Received: 16 March 2020 | Revised: 8 April 2020 | Accepted: 27 April 2020

DOI: 10.1111/hae.14040

ORIGINAL ARTICLE

Haemophilia WILEY

Unravelling the knowledge, beliefs, behaviours and concerns of Persons with Haemophilia and their carriers in Senegal

Awa Babington-Ashaye[1] | Saliou Diop[2] | Antoine Geissbuhler[1] | Philippe de Moerloose[1]

[1]Faculty of Medicine, University of Geneva, Geneva, Switzerland

[2]Cheikh Anta Diop University, Dakar, Senegal

Correspondence
Awa Babington-Ashaye, Faculty of Medicine, University of Geneva, Campus Biotech, Chemin des Mines 9, 1202 Geneva, Switzerland.
Emails: awa.c.sarr@gmail.com, awa.sarr@etu.unige.ch

Abstract

Introduction: In many sub-Saharan countries, haemophilia exists in an environment of poor knowledge and poor access to treatment. To improve the quality of life of Persons with Haemophilia (PWH), understanding their unmet needs and the socio-cultural realities is essential.

Aim: This study aims to explore disease knowledge, beliefs, behaviours and concerns of PWH and carriers as a way to find adapted solutions to address the unmet needs.

Methods: Based on an interview guide, we performed a qualitative study with in-depth interviews of 26 PWH and 14 carriers.

Results: Eighty per cent of adult PWH were able to name the severity of haemophilia, but only 32% could describe with accuracy the mode of transmission of haemophilia. Only 23% of carriers were able to inform the severity of the disease. All carriers and adult PWH acknowledged at least one visit to a traditional healer. Acceptance of the disease through religion is the dominant coping strategy observed. High costs of treatment, fear of social rejection, difficulty of management of pain and bleeding at home were the main concerns.

Conclusions: Results demonstrate important gaps in knowledge, especially within the carrier population, mothers in Africa playing particularly an important role in the survival and empowerment of PWH. Findings also indicate the important weight of cultural determinants in disease management and behaviours of PWH and thus their important role in the development of educational materials taking into account these determinants.

KEYWORDS

carriers, developing countries, haemophilia, qualitative study, Senegal

1 | INTRODUCTION

Haemophilia is a genetic condition affecting 1 out of 10 000 people around the world. The lack of diagnosis is one of the main health challenges for PWH, as only 25% of the global haemophilia patient population have been identified.[1] In Senegal, for example, a study reports a prevalence of 2.3/100 000 male births, accounting for only 11.6% of the expected figure in the country.[2] Very few studies have focused on haemophilia in sub-Saharan Africa, the epidemiological data being mainly obtained from Senegal,[2] South Africa,[3] Zimbabwe,[4] Nigeria,[5] Cameroon [6] and Ivory Coast.[7,8] These studies highlight worrying trends as reported by Diop et al[2] who emphasize

that most of the patients die very early in their childhood, during untreated haemorrhagic episodes due to the inaccessibility of factor concentrates replacement. Indeed, the life expectancy of PWH remains low, as the mean age of the patients in 2014 was 15 years in Senegal.[2] Prophylaxis is today the main goal of treatment, though prophylaxis access remains very limited in low-resource countries. Thanks to the World Federation of Haemophilia (WFH) humanitarian aid programme, 14 children were under prophylaxis in 2018 in Senegal.

This study aims to explore disease knowledge, beliefs, behaviours and concerns of PWH and carriers as a way to find adapted solutions to address the unmet needs.

2 | STUDY POPULATION AND METHODS

2.1 | Population

Participants were enrolled on a volunteering basis and contacted by telephone for recruitment using the registry of the Senegalese National Member Organization and with the help of the National Blood Transfusion Center (CNTS). Semi-structured interviews with PWH and carriers were conducted. Knowledge, beliefs, attitudes and behaviours were investigated during the study. The interviews were mainly performed in Dakar at the CNTS. In Thiès (located at 70 km from Dakar), home visits to PWH and relatives were performed. The inclusion criteria were the following: adult PWH with expressed willingness to communicate their experience, children with haemophilia recruited during the World Haemophilia Day in 2018 and obligate carriers with at least one son with haemophilia.

