

# Assessing the factors affecting the accessibility of primary dental care for people with haemophilia

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## Abstract

**Introduction:** Patients with haemophilia (PWH) often have difficulty accessing dental services.

**Aim:** To determine the accessibility of dental care for PWH and to examine their perceptions of how coronavirus type-2 (CoV-2) disease (COVID-19) has affected their ability to access dental treatments following the pandemic.

**Methods:** The questionnaire survey was conducted between July 2022 and December 2022 at haemophilia treatment centres in Hungary. Variables with statistical significance (Pearson's Chi-squared test;  $p < .05$ ) were included in logistic regression analyses. Least absolute shrinkage and selection operator (LASSO) regression was used as a machine learning technique to identify the most predictive variables.

**Results:** Twenty-one percent of the sixty-eight participants reported that they had been refused dental treatment, mainly in primary care (86%). Dental refusal was influenced by infectious disease (OR: 4.48, CI: 1.14–17.69) and previous dental bleeding complications (OR: 4.23, CI: 1.10–16.27). There was correlation between dental visits and having a permanent dentist or receiving oral hygiene advice (OR: 9.95, CI: 2.86–34.62 and OR: 3.84, CI: 1.09–13.58). Participation in an oral hygiene consultation increased patients' satisfaction with their dental care (OR: 6.28, 95% CI: .71–55.88). Twenty-eight percent of patients had experienced difficulties since the start of the COVID-19, but 84% had visited their dentist at least once between 2021 and 2022 ( $p = .002$ ). Nevertheless, 16% of respondents went for only the most necessary treatments due to pandemic.

**Conclusion:** Refusal of dental care was high among participants, especially in primary care. The COVID-19 pandemic has exaggerated the difficulties of PWH in accessing dental treatment.

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**KEYWORDS**

COVID-19, dental care, haemophilia A, haemophilia B, machine learning, surveys and questionnaires

**Highlights**

- Patients with haemophilia (PWH) have difficulty accessing dental care, and the coronavirus type 2 (CoV-2) disease pandemic (COVID-19) has created a new barrier.
- The study revealed a high prevalence of dental care refusal (21%), particularly in primary care (86%).
- This 2022 survey found that 28% of patients experienced difficulties since the pandemic started and 16% only sought necessary treatments.

**1 | INTRODUCTION**

Oral health is an important aspect of quality of life (QoL) in general and in particular in patients with bleeding disorders (BD).<sup>1,2</sup> Dental care of patients with haemophilia (PWH) represents a largely unmet need of their comprehensive management program.<sup>3–8</sup> In addition to previously existing difficulties, patients had to face challenges due to the outbreak of coronavirus type-2 (CoV-2) disease (COVID-19) pandemic since 2019.<sup>9–11</sup> The obstacles to obtaining adequate dental care can result in poor oral hygiene, thereby increasing the necessity for more invasive dental treatments.<sup>4</sup> Prevention and early detection of dental diseases is of paramount importance among PWH, as most common non-surgical procedures can be performed in a general dental practice (GDP) provided that a haematologist is involved, and guidelines are followed.<sup>2,4,12</sup>

Only a few groups from the Americas and the United Kingdom investigated the access to dental care for PWH.<sup>3–8</sup> No survey on dental care experience of PWH has yet been published from continental Europe.

The aim of this study was to extend the research on the accessibility of dental care for PWH, with a particular focus on primary dental care. We also aimed at collecting data on patients' perceptions of how COVID-19 pandemic has affected access to dental treatments.

**2 | MATERIALS AND METHODS****2.1 | Study design**

This multicentre cross-sectional study was performed between July and December 2022 in four major Hungarian haemophilia treatment centres (HTCs) (National Hemophilia Center and Hemostasis Department, Medical Center of the Hungarian Defense Forces—Budapest; Heim Pal Children's Hospital—Budapest; Clinical Center of the University of Debrecen—Debrecen; Mohács Hospital—Mohács). Inclusion and exclusion criteria are shown in Table 1.

We used a self-administered anonymous questionnaire developed for this study based on literature review and expert opinion of the senior dentist and haematologist (IM and CK).<sup>3–8,13</sup> Children under 16 years of age completed the questionnaire with the help of their guardians. A pilot test of the questionnaire was carried out with 10 PWH (eight adults, two children) from the Debrecen centre. Ethical approval was obtained (Regional and Institutional Ethics Committee, Clinical Center, University of Debrecen; No. DE RKEB/IKEB: 6087-2022). Study was conducted in compliance with the Helsinki Declaration.

