcribed verbatim and analysed by two independent researchers through thematic content analysis using MAXQDA® software. The results were then discussed within the research team until consensus was reached. The constructed themes were shared with and reviewed by the HC's.

Results: Fifteen HC's were included in three FGDs and four interviews. Five central themes were constructed: (1) communication by healthcare professionals, (2) lack of knowledge, (3) feeling insecure, (4) autonomy and (5) family experiences with haemophilia. Desired improvements in care mainly concerned counselling during preconception and pregnancy. This included timely access to comprehensive information during each consecutive phase, acceptance of HC's' choices by healthcare providers and healthcare tailored to the HC's family experience with haemophilia.

Conclusions: In recent years, haemophilia treatment has seen major advances, which could impact general and reproductive care for HC's. HC's indicated that reproductive care would benefit from a more personal and informative approach. Healthcare professionals could use these insights to adapt their consultations to meet the needs of these women when they are preparing for having children.

KEYWORDS

haemophilia, healthcare, postpartum period, preconception, pregnancy, qualitative study

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1 | INTRODUCTION

Haemophilia carriers (HCs) face considerable haemostatic and psychological challenges during their lives.¹⁻³ The high probability of transmitting haemophilia to offspring complicates reproductive decision-making and introduces additional choices, including preimplantation genetic diagnosis, prenatal diagnostics and termination of pregnancy.⁴

Four reproductive phases can be distinguished: preconception, pregnancy, delivery, and the postpartum period. Preconception allows for preparation for the upcoming pregnancy and family life, with preimplantation genetic diagnosis being one of the first major choices women and their partners have. After conception, challenging dilemmas regarding prenatal diagnostics emerge for HCs.^{5,6} Prenatal diagnostics, while potentially having a medical and psychosocial impact, contributes to peripartum management and enables parents to psychologically prepare for having an affected child.^{7,8} Previous studies on pregnancy outcomes have reported a higher prevalence (30-57%) of postpartum haemorrhage in HCs compared to the healthy population (19%).⁹ Postpartum haemorrhage is associated with serious maternal morbidity and traumatic impact.^{10,11} During all reproductive phases feelings of guilt due to having an affected child as well as the threat of bleeding may cause a physical and emotional burden.¹²

Despite a considerable number of qualitative reports on the experiences of people with haemophilia and their caregivers, few studies have examined the psychosocial issues of HCs.¹³ A systematic review on HCs experiences before, during and after pregnancy showed that the few available studies have focused on prenatal diagnostics and the impact of HC status on reproductive choices.¹⁴⁻¹⁹ This information is partially outdated as the prospects of living with haemophilia still change favourably due to introduction of non-replacement therapy and gene therapy.²⁰⁻²³ However, people with haemophilia still experience bleeds and joint disease to some extent, which can affect their day-to-day life.^{24,25}

Up-to-date knowledge on all reproductive phases in the current era of haemophilia treatment in addition to current research on the psychosocial impact of haemophilia is essential to ensure quality of haemophilia reproductive healthcare. We aimed to explore HCs perspective during preconception, pregnancy, delivery, and the postpartum period to assist clinicians in meeting the needs of HCs.

2 | METHODS

2.1 | Study design and research team

This study was conducted in the Netherlands, where HC care is provided in Comprehensive Care Centres (CCCs). Face-to-face focus group discussions (FGDs) and semi-structured interviews by telephone were conducted to assess the impact of haemophilia on each reproductive phase. FGDs provided the opportunity for interaction and discussion between HCs, while the semi-structured interviews enabled result validation and confirmation of data saturation. Two CCCs were

involved: University Medical Centre Utrecht (UMCU) and Amsterdam University Medical Centre (AUMC) of which both ethical committees confirmed that ethical approval was not required (reference numbers 19-626(1-10-2019)/19-434(14-11-2019)). The research team included medical doctors (MP, KG, KF, KB), psychologists (LT, LH) (all familiar with haemophilia), and patient representatives from the Dutch haemophilia patient organization (NVHP: LP, MHED). The consolidated criteria for reporting qualitative research (COREQ) were followed to ensure comprehensive reporting.²⁶