In accordance with the Declaration of Helsinki, informed consent was obtained from all participants, parents of children, and data were anonymously transcribed.

2.2 | Methods

A total of 40 interviews were performed using semi-structured interviews and focus groups discussions. The main researcher (AWB) followed an interview guide; face-to-face discussions were recorded. Written notes were also taken. The interviews lasted between 45 minutes and 1 hour. The discussions were conducted in French for 76% of adult PWH and the majority of children whereas 93% of the interviews of carriers were conducted in Wolof, a local language spoken in Senegal. A staff of the CNTS or a member of the Senegalese association of PWH supported the researcher ensuring understanding of questions and answers from the study population.

The interview guide (see Table S1) included several categories, combining predetermined open questions and themes with the opportunity for the interviewer to explore specific responses further.

2.3 | Analysis

The results of the interviews were analysed using the thematic analysis framework.[9] Emerging themes were then defined based on the codes and clustered into categories. A theme was considered emerging if it appeared in at least half in each population of PWH and carriers. Relevant quotes were assigned to the different themes. In order to preserve the anonymity of the testimonials, participants have been referred as follow: Persons with Haemophilia: PWH1 to PWH26, Carriers C: C1 to C14.

3 | RESULTS

3.1 | Study population and circumstance of diagnosis

As shown in Table 1, 3 main categories of participants were considered (adult PWH, children PWH and carriers).

Sixty-six per cent of carriers mentioned bleeding following post-traumatic events as the main circumstances of diagnosis of their sons. Carriers shared their ignorance about the cause of their children's bleeding and even after that they were told that their son has a disease named haemophilia. 33% of adult participants shared that circumcision was the main event that triggered the diagnosis, a percentage also found in Ivory Coast.[8] Only 16% of carriers expressed that circumcision was the main trigger to diagnosis of their sons, and this can be explained by the young age of their children, which consequently have not yet been circumcised at the time of their diagnosis. Circumcision represents an act of great religious value and social recognition for the boys and is performed in Senegal.[10] The failure of this step induces stigmatization and a social burden to carry, as expressed by the participants.

3.2 | Learnings and challenges experiences by PWH and carriers

3.2.1 | Disease knowledge gap

Eighty per cent of carriers were able to name the disease but only 23% were able to inform the severity of the disease. Carriers shared ignorance, daily difficulties and lack of knowledge about the actions to prevent their child to be injured.

Eighty per cent of adult PWH were able to name the type and severity of haemophilia, hesitancy shown when they were asked to mention the severity of their haemophilia in some rare cases. Only 32% spontaneously mentioned the inheritance of the disease. Moreover, we noticed a lack of accurate knowledge about the genetic transmission. 69% of adult PWH declared having relatives with haemophilia (including brothers, uncles and nephews), and also, 25% declared at least one sibling deceased from haemophilia.

All adult PWH could express the main clinical aspects of the disease. Unlike carriers, all adults could mention the name of their

TABLE 1 Socio-demographic data of Senegalese PWH and carriers

Characteristics	Total participants N = 40	Person with Haemophilia (PWH) N = 26	PWH adults N = 17	PWH-children N = 9	Carriers N = 14
Mean Age (y), range (min-max)		24.6 (6-59)	33.9 (19-59)	12.1 (6-17)	34.5
Place of residence, n (%)					
Dakar		18 (69.3)	12 (70.6)	6 (66.6)	4 (28.6)
Outside Dakar		8 (30.7)	5 (29.4)	3 (33.3)	10 (71.4)
Marital status, n (%)					
Married			5 (29.4)	N/A	14 (100)
Divorced			2 (11.8)	N/A	0
Single			10 (58.8)	N/A	0
With children, n (%)			6 (35.2)	N/A	0
Children with haemophilia				N/A	1.21
Professional activity n (%)				N/A	
Employed			4 (23.5)	N/A	1 (7.1)
Self-employed			6 (35.3)	N/A	3 (21.4)
Total with professional activity (Employed + Self-employed)			10 (58.9)	N/A	4 (28.6)
Unemployed			3 (17.6)	N/A	
Student			4 (23.5)	0	
Pupils (Koranic or State School)				9 (100)	
Housewives			N/A	N/A	10 (71.4)
Diagnosis (n, %)					
Haemophilia A		24 (92.3)	17 (100)	7 (77.7)	
Severe		18 (69.2)	13 (76.5)	5 (55.5)	
Moderate		5 (19.2)	3 (17.6)	2 (22,2)	
Mild		1 (3.8)	1 (5.9)	0	
With inhibitors		4 (15.4)	3 (17.6)	1 (11.1)	
Haemophilia B		2 (7.4)	0	2 (22.2)	
Severe		2 (7.4)	0	2 (22.2)	
With inhibitors		0	0	0	