**2.2 | Sample population and data collection**

Study participants were recruited from the participating HTCs. Participation was voluntary. Written informed consent was obtained from study patients and/or by legal guardians. A priori sample size calculation, incorporating a 10% allowance for dropout and nonresponse, determined the need for 58 participants (adjusted to 64) to detect a difference with means of 64.01 and 82.43, standard deviations (SD) of 26.26 and 22.85, respectively, with 80% power and a two-sided alpha of .05.<sup>14</sup> During data collection period, 80 patients were invited, and 68 patients enrolled (response rate: 85%). The questionnaires were completed in HTCs ( $n = 63$ ; 93%) or online via a Microsoft Forms link ( $n = 5$ ; 7%).

**TABLE 1** Inclusion and exclusion study criteria.

Inclusion criteria	Patients with haemophilia A and B (any severity) Female conductors with low FVIII/IX level (FVIII/IX activity <50%)
Exclusion criteria	Additional haemostatic disorder Severe cognitive impairment Lack of signed written informed consent Participating in the pilot study to develop the study questionnaire

**TABLE 2** Variable specification.

Variable	Description
Questionnaire sections	Nos. 1–2: Questions on birth and gender
	Nos. 3–9: Health condition
	Nos. 10–29: Dental care
	No. 30: Question on the impact of COVID-19 on dental care
Level of dental care	General dental practitioner: primary care level—free of charge financed by the National Health Insurance Fund of Hungary
	Private dental care: primary care level—fee-paying Specialist institution or care centre: advanced level of care—free of charge financed by the National Health Insurance Fund of Hungary
Age groups	0–18 years
	19–44 years
	>45 years
Haemophilia severity	Severe <1%
	Moderate 1%–5%
	Mild 5%–40%
Inhibitors	Low-titer inhibitors <5 BU/mL
	High-titer inhibitors ≥5 BU/mL

Abbreviations: BU, Bethesda unit; COVID-19, coronavirus type-2 (CoV-2) disease.

## 2.3 | Variable specification

The questionnaire contained 30 questions (Table S1) and was divided into four sections (Table 2). In regard to the level of dental care, the statistical analysis was based on the Hungarian healthcare system, and three age groups were delineated (Table 2).<sup>15</sup> Haemophilia severity defined conventionally by factor VIII and IX levels.<sup>2</sup> Patients with severe or moderate haemophilia and those with mild haemophilia were assessed separately. Inhibitors were categorized conventionally by Bethesda titer (BU/mL).<sup>16</sup>

## 2.4 | Variable selection and statistical analysis

Categorical variables were compared using Pearson's Chi-squared test. Multiple logistic regression models were built to investigate the factors influencing the frequency of visits to the dentist, refusal of dental treatment and patients' views on the dental care options for PWH. To enhance the robustness of our multiple logistic regression models and ensure the selection of the most predictive variables, we employed the Least Absolute Shrinkage and Selection Operator (LASSO) regression technique. Findings from the logistic regression analyses were represented as odds ratios (ORs) and 95% confidence intervals (CIs). Statistical evaluations were executed using STATA IC Version 17.0 software.<sup>17</sup> A  $p$ -value < .05 was considered significant.

## 3 | RESULTS

### 3.1 | Patient demographic and disease characteristics

Demographic data and disease characteristics are presented in Table 3.

### 3.2 | Descriptive and multivariate analyses of access and quality of dental care

#### 3.2.1 | Frequency of visits to dental practice

Severe and moderate versus mild PWH were compared to investigate the effect of haemophilia severity on frequency of dental visits (Table 4). Results indicated that a significantly higher proportion of patients with mild haemophilia visited a dentist in the preceding year than patients with severe or moderate haemophilia ( $p = .026$ ). Furthermore, age had a significant effect on the frequency of dental visits ( $p = .033$ ) (Table 4). The data revealed a markedly higher frequency of dental visits among individuals within the 0–18 age group (51.35%). Results indicated that comorbidities and negative experiences of refusal by dentists did not significantly impact the frequency of visits (Table 4).

A multiple logistic regression model based on LASSO selection was used to examine the effect of a permanent dentist, oral hygiene consultation attendance, age and type of haemophilia on the frequency of visiting a dental office. Participants having a permanent dentist ( $n = 37$ ; 54%) had higher odds of visiting a dental practice (OR: 9.95, 95% CI: 2.86–34.62). Moreover, patients who have ever attended an oral hygiene consultation ( $n = 28$ ; 41%) had higher odds of visiting a dental office (OR: 3.84, 95% CI: 1.09–13.58), mostly “in the workplace/school” ( $n = 15$ ; 54%) or “in a private dental practice” ( $n = 11$ ; 39%). The analysis revealed that the age and type of haemophilia did not have a significant impact on the likelihood of visiting the dentist (OR: 1.01, 95% CI: .97–1.05 and OR: .20, 95% CI: .02–1.85).