2.2 | Participant selection

HCs from the UMCU, AUMC and NVHP were invited if they met the following inclusion criteria: (1) obligate/known HC, without concomitant coagulation disorders; (2) ≥ 18 years of age; (3) ability to provide informed consent and proficiency in the Dutch language; and (4) experienced childbirth within 5 years before study commencement. HCs were invited through pragmatic inclusion, after which the need for a selective approach was determined. A broad representation of HCs was desired: (1) carriers of mild/moderate/severe haemophilia; (2) being patients from different CCCs; (3) experienced childbirth inside/outside CCCs; (4) experienced different situations during reproductive phases (i.e. choosing/opting out of prenatal diagnostics, affected/unaffected children); and (5) NVHP members/non-members. Participants of the interviews were recruited after the last FGDs and consisted of women for who the timing or location of the FGD was inconvenient. Informed consent was provided by all participants.

2.3 | Data collection and analysis

Sociodemographic data were collected through questionnaires before the FGD and from the medical records. FGDs were held at the UMCU and were conducted by at least one facilitator and one observer (MP, LT or LH), whereas the interviews were conducted by one interviewer (MP or LT). These female researchers (MP, LT, LH) had been trained in qualitative research and did not have a current healthcare provider relationship with the participants. FGDs were conducted according to qualitative research guidelines²⁷ and consisted of 120-min face-to-face meetings with 3-5 HCs during which each reproductive phase was discussed in chronological order. All FGDs were audiotaped and transcribed verbatim, anonymizing the participants. Field notes were taken to assess non-verbal communication and to evaluate the FGD. A non-judgmental atmosphere was promoted by emphasizing the need to understand the women's experiences and the importance of keeping personal information confidential. The topic list was based on identified knowledge gaps by a literature review,¹⁹ questionnaires from the on-going observational study (PRIDES study: Dutch trial registry number NL6770) and the clinical experience of the haemophilia treatment team (Supplement 1). FGDs were continued until data saturation was reached i.e. when the last FGD showed no new insights.

TABLE 1 Socio-demographic characteristics of the participants (n = 15 haemophilia carriers)

		N = 15	N = 15
Maternal	Age in years (median, range)	33 (27-37)	Bleeding disorder - Haemophilia A carrier
	Clotting factor level ^a - < 40%	3 (20%)	Bleeding tendency ^a - Yes
	Severity of haemophilia in family - Severe	6 (40%)	DNA analysis carrier - No
	- Moderate	4 (27%)	- Yes, before the first pregnancy
	- Mild	5 (33%)	- Yes, during pregnancy
	Member of Dutch patient society - Yes	4 (27%)	Comprehensive Care Centre - University Medical Centre Utrecht
	Ethnicity - Dutch	15 (100%)	Education level - High school
Prenatal	Parity (1/2/3)	6/7/2	Gender known during pregnancy - Yes
	Prenatal diagnostics ^b - Yes	7 (78%)	Termination of pregnancy (affected son) - Yes
Delivery	Delivery location ^c - CCC	6 (40%)	Peripartum prophylactic treatment - Yes
	- CCC & non-CCC	5 (33%)	
	- Non-CCC	2 (13%)	
	- Home	2 (13%)	
	Postpartum haemorrhage \geq 500 cc - Yes	4 (40%)	
Neonatal	Children - Boys (severe -/ moderate -/ mild haemophilia / not affected)	0/3/2/4	
	- At least 1 girl	14 (93%)	

Legend: a. Self-reported, outside of pregnancy, b. Out of nine women who were pregnant with a boy, c. CCC, Comprehensive Care Center, location(s) of each delivery per woman.

All data from FGDs/interviews was anonymized prior to analysis. Sociodemographic data were summarized with descriptive statistics using IBM SPSS Statistics Version 25. MAXDQA version 10 was used for the qualitative analyses. Two researchers (MP, LT) conducted the coding: (1) independent open coding; linking relevant fragments to an appropriate code; (2) adding new codes to cover the derived themes; (3) axial coding; categorizing codes into themes and discussing those within the research team (MP, LT, LH, KG).²⁸ Main and subthemes were determined during the FGDS/interviews i.e. we asked which healthcare experiences are most impactful and which healthcare aspects mostly require change. Pre-final themes were shared once by email with participants and NVHP members (LP, MHED) to allow for feedback. The results were examined again in depth by the research team to finalize the themes. Original, anonymized data is available upon reasonable request.