N/A, not applicable.
*Aged above 18 years old.

treatment (referred as factors) and were able to describe its primary functionality.

Children were able to describe accurately the symptoms. They had a very limited knowledge about the inheritance. Interestingly, none of the children see the disease as an obstacle for their future. The majority said that they want to embrace a medical career perceived as a profession compatible with their disease and considered safe.

3.2.2 | Expressed needs in terms of knowledge

The main areas of knowledge gaps expressed by participants are described in Table 2.

Adult PWH shared their willingness to manage the symptoms while at home. They also aimed to know more about prophylaxis with the hope that it would be available for them 1 day and seek for a better understanding of the mode of transmission of haemophilia.

Carriers unanimously acknowledged not having enough information. They expressed the little they know: the child is sick, and when bleedings do not stop, they have to take him to the hospital. Carriers also shared that they want to know whether homemade solutions (applying shea butter or other plant-based lotions) are correct approaches and what should be done in case of emergency.

3.2.3 | Beliefs and behaviours

We observed noticeable rooted cultural beliefs. The first expression of belief in spiritual forces appeared while the participants

TABLE 2 Education needs expressed by adult PWH and carriers

Adult PWH N = 26	Carriers N = 14
1. Transmission of the disease, inheritance pattern	1. Why does my child have haemophilia?
2. Prevention of the risks related to their condition	2. How to take care of bleedings and pain before getting to hospital?
3. Home management of joint pain and bleedings	3. Level of severity of symptoms that should urge them go the hospital
4. Treatment: Prophylaxis knowledge and other existing treatments	4. Risk preventions at home and school

shared the circumstances of the first symptoms of haemophilia. Carriers mentioned spiritual forces called 'Djinns' during their explanation of the origin of their son's symptoms. The belief in Djinns is anchored in a cultural rather than religious belief. The ignorance of the cause of a phenomenon is a trigger; in this case, symptoms of haemophilia sometimes trigger associated beliefs that the symptoms are a sort of punishment targeted to the specific individuals.

Participants mentioned visits to traditional healers in their seek of remedy to haemophilia. In our study, the reference to traditional healers was cited by the participants by evoking terms such as 'Fajuwolof' a person using medical care based on the use of traditional pharmacopeia and they mentioned as well visiting '*Serigne*' (Practitioner using prayers and religious esoteric formulas).

Carriers and adult PWH acknowledged at least one visit to a traditional healer at different stages of their journey with haemophilia. When they were ignorant about haemophilia, it was the only solution perceived to treat the symptoms. Interestingly, even after a confirmed diagnosis of haemophilia, visits continued to traditional healers in a way to keep the hope alive to be completely cured.

This specific thematic captured the different behaviours and attitudes of carriers and adult PWH towards haemophilia. The expression and observation of psychological and emotional impact of haemophilia on both populations were also analysed (Table 3). PWH often have to deal with challenges regarding their social integration and psychological impairments, so we aimed to seek comparatively the status of Senegalese PWH.