Most participants not attending a dentist in the previous year denied any dental problems ( $n = 24$ ; 77%) (Table 5). The second most frequent response was delayed treatments due to the COVID-19 epidemic ( $n = 5$ ; 16%). Impact of COVID-19 pandemic on access to dental care is shown in Table 6.

#### 3.2.2 | Refusal by dental care-providers

Fourteen (21%) respondents have already been refused at least once because of their BD (Table 7). Twelve (86%) such events happened in primary dental care financed by the National Health Insurance Fund of Hungary. Pearson's Chi-squared test revealed a statistically significant association between age group and the refusal of dental care ( $p = .020$ ) (Table 7). Furthermore, refusal rate was found to be significantly higher in the presence of comorbidities ( $p = .006$ ). Neither the severity of haemophilia nor the availability of a regular dentist

**TABLE 3** Demographic and disease characteristics of participants.

Characteristics (n = 68)			
Gender			
Female	1.4% (1)		
Male	98.5% (67)		
Age (years)			
Mean (SD)	28.59 (±17.08)		
Median with interquartile range	26 (14–41.50)		
Range			
Children (<18 years of age)	36.7% (25)		
Adults (≥18 years of age)	63.2% (43)		
Type and severity of haemophilia			
<u>Haemophilia A</u>		<u>Haemophilia B</u>	
88.2% (60)	11.8% (8)		
Severe	90.0% (54)	Severe	25.0% (2)
Moderate	3.3% (2)	Moderate	12.5% (1)
Mild <sup>a</sup>	6.7% (4)	Mild	62.5% (5)
Presence of inhibitors			
10.2% (7)			
Low-titer inhibitors	42.8% (3)		
High-titer inhibitors	0% (0)		
Titer not available	57.1% (4)		
Comorbidities			
70.5% (48)			
Infectious disease			
60.4% (29)			
Current	5.8% (4)		
Previously	36.7% (25)		
HCV	65.5% (19)		
HBV	17.2% (5)		
HIV	3.4% (1)		
Other	20.6% (6)		
Hypertension			
25.0% (12)			
Other <sup>b</sup>			
14.5% (7)			
Diagnosis of haemophilia due to oral bleeding			
7.3% (5)			
Haemophilia A <sup>c</sup>		Haemophilia B <sup>d</sup>	
40.0% (2)		60.0% (3)	

Abbreviations: HBV, hepatitis B virus; HCV, hepatitis C virus; HIV, human immunodeficiency virus; SD, standard deviation.

<sup>a</sup>Female carrier.

<sup>b</sup>Other comorbidities include Gastroesophageal reflux disease (GERD) ( $n = 2$ ); Gilbert's syndrome ( $n = 2$ ); Crohn's disease ( $n = 1$ ); Diabetes mellitus ( $n = 1$ ); Insulin resistance ( $n = 1$ ); Osteoporosis ( $n = 1$ ); Clubfoot ( $n = 1$ ); Cataract ( $n = 1$ ); Glaucoma ( $n = 1$ ); Allergy (not specified) ( $n = 1$ ).

<sup>c</sup>Severe ( $n = 1$ ) and mild ( $n = 1$ ) haemophilia A.

<sup>d</sup>Severe ( $n = 1$ ), moderate ( $n = 1$ ) and mild ( $n = 1$ ) haemophilia B.

significantly affected the refusal rate (Table 7). Sixty-nine percent of respondents were completely ( $n = 20$ ) or mostly ( $n = 27$ ) satisfied with the dental care options for PWH. There was a significant difference ( $p < .001$ ) in the refusal by dental care-providers between satisfied ( $n = 47$ ) and dissatisfied patients ( $n = 7$ ) (Table 7). All dissatisfied patients were refused. A multiple logistic regression model based on LASSO selection was used to examine the effect of infectious disease, bleeding episodes following dental procedures and type of haemophilia on refusal of dental care. Those patients who have or ever had an infectious disease ( $n = 29$ ; 43%) had higher odds for refusal (OR: 4.48, 95% CI: 1.14–17.69). Patients who ever experienced bleeding or swelling after dental procedures ( $n = 24$ ; 35%) were also more likely to experience refusal (OR: 4.23; 95% CI: 1.10–16.27). In the course of the questionnaire survey, three (4%) respondents concealed their infectious disease for fear of rejection, however, none of the respondents concealed their haemophilia.