The narrative synthesis of the themes will be presented as an overview of the overall themes and subsequently for each reproductive phase in chronological order from preconception up to the postpartum period. Representative quotes are provided to support the themes.

3 | RESULTS

3.1 | Participant selection and characteristics

Of the 42 women approached, 15 HCs agreed to participate; 11 HCs participated in three FGDs and four HCs were interviewed. These women reflected a variety of experiences and choices made regarding prenatal diagnostics, delivery locations and whether their child was affected (Table 1). No selective invitations were made after this initial pragmatic inclusion as the study population met the diverse characteristics we desired beforehand.

3.2 | Overall themes

While HCs generally reported being content with the current standard of haemophilia care, many HCs expressed their wish to help other women. Five central themes were derived throughout each reproductive phase: (1) communication by clinicians, (2) lack of knowledge, (3) feeling insecure, (4) autonomy, and (5) family experiences (Table 2).

TABLE 2 Overview of the main results

		Phase			
		Preconception	Pregnancy	Childbirth	Postpartum
Themes	Communication by healthcare professionals	<ul style="list-style-type: none"> Timely, comprehensive information on carriership and consequences 	<ul style="list-style-type: none"> Timely, comprehensive information on different scenarios Communication adjusted to personal experiences Efficient/coordinated care Information exchange between involved healthcare professionals 	<ul style="list-style-type: none"> Delivery plan and discharge information exchange among involved healthcare professionals and HCs^a Clear instructions at discharge 	<ul style="list-style-type: none"> Information transfer to midwife/ maternity care Written take-home information for the mother and baby
	Lack of knowledge	<ul style="list-style-type: none"> Healthcare professionals outside Comprehensive Care Centres 	<ul style="list-style-type: none"> Obstetrician/gynaecologist, midwife 	<ul style="list-style-type: none"> Obstetrician/gynaecologist, midwife 	<ul style="list-style-type: none"> Maternity care
	Feeling insecure	<ul style="list-style-type: none"> Upcoming choices during consecutive phases 	<ul style="list-style-type: none"> Potentially affected child Safer in Comprehensive Care Centres Deciding on prenatal diagnostics 	<ul style="list-style-type: none"> Distance to Comprehensive Care Centres Lack of healthcare professionals' experience with HCs Mode of delivery Maternal and neonatal (male/female) bleeding risk 	<ul style="list-style-type: none"> Testing baby (male/female) Maternal and neonatal (male/female) bleeding risk
	Autonomy	<ul style="list-style-type: none"> Timing of carrier diagnosis Timing and method of preconception counselling 	<ul style="list-style-type: none"> Gender assessment, prenatal diagnostics, termination of pregnancy Choice of midwife versus gynaecologist Intensity and timing of outpatient clinic visits 	<ul style="list-style-type: none"> Choice of midwife versus gynaecologist Location of delivery 	<ul style="list-style-type: none"> Timing testing baby Timing counselling on future pregnancies
	Family experiences	<ul style="list-style-type: none"> Severity of haemophilia and coping of family determines the attitude towards pregnancy 	<ul style="list-style-type: none"> Severity of haemophilia and family attitudes influence prenatal diagnostics and decisions regarding termination of pregnancy 	<ul style="list-style-type: none"> Home delivery possible 	<ul style="list-style-type: none"> Adjust frequency of clinic visits to severity of haemophilia and coping of family

Legend: a. HCs, haemophilia carriers.

Most HCs chose preconception and pregnancy to be in greatest need of improvement. Improvements could focus on providing timely, comprehensive information on each consecutive phase. An important finding was that HCs needed to feel that their personal choices were being accepted by clinicians; for some HCs, even the knowledge that they had, in fact, a choice. During each reproductive phase, some HCs mentioned that care provision felt impersonal. Moreover, healthcare provision should be tailored to each individual's family experience with haemophilia. There was a great diversity in the overall feeling HCs expressed when talking about haemophilia and carriership – ranging from haemophilia not playing any role up to having a significant emotional impact on daily family life.