Most (94%) of the adult PWH population expressed an acceptance of their condition through their faith to God; however, at the same time 29.1% expressed also denial. The majority (58.8%) of adult PWH shares a 'low profile behaviour', which means that they were hiding their haemophilia to friends and even within family by fear of rejection. Despite important absenteeism at work in some cases, PWH rather not share their condition with their colleagues anticipating ignorance that could lead to exclusion. Absenteeism at work is felt as a real barrier to career progression. Interestingly, PWH were socially integrated with either a job (58.9%) or as a student (23.5%). The diagnosis of haemophilia was described as a 'Wake-up' call of God as shown in Table 3.

The adolescent years were often cited as the life period where PWH suffered the most from the different physical and social limits of the disease. The constraints triggered behaviours such as transgression of certain rules and willingness to live like their friends. PWH shared a willingness to pursue different activities even if it implied disobeying to parents and putting their health at risk. The expression of rebellion and denial were mostly related to childhood and adolescence years.

Coping strategies associated with their faith in God were mentioned by all carriers. The reference to God is related to fate, a

TABLE 3 Behaviours and attitudes of PWH towards their condition

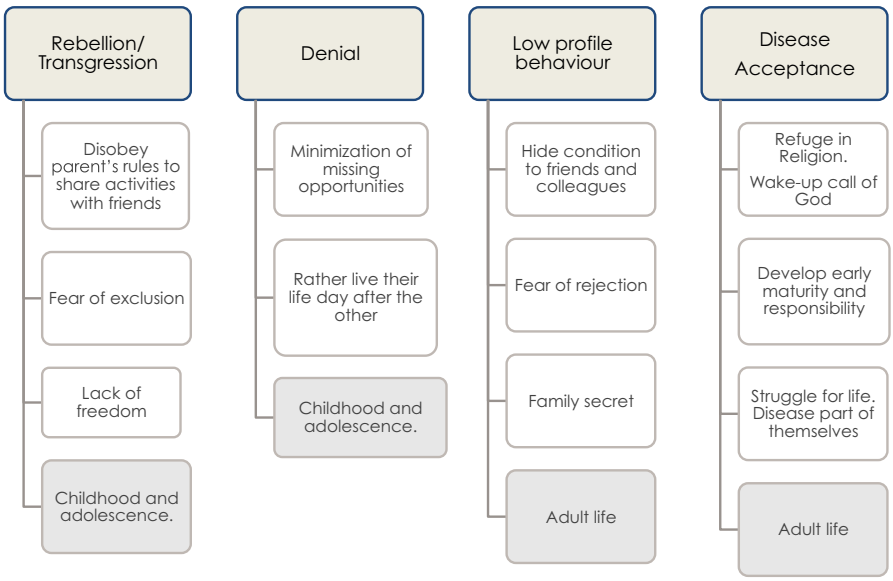
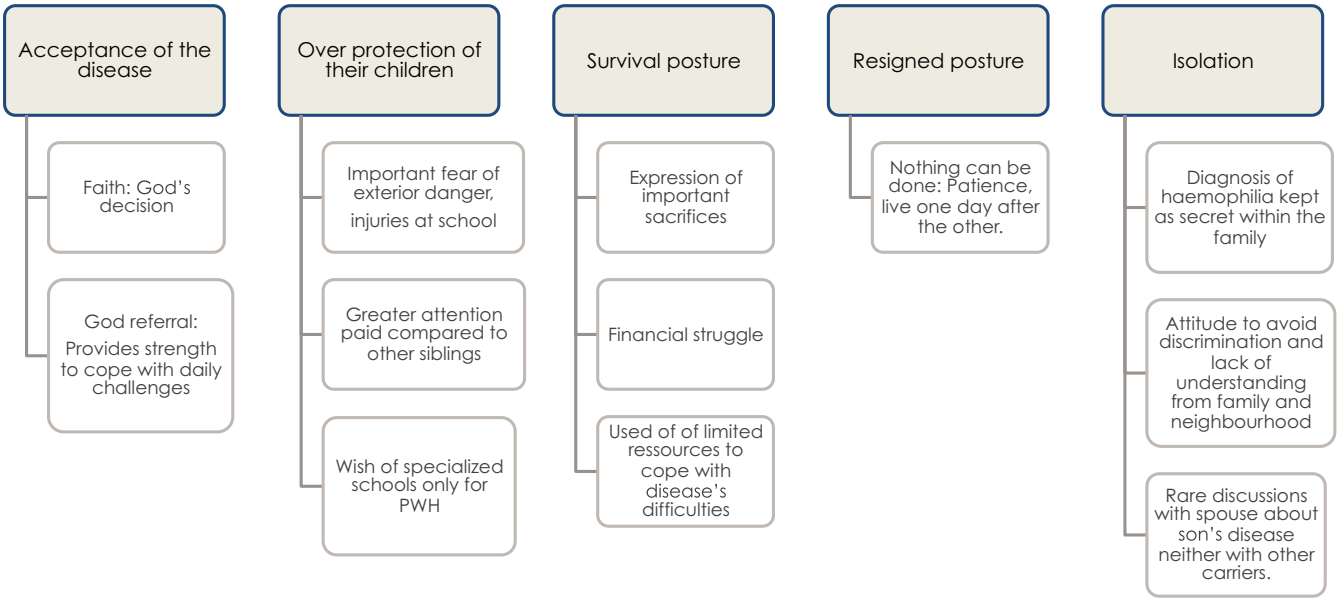