A multiple logistic regression model based on LASSO selection was used to examine the effect of oral hygiene consultation attendance, type of haemophilia, infectious disease and bleeding episodes following dental treatment on patients' assessment of their dental care options. Participants who had ever attended an oral hygiene consultation ( $n = 28$ ; 41%) had higher odds of being satisfied with the dental care options for PWH (OR: 6.28, 95% CI: .71–55.88). Patients with severe and moderate haemophilia, infectious disease, or experienced bleeding signs or swelling after a dental procedure were characterized by lower odds of satisfaction (OR: .60, 95% CI: .04–8.94, OR: .07, 95% CI: .01–.89 and OR: .13, 95% CI: .02–1.03, respectively).

### 3.2.3 | Dental complaints and treatments

Table 8 presents a summary of patients' responses to questions regarding dental complaints and treatments. The majority of respondents reported no dental complaints ( $n = 58$ ; 85%). Fifty-four (79%) participants had some dental procedure, with the most common interventions being restorative treatments. Twenty-four (35%) respondents experienced prolonged oozing, bleeding, or swelling after a dental treatment. Most bleeding occurred after surgical procedures ( $n = 17$ ). In case of dental pain, the majority ( $n = 41$ ; 60%) of participants would consult a dentist first. If patients had a choice of any level of care for dental procedures, 15% of the participants would choose a treatment to be performed by GDP, while most (65%) of them would go to a private practice.

### 3.2.4 | Knowledge on dental care options and patients' evaluation of dentists

The majority of participants ( $n = 45$ ; 66%) considered themselves to be mostly informed about the dental care options available for PWH (Table 9). Patients who were fully informed ( $n = 9$ ; 13%), expressed satisfaction. Moreover, the majority of participants in both the mostly

**TABLE 4** Patient characteristics on factors influencing frequency of dental visits.

Frequency of visits to dental practice	Attended visit % (n = 37)	Did not attend visit % (n = 31)	p-value
Severity of haemophilia			
Severe/Moderate	49.15% (29)	50.85% (30)	
Mild	88.89% (8)	11.11% (1)	.026*
Age			
0–18	51.35% (19)	22.58% (7)	
19–44	29.73% (11)	58.06% (18)	
45+	18.92% (7)	19.35% (6)	.033*
Comorbidities			
Infectious disease			
Present	75.00% (3)	25.00% (1)	.394
Past	56.00% (14)	44.00% (11)	.841
Other comorbidity	52.63% (10)	47.37% (9)	.854
Negative experience of refusal			
	57.14% (8)	42.86% (6)	.818

**TABLE 5** Barriers to access to the dentist.

Patients who have not been to a dentist in the last year	
45.5% (n = 31)	
No complaints	77.4% (24)
Lack of time	12.9% (4)
Difficulties in getting to the dentist <sup>a</sup>	3.2% (1)
Fear of dental procedures	9.6% (3)
Fear of bleeding or injury during dental treatment	6.4% (2)
Fear of bleeding after a dental procedure	6.4% (2)
Negative experiences because of haemophilia/infectious disease	12.9% (4)
Financial considerations	3.2% (1)
COVID-19 epidemic	16.1% (5)

Abbreviation: COVID-19, coronavirus type-2 (CoV-2) disease.

<sup>a</sup>For example, distance or transport.

**TABLE 6** The impact of COVID-19 on dental visit.

Question 30 (Multiple choices)	n	%	Visiting dentist	No dental visit	p-value
1	19	27.94	84.21	15.79	.002*
1;3	2	2.94	50	50	.899
2	1	1.47	100	0	.356
3	9	13.24	55.56	44.44	.941
4	15	22.06	73.33	26.67	.096
5	22	32.35	13.64	86.36	<.001*

1 = At the start of the COVID-19 outbreak, I experienced significant difficulties, such as difficulty getting an appointment with a dentist or being refused by dentists.

2 = Even with the improvement of the COVID-19 epidemic situation, I still don't feel that it is easier to get an appointment at the dentist, it is still difficult to get care.

3 = In my opinion, dental care has always been difficult, and the COVID-19 epidemic has only exacerbated the problem.

4 = In my opinion, the COVID-19 epidemic has not affected access to dental care.

5 = I do not know.