"I don't feel like I am any different than the average pregnant woman who has to deliver. So, decide for yourself what feels comfortable during childbirth, what you want." FGD2, Respondent (R)4

"I told it [being pregnant with a boy] to my mother and I could hear her thinking "oh no, a boy". [...] It still gets to me, thinking how can you still feel that; that really shocked me. (Cries) Yes, especially for her, exactly that, the grief, and how do I make sure to remain true to myself in this." FGD1, R3

3.2.1 | Preconception

Preparation varied widely among HCs: some HCs were uncertain about care pathways, carriership testing and where/how to receive preconception care (Supplement 2). HCs were surprised about the limited knowledge general practitioners and midwives have about haemophilia, impairing proper preparation for pregnancy. Some HCs felt as if no choices were available; such as preimplantation genetic diagnosis being the only option. Several HCs also reported that within

their own families there was little knowledge about haemophilia carriership.

HCs felt that their family was the greatest influence on their views about the burden of their carriership and reproductive choices. Some women had experienced severe haemophilia with extensive consequences within their families and felt anxious when preparing to have children themselves. They did not want their child to face the same challenges as their affected male family members and were more likely to discuss options including preimplantation genetic diagnosis (Supplement 3). A positive family experience, where affected male members experienced few consequences, led HCs to feel that haemophilia did not impact their reproductive choices. HCs believed that the information provided regarding consecutive phases (i.e., preimplantation genetic diagnosis, prenatal diagnostics and termination of pregnancy) should be comprehensive, though always carefully patient-tailored. In addition to medical consultations, several women were also guided by a social worker who brought them into contact with other families. This allowed HCs and their partners to gain new insights, because current standards of treatment might mean that the experience of families today might nowadays be dramatically different.

"I have to say because of that, the experience around haemophilia for us was quite tough, because my uncle was very seriously affected, including lot of damage to the joints. Well, fused knees, ankles etcetera." FGD3, R1

"[...] for us it has always been an issue, that a child was not here because my parent made the conscious decision at the time not to do it. And that I still sometimes think the grief, for them it is mostly knowing that if the knowledge at the time had been like it is now, then they would never have made that decision." FGD3, R2

During preconception, HCs mentioned being able to access timely, comprehensive information on consecutive reproductive phases was most important. This is especially relevant for couples considering preimplantation genetic diagnosis, a potentially time-consuming process. Most HCs did not identify a specific age for an outpatient clinic visit, but agreed on the need to educate families and clinicians regarding access to information and preconception outpatient clinics. This allows HCs and their partners to initiate preconception care when they feel ready. For some individuals, this occurs during adolescence; for others when starting a family is near. HCs did agree that once they are notified about their carriership and began seeking additional information, sufficient information was available. Knowing that the CCC can be contacted at any time was helpful for some HCs, whereas others would have preferred to be informed earlier.

"I can still clearly remember that my clotting levels were tested when I was a child, and that I was told that if I ever, in the future, when you are really old, I think I was ten years old, if you are ready to have children, then come visit me, then I will explain things. That has stayed with me." FGD2, R4

"It was more like, if you are pregnant, then just call the clinic. So that's what I did, while I might have preferred to know, like, if you become pregnant then these are the options." FGD3, R2

3.2.2 | Pregnancy

Even though HCs felt that Dutch healthcare for pregnant HCs is generally well structured, several improvements can be made (Supplement 4). During pregnancy, family experience and attitudes towards haemophilia affected the women's views about gender assessment, prenatal diagnostics and termination of pregnancy. Emotions of HCs varied greatly, from feeling 'like any other woman' to experiencing anxiety and feeling the burden of decision-making. Women mentioned that it was an eye-opening experience to hear each other's stories. HCs realized that, for example, prenatal diagnostics comes with certain medical benefits, but many HCs did not feel that this was, in fact, a choice. The conclusion of HCs was that each decision is acceptable and understandable and should always be respected. These decisions, together with general care plans, should be carefully noted and shared with involved clinicians. This prevents HCs from having to repeat their choices and to have a clear pregnancy/delivery care plan for all involved parties.