TABLE 4 Behaviours and attitudes of carriers towards their son's haemophilia



decision from God to give them a child with haemophilia. Their faith also explained as way to help them to overcome the challenges related to the disease. They also explained that they have difficulties to avoid any harm done to their sons as shown in Table 4.

They also expressed regrets to not always provide appropriate emergency care to their children, and this observation triggered behaviours such as overprotecting their children for 71% of them. Solutions were found such as to lock their children in their bedroom or to bring them along to their work place. This behaviour was understood as the immediate and accessible way to ensure well-being of their children. Overcoming the difficulties of living with the disease implies a lot of sacrifices and carriers adopted what we qualified as a survival posture expressed by 78% of this population. As shown also in Table 4, carriers disclosed their reluctance to share their son's haemophilia and their own daily difficulties even within the family-fearing stigmatization and rejection; thus, they tend to isolate themselves. Three carriers shared the negative comments and blame received in their family house especially by the in-law family. Carriers expressed gratitude to be able to share their challenges during the interview as they said having rarely such opportunity. Finally, they also shared social pressure of being a carrier and deal with the other incumbents of mother, spouse and housewife.

3.2.4 | Main concerns: better access to care and improve disease management

Participants pointed out different challenges in their daily management of the disease. The predominant key challenges expressed were the financial difficulties, the social impact of haemophilia, the lack of treatment access and the difficulty of pain management in home setting. The difficulty to manage the financial costs associated with the disease (hospitalization, medical prescriptions and transport costs) is undoubtedly important. While acknowledging the ongoing efforts to reduce the costs of treatment, they complained about the cost of overall medical care. In addition to the burden of financial cost carriers, they also cited the daily disease management at home as a key difficulty, particularly the supervision while having to take care of other siblings. Carriers living outside Dakar deplored the cost and distance to reach the CNTS.

The main concerns shared by adult PWH, in addition to the cost of clotting factors, are the treatment access, the lack of prophylaxis, the poor supply in regions outside Dakar and the difficulty of pain management. Both PWH and carriers tackled differences in the quality of care management between the capital and other cities; while they praised the availability and professionalism of the medical staff of the CNTS of Dakar, as they feel listened and have answers to their questions, they expressed dissatisfaction regarding the management of haemophilia in cities outside Dakar, emphasizing the need to develop a close collaboration between CNTS and proximity care centres.

4 | DISCUSSION

The main findings of our research are (a) the gaps in knowledge of haemophilia of Senegalese carriers and PWH and (b) the importance of socio-cultural determinants such as the weight of religion and visits to the traditional healers, both having impact on carriers and PWH's day-to-day experiences with the disease. These findings are both original and complementary to the existing literature.

Results demonstrate striking gaps in knowledge, especially concerning the exact mode of transmission of the disease. Above all, God's decision and Djinns are often mentioned as explanations of

symptoms. Religion plays a central role in Senegal, and this is also expressed in this situation. All carriers and 93% of adult PWH expressed their faith to God as a main coping strategy to deal with the difficulties. Religiosity and spirituality are actually amongst the methods cited in the literature that contribute to support patients and family to cope with chronic illness.[11] As an example, in the case of sickle-cell disease, Strickland et al[12] concluded 'Religion serves as a coping insulator'.

The poor understanding of the genetic transmission of haemophilia explains also the frequent visits to traditional healers not only to treat the symptoms but also to be completely cured. Carriers expressed that they felt pressure of family to do so, even when diagnosis was confirmed. In African countries, traditional healers are recognised as legitimate healers.[13] Traditional healers in communities could be better informed and integrated in care management. This is consistent with conclusions of Mac-Seing et al[14] claiming the necessity to promote health equity for all and the sharing of both resources and knowledge within partnerships in global health researches. The acceptance of haemophilia is high in adults who however share a 'low profile' attitude, which means hiding haemophilia by fear of rejection.

The situation is especially difficult for carriers who are sometimes blamed to have a son with haemophilia. Few studies have been conducted so far in sub-Saharan countries on haemophilia carriers.[15-17] A recent study conducted in Ivory Coast illustrates the lacking awareness of the haemophilia carrier condition and its implications for carriers, PWH's families, and medical community.[18] While improving the level of education is essential, the need of thorough adapted guidelines to improve local identification of carriers and provide them necessary psychological and genetic counsel is also suggested.[18,19] Problematic access to treatments due to financial difficulties is mentioned by all participants as a major concern; carriers mentioned specifically the costs of transportation since in this study most of them lived outside Dakar. The WFH has taken several actions in sub-Saharan Africa, which have already important beneficial impact of haemophilia care.[20]

By being more empowered, PWH should not only gain better skills to cope better with their daily disease requirements, but this would encourage them to spread knowledge and thereby empower others. Appropriate, culturally adapted educational tools are likely to contribute to better disease awareness, prevention and self-management, as also recently demonstrated in Ivory Coast.[21] The use of information and communication technology (ICT) in health care, E-health, is explored as a tool to improve education and the access to information the quality of care.[22]

5 | CONCLUSION

The important gaps in knowledge observed in PWH and carriers suggest that improving education is crucial. Our study yielded insights into some fundamental components of socio-cultural determinants regarding haemophilia in Senegal. The results can support intervention in three main complementary directions: (a) empowerment and education of young Senegalese PWH from early age, so that they acquire sufficient health literacy and haemophilia knowledge management, (b) leverage carriers' fundamental role in the empowerment and education of PWH through disease management training and (c) development of culturally adapted educational solutions.

ACKNOWLEDGEMENTS

The authors thank all participants, the translators (I. F and A. N), the members of the Senegalese patient Association (ASH) and the medical team of the National Blood Transfusion Center.

DISCLOSURES

The authors stated that they had no interests, which might be perceived as posing a conflict of interest or bias.

AUTHOR CONTRIBUTIONS

AWB participated in the design of research, conducted the field research and wrote the manuscript. SD, AG and PDM provided expert consultation and reviewed and approved the final version of the manuscript.

ORCID

Awa Babington-Ashaye https://orcid.org/0000-0001-6595-7645
Saliou Diop https://orcid.org/0000-0002-2354-3839
Antoine Geissbuhler https://orcid.org/0000-0001-5039-3373