"I felt like I had to defend myself that I did not want to undergo that test. And actually, during each conversation with a doctor I was told, you haven't done that test right? Why not? So during each consultation I had to say that again." FGD1, R3

HCs discussed how they pregnancy care can be optimized. Joint gynaecologic and hematologic clinics provided trust, and women felt comfortable that all clinicians were familiar with haemophilia. HCs wanted to receive timely, clear information on where care can be received (midwife/general hospital/CCC), which choices can be made (e.g., prenatal diagnostics, delivery location) and possible scenarios. Each of these options comes with certain benefits or drawbacks, but neutral advice is valued. This allows HCs to prepare for upcoming events by selecting their own optimal care route. Choices of prenatal diagnostics and termination of pregnancy were predominantly based on weighing maternal/foetal risks versus the obstetric or emotional benefits (Supplement 3). For some women, haemophilia was not severe enough to opt for preimplantation genetic diagnosis or termination of pregnancy, whereas others expressed that an unaffected foetus felt like the only acceptable option, thus preimplantation genetic diagnosis was chosen. Women felt safe when at a CCC, but midwives provided more personal care. While receiving advice from clinicians, HCs also expressed the need for autonomy in deciding to combine midwifery and hospital care and when to switch to secondary care.

"Because you can also get somebody who indeed only mentions all the risks and everything, but she just... You have to

watch out, but you should also just do your thing, that felt really good to me." FGD3, R2

"So my whole pregnancy from week 1 to 38 was guided by a gynaecologist. Each visit, it looked great. And after 5 minutes I could leave again. Each time a really nice ultrasound was done, that was really nice. [...] You know, with a midwife you can have a nice general chat. With a gynaecologist, however nice the man was, there was no room for a nice chat." FGD1, R2

3.2.3 | Delivery and hospital admission

Many HCs felt their delivery being similar to any other woman, whereas some HCs felt insecure about both their own and their child's bleeding risk (Supplement 5). When reflecting on their delivery, women wanted to follow their personal needs and mentioned that their main concern was their child's health.

"I didn't notice any different postnatal care than you would normally get. I believe that wouldn't have been necessary anyway." FGD3, R1

"Well that made me, well, that I really went through the booklet. I thought, if they [clinicians] don't know it [about delivery in HCs], then I should know something about it." FGD3, R4

Well before delivery started, a main concern of HCs was the distance to the CCC. Several women were nervous about having to travel far during labour and feared delivering before reaching their destination. Some preferred to deliver in a hospital closer by, even though that might not be a CCC. HCs also suggested the potential role for midwives in assessment at home to assist in deciding when to go to the hospital.

"In case I have to deliver here [at the CCC], then I can imagine that I deliver that quickly that I deliver next to the highway, I don't really feel like that. So I am quite nervous about, well, how will that go?" FGD1, R1

HCs were not only concerned about the bleeding risk for affected boys, but also the bleeding risk for girls who are potential carriers. Additional information on the delivery and postpartum period for mother and neonates (male/female) on paper and scheduled outpatient clinic visits would be valued.

"In hindsight I thought, because for the last part they used a vacuum and did a serious episiotomy, I thought, even though it is a girl, I have low clotting factor levels, she has maybe as well, have they fully determined the vacuum to be possible? I am not sure myself." FGD3, R1

"What if bleed afterwards, I really... I was really aware of that. I specifically mentioned to the midwife that she had to pay attention to it [excessive bleeding], because I would be needing medication." FGD2, R3

Furthermore, a detailed delivery plan including prophylaxis and anaesthetic pain management was to be shared between all involved clinicians (midwife, gynaecologist, haematologist and anaesthetist). Without this, HCs felt that they had to watch over themselves. Clinicians seemed unsure and differed in opinion about neuraxial techniques, the side effects of prophylaxis (i.e., fluid restriction during DDAVP) and duration of hospital stay. A visit from the haematologist or haemophilia nurse to the postnatal ward contributed to feeling well cared for personally.

"So the next day I was like a swollen doll, full with fluid. [...] But they don't know that [Fluid restriction after DDAVP]." FGD2, R4

"I had people from the haemophilia department on my bedside twice. I received the information. [...] That is like a warm blanket, I am really pleased about that." FGD2, R3

3.2.4 | Postpartum period

The postpartum period was mostly characterized by testing male neonates when no prenatal diagnostics had been done and by often limited contact with the haematology department (Supplement 6). HCs felt that testing of neonates was stressful; especially collecting blood, but HCs preferred to complete it as early as possible. Women who had a daughter were unsure whether their daughters were carriers and their daughter's bleeding risk. HCs preferred testing of clotting factor levels of female neonates as early as possible as well and genetic testing earlier than the current Dutch practice, which is to test from the age of 16 years onwards. A telephone call from a haematologist, preferably someone who is updated on the delivery, is welcomed: feels personal and provides the opportunity to ask questions.

"I have a daughter, you don't have to come. Even though she might still have the same as I do, in terms of clotting factor level. But there is no regular clinic visit for that. Actually, a bit strange." FGD2, R4

"Yes, after a few days I received a call on how it all had been. And whether I had any complaints. I appreciated that." FGD1, R1

In general, HCs were not worried about their own bleeding risk. However, they valued receiving clear information from the midwife on possible scenarios. HCs required information on criteria for excessive maternal bleeding, when to call the hospital and scheduling outpatient

clinic visits (such as at 6 weeks for maternal/neonatal care). The ideal timing for information provision on future pregnancies should be chosen by the HCs, generally not too soon after delivery.

"Maybe a bit of information on the postpartum period, where you, yes, maybe on paper or something, what you have to be aware of. Because you do forget everything when somebody tells you." TC1

"Maybe they could have asked beforehand. I don't know whether we are thinking of a second [child], whether we would like information now, or whether that is something for later." FGD1, R3

4 | DISCUSSION

This qualitative study describes the experiences of HCs before, during and after pregnancy and their suggestions for healthcare improvement. Care improvements should be aimed at timely preparation, respecting HCs' autonomy, increasing knowledge, and improving communication among clinicians. In addition, tailoring care and education to families with haemophilia appears important.

Previous studies on HCs have described the challenges of timely diagnosis of carriership and the impact of bleeding tendencies on day-to-day life.²² In clinical practice, a diagnosis of HCs often follows when a male family member is diagnosed with haemophilia.³⁰ Only clotting factor levels are determined in Dutch female neonates, whereas genetic analysis is generally performed from 16 years onwards to allow the neonate to be able to decide for themselves, causing uncertainty in mothers about the bleeding risk of their new-born daughter in the meantime. Early diagnostics based on emerging techniques, such as genetic analysis on cell-free foetal DNA isolated from maternal blood, can offer the opportunity to provide early, safe, patient-tailored counselling. Even with a clear diagnosis, healthcare protocols are not as clear for carriers as they are for men with haemophilia. Preconception counselling might come too late: the search for information and realization of choices only arise when HCs are aware of their carriership. The HCs in this study indicated that timely diagnosis, genetic counselling and preconception counselling should always be offered. Educating families and young HCs frequently on the availability of preconception care at CCCs will empower them to seek advice when they are ready.

Major choices during each reproductive phase requires patient-tailored guidance. Women appreciated clear information provision and multi-disciplinary counselling. Here, common insecurities of HCs regarding bleeding risks and concerns about the distance to the a CCC when delivery starts, can jointly be discussed and acted upon (e.g., inducing labour). HCs desired care provision with a personal and accepting atmosphere, as in midwifery care, or even shared-care with a midwife. Good communication among the different clinicians is important, as some HCs who opted out of prenatal diagnostics felt defensive when having to clarify their choice repeatedly. Some of these themes might be applicable to the general obstetric population as well, though

likely not to the same extent as in HCs. Overall, HCs indicated that they should be able to make well-informed decisions for themselves, even though some choices may be difficult to understand for clinicians.