REFERENCES

1. World Federation of Hemophilia. Global annual survey 2017. https://www.wfh.org/en/data-collection. Accessed July 17, 2019.
2. Diop S, Seck M, Sy-Bah D, et al. Implementing haemophilia care in Senegal. West Africa. *Haemophilia*. 2013;20:73-77.
3. Mahlangu JN. Haemophilia care in South Africa: 2004–2007 look back. *Haemophilia*. 2019;15:135-141.
4. Adewuyi JO, Coutts AM, Levy L, Lloyd SE. Haemophilia care in Zimbabwe. *Centr Afr Med*. 1996;42:153-156.
5. Mba EC, Kulkami AG, Fleming A. Haemophilia in the Northern Nigeria. *Centr Afr Med*. 1995;41:59-62.
6. Yimlefack NC, Tagny CT, Ndoumba AM, Pauline NB, Ngum MD. Assessing the quality of care for haemophilia at the Yaounde reference treatment centre for Cameroon. *Blood Coagul Fibrinolysis*. 2017;28:176-180.
7. Sangare A, Sanogo I, Koffi CI, et al. Prevalence and clinical profile of haemophilia of black African in urban areas of the Ivory Coast. *Med Trop*. 1990;50:176-180.
8. Lambert C, Meité N, Sanogo I, et al. Haemophilia in Côte d'Ivoire (the Ivory Cosat) in 2017: extensive data collection as part of the World Federation of Hemophilia's twinning programme. *Haemophilia*. 2019;25:236-243.
9. Brown V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol*. 2006;3:77-101.
10. Seck M, Sagna A, Guéye MS, et al. Circumcision in hemophilia using low quantity of factor concentrates: experience from Dakar, Senegal. *BMC Hematol*. 2017;17:4-9.
11. Rambod M, Sharif F, Molazem Z, Khair K. Spirituality experiences in hemophilia patients: a phenomenological study. *J Relig Health*. 2019;58:992-1002.

12. Strickland OL, Jackson G, Gilead M, McGuire DB, Quarles S. Use of focus groups for pain and quality of life assessment in adults with sickle cell disease. *J Natl Black Nurses Assoc*. 2001;12:36-43.
13. Yeboah T. Improving the provision of traditional health knowledge for rural communities in Ghana. *Health Libr Rev*. 2000;17:203-208.
14. Mac-Seing M, Perez Osorio C, Tine S, et al. Une santé mondiale repensée : la perspective de futures chercheuses-praticiennes. *Can J Public Health*. 2017;108:e452-e454.
15. Dangerfield BT, Manga P, Field SP, Hartman E, Jenkins T, Krause A. Feasibility of prenatal diagnosis and carrier detection in South African haemophilia A patients. *Br J Haematol*. 1997;97:558-560.
16. Gillham A, Greyling B, Wessels TM, et al. Uptake of genetic counseling, knowledge of bleeding risks and psychosocial impact in a South African cohort of female relatives of people with hemophilia. *J Genet Couns*. 2015;24:978-986.
17. Seck M, Faye BF, Sall A, et al. Bleeding risk assessment in hemophilia A carriers from Dakar, Senegal. *Blood Coagul Fibrinolysis*. 2017;28:642-645.
18. Lambert C, Meité ND, Sanogo I, et al. Hemophilia carrier's awareness, diagnosis, and management in emerging countries: a cross-sectional study in Côte d'Ivoire (Ivory Coast). *Orphanet J Rare Dis*. 2019;14:26.
19. Naicker T, Aldous C, Thejpal R. Haemophilia: a disease of women as well. *South Afr J Child Health*. 2016;10:29-32.
20. Diop S, Haffar A, Mahlangu J, Chami I, Kitchen S, Pierce G. Improving access to hemophilia care in sub-Saharan Africa by capacity building. *Blood Adv*. 2019;3(Suppl 1):1-4.
21. Lambert C, Meité N, Sanogo I, Lobet S, Hermans C. Development and evaluation of appropriate, culturally adapted educational tools for Ivoirian patients with haemophilia, haemophilia carriers and their families. *Haemophilia*. 2019;25:838-844.
22. Bediang G, Perrin C, Ruiz de Castañeda R, et al. The RAFT telemedicine network: lessons learnt and perspectives from a decade of educational and clinical services in low- and middle-incomes countries. *Front Public Health*. 2014;2:180.

SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

How to cite this article: Babington-Ashaye A, Diop S, Geissbuhler A, de Moerloose P. Unravelling the knowledge, beliefs, behaviours and concerns of Persons with Haemophilia and their carriers in Senegal. *Haemophilia*. 2020;00:1–7. https://doi.org/10.1111/hae.14040