The central role of family experience has previously been acknowledged as both an information source and a main contributor to HC perspectives.^{14-16,18} Within families, knowledge on carrier bleeding problems and therapeutic management options is often limited. Educating families on this topic seems to be a good starting point. Throughout each reproductive phase, the perceived severity of haemophilia and the coping of family members strongly influences reproductive choices. For some women, these family experiences were traumatic, commonly originating from a period of time when haemophilia treatment was limited. In these cases, inclusion of a social worker in the multidisciplinary team and peer support is highly valued. Therefore, family experience should be taken into account while further guiding consecutive reproductive phases.

The reliability and consistency of this study were promoted through researchers trained in qualitative research, independent coding by two researchers and theme discussion in a multidisciplinary research team. Validation was embedded through triangulation of researchers and respondent validation. Limitations of the study are linked to the generalizability of the results: from a high-income country, all participants of Dutch ethnicity and the majority of originating from one CCC. HCs who have received prophylaxis and experienced a postpartum haemorrhage were underrepresented in our study population; thus the themes related to these events should be interpreted with caution. Postpartum haemorrhage experiences have been previously investigated by another research group.²⁹

5 | CONCLUSION

The challenges HCs face during reproductive phases call for awareness. In the current era of rapidly progressing haemophilia treatment and obstetrical diagnostic procedures, the reproductive choices of HCs have evolved but remain as important as ever. Clinicians from different backgrounds can use these insights to meet the needs of these women. The variation in the experiences HCs have with haemophilia within their families requires a patient-tailored approach. Future efforts to improve haemophilia care should be aimed at constructing accessible educational strategies and information transfer for involved clinicians and families with haemophilia.

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CONFLICTS OF INTEREST

Punt MC, Teela L, Bloemenkamp KWM, Pekel L, Driessens MHE, Haverman L, Lely AT: none. Fischer K: KF has received speaker's fees from Bayer, Baxter/Shire, Biotest, CSL Behring, Octapharma, Pfizer and Novo Nordisk; performed consultancy for Bayer, Baxter/Shire, Biogen, CSL Behring, Freeline, Novo Nordisk, Pfizer, Roche and SOBI;

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AUTHOR CONTRIBUTIONS

Marieke C. Punt, Lorynn Teela and Lotte Haverman interviewed the participants. Marieke C. Punt and Lorynn Teela conducted the coding and ran the first analyses. Marieke C. Punt, Lorynn Teela, Lotte Haverman, Kathelijin Fischer and Karin P.M. van Galen formed the research team and analysed and interpreted the coding together. Marieke C. Punt prepared the first draft of the article, which was revised by Lorynn Teela, Lotte Haverman, Karin P.M. van Galen, Kitty W.M. Bloemenkamp, A. Titia Lely, Mariette H.E. Driessens and Lynnda Pekel.

DATA AVAILABILITY STATEMENT

The anonymous data that support the findings of this study are available from the corresponding author upon reasonable request.

REFERENCES

- McIntock C. Women with bleeding disorders: clinical and psychological issues. *Haemophilia*. 2018;24:22-28.
- Plug I, Mauser-Bunschoten EP, Bröcker-Vriens AHJT, et al. Bleeding in carriers of hemophilia. *Blood*. 2006;108(1):52-56.
- Paroskie A, Gailani D, Debaun MR, Sidonio RF. A cross-sectional study of bleeding phenotype in haemophilia A carriers. *Br J Haematol*. 2015;170(2):223-228.
- Lavery S. Preimplantation genetic diagnosis of haemophilia. *Br J Haematol*. 2009;144(3):303-307.
- Ljung R, Tedgård U, Mcneil T, et al. How do carriers of hemophilia experience prenatal diagnosis by fetal blood sampling?. *Clin Genet*. 1987;31(5):297-302.
- Morris M, Glass M, Wessels T-M, Kromberg JGR. Mothers' experiences of genetic counselling in Johannesburg, South Africa. *J Genet Couns*. 2015;24(1):158-168.
- Mårtensson A, Tedgård U, Ljung R. Prenatal diagnosis of haemophilia in Sweden now more commonly used for psychological preparation than termination of pregnancy. *Haemophilia*. 2014;20(6):854-858.
- Tabor A, Alfirevic Z. Update on procedure-related risks for prenatal diagnosis techniques. *Fetal Diagn Ther*. 2010;27(1):1-7.
- Punt MC, Waning M, Mauser-Bunschoten EP, et al. Peripartum management and outcomes in hemophilia carriers: a systematic review. *EAHAD*. 2019. Abstract.
- Carroll M, Daly D, Begley CM. The prevalence of women's emotional and physical health problems following a postpartum haemorrhage: a systematic review. *BMC Pregnancy Childbirth*. 2016;16(1).
- Zaat TR, Van Steijn ME, De Haan-Jebbink JM, et al. Posttraumatic stress disorder related to postpartum haemorrhage: a systematic review. *Eur J Obstet Gynecol Reprod Biol*. 2018;225:214-220.
- Myrin-Westesson L, Baghaei F, Friberg F. The experience of being a female carrier of haemophilia and the mother of a haemophilic child. *Haemophilia*. 2013;19(2):219-224.
- Wiley RE, Khoury CP, Snihur AWK, et al. From the voices of people with haemophilia A and their caregivers: challenges with current treatment, their impact on quality of life and desired improvements in future therapies. *Haemophilia*. 2019;25(3):433-440.
- Gillham A, Greyling B, Wessels T-M, et al. Uptake of genetic counselling, knowledge of bleeding risks and psychosocial impact in a South African cohort of female relatives of people with hemophilia. *J Genet Couns*. 2015;24(6):978-986.
- Kadir RA, Sabin CA, Goldman E, et al. Reproductive choices of women in families with haemophilia. *Haemophilia*. 2000;6(1):33-40.
- Markova I, Forbes CD, Inwood M, Optiz JM. The consumers' view of genetic counseling of hemophilia. *Am J Med Genet*. 1984;17(4):741-752.
- Tedgard U, Ljung R, Mcneil T, et al. How do carriers of hemophilia experience prenatal diagnosis (PND)? Carriers' immediate and later reactions to amniocentesis and fetal blood sampling. *Acta Paediatr Scand*. 1989;78(5):692-700.
- Thomas S, Herbert D, Street A, et al. Attitudes towards and beliefs about genetic testing in the haemophilia community: a qualitative study. *Haemophilia*. 2007;13(5):633-641.
- Punt MC, Aalders TH, Bloemenkamp KWM, et al. The experiences and attitudes of hemophilia carriers around pregnancy: a qualitative systematic review. *J Thromb Haemost*. 2020;18(7):1626-1636.
- Perrin GQ, Herzog RW, Markusic DM. Update on clinical gene therapy for hemophilia. *Blood*. 2019;133(5):407-414.
- Weyand AC, Pipe SW. New therapies for hemophilia. *Blood*. 2019;133(5):389-398.
- Noone D, Skouw-Rasmussen N, Lavin M, et al. Barriers and challenges faced by women with congenital bleeding disorders in Europe: results of a patient survey conducted by the European Haemophilia Consortium. *Haemophilia*. 2019;25(January):468-474.
- Balkaransingh P, Young G. Novel therapies and current clinical progress in hemophilia A. *Ther Adv Hematol*. 2018;9(2):49-61.
- Brown LJ, La HA, Li J, Brunner M, et al. The societal burden of haemophilia A. I - A snapshot of haemophilia A in Australia and beyond. *Haemophilia*. 2020;26(S5):3-10.
- Lindvall K, Von Mackensen S, Elmståhl S, et al. Increased burden on caregivers of having a child with haemophilia complicated by inhibitors. *Pediatr Blood Cancer*. 2014;61:706-711.
- Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Heal Care*. 2007;19(6):349-357.
- Green J, Thorogood N. *Qualitative Methods for Health Research*. Sage Publications Ltd; 2018.
- Dierckx De Casterlé B, Gastmans C, Bryon E, et al. QUAGOL: a guide for qualitative data analysis. *Int J Nurs Stud*. 2012;49(3):360-371.
- Vandermeulen H, Petrucci J, Floros G, et al. The experience of postpartum bleeding in women with inherited bleeding disorders. *Res Pract Thromb Haemost*. 2019;3(4):733-740.
- Arya S, Wilton P, Page D, et al. "Everything was blood when it comes to me": understanding the lived experiences of women with inherited bleeding disorders. *J Thromb Haemost*. 2020. Published online.

SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

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