**TABLE 7** Patient characteristics associated with refusing dental treatment.

Refusal by dental care-providers	Refused patients % (n = 14)	Non-refused patients % (n = 54)	p-value
Level of denial of health care (Multiple choices)			
Primary care dentist	85.71% (12)		
Private practice	28.57% (4)		
Specialist institution or care centre	7.14% (1)	-	-
Age			
0–18	14.29% (2)	44.44% (24)	
19–44	42.86% (6)	42.59% (23)	
45+	42.86% (6)	12.96% (7)	.020*
Comorbidities			
Infectious disease (Present/Past)	71.43% (10)	31.48% (17)	
No infectious disease	28.57% (4)	68.52% (37)	.006*
Other comorbidity	57.14% (8)	20.37% (11)	
No other comorbidity	42.86% (6)	79.63% (43)	.006*
Severity of haemophilia			
Severe/Moderate	92.86% (13)	85.19% (46)	
Mild	7.14% (1)	14.81% (8)	.450
Regular dentist			
Yes	64.29% (9)	51.85% (28)	
No	35.71% (5)	48.15% (26)	.405
Satisfaction			
Satisfied patients (Completely/Mostly)	50% (7)	100% (40) <sup>a</sup>	
Dissatisfied patients	50% (7)	.00% (0)	<.001*

<sup>a</sup>Totals may sum to less in some strata due to non-applicable responses.

informed and uninformed ( $n = 14$ ; 21%) groups reported no significant obstacles to accessing dental care.

A total of 14 parents of 25 children (56%) with haemophilia were unaware that the first dental check-up is recommended before the age of 1 year.

The survey respondents reported that dentists were fully ( $n = 7$ ) or mostly prepared ( $n = 13$ ) for the management of PWH (together, 29%). None of the participants considered dentists unprepared, although 34 (50%) of them said dentists know the disease, but their knowledge was very incomplete.

## 4 | DISCUSSION

This survey is the first study reporting on access to dental care for PWH in continental Europe and one of the very few studies reporting on the impact of COVID-19 on dental treatments for people with BD. The study paid particular attention to primary care. To be treated in a specialized care centre, patients face many obstacles, such as travel distance or long waiting times for treatments. In addition to haemophilia, these barriers place an additional burden on patients, which can put dental care at the bottom of the priority list. Fortunately, most of the

participants (54%) of this study had visited a dentist at least once in the previous year, similar to the findings of other investigators.<sup>4,8</sup> According to our data, severity of haemophilia exerted a significant impact on frequency of dental visits among patients. Possible explanation is that patients at a higher risk of bleeding encounter greater difficulties in finding dentists willing to provide them with care as suggested by the results of Frusca do Monte et al.<sup>8</sup> Nevertheless, according to our results, dentists who actually treated the patients were not influenced by the severity of haemophilia.

Similar to Fiske et al.,<sup>3</sup> we demonstrated that children (0–18 years of age) were more likely to attend dental appointments. This observation highlights the need for oral hygiene education at an early age and making children familiar with dental visits, which can alleviate anxiety and promote oral health.<sup>18,19</sup>

The survey found a high rate of dental treatment refusal among participants. Twenty-one percent of patients were refused at least once by a dentist because of haemophilia and 86% of the refused participants had been rejected by GDP. However, primary care would give patients faster access to treatments that do not require advanced level of care. The refusal rate was similar to that found by three groups from the US and UK varying between 18% and 29%.<sup>3,4,8</sup> A study from Saudi Arabia reported a much higher refusal rate (67%).<sup>6</sup> Prevalence of refusal

**TABLE 8** Patients' responses to questions about dental complaints and treatments.

Dental complaints and treatments	
<i>n</i> = 68	
Dental complaints (multiple choices)	
No complaints	85.29% (58)
Sensitive teeth	10.29% (7)
Intermittent pain	1.47% (1)
Acute pain	.00% (0)
Gum bleeding	1.47% (1)
Jaw problem	1.47% (1)
Other <sup>a</sup>	4.41% (3)
Dental procedure (multiple choices)	
Never had a dental procedure	20.58% (14)
Restorative dental treatments <sup>b</sup>	55.88% (38)
Tooth extraction/wisdom tooth removal	41.17% (28)
Scaling and polishing/cleaning	39.70% (27)
Orthodontic treatment	14.70% (10)
Prosthetic treatments <sup>c</sup>	13.23% (9)
Other dental surgical intervention <sup>d</sup>	2.94% (2)
Prolonged oozing, bleeding or swelling after a dental procedure (multiple choices)	
Not happened	51.47% (35)
After tooth extraction/wisdom tooth removal	23.52% (16)
After restorative treatments	4.41% (3)
After scaling and polishing/cleaning	2.94% (2)
After making dentures/while wearing dentures	1.47% (1)
During orthodontic treatment	1.47% (1)
After other dental surgical intervention <sup>e</sup>	1.47% (1)
In case of dental pain consultation with	
Dentist	60.29% (41)
Haematologist	30.88% (21)
General Practitioner	.00% (0)
None; tolerate the pain	4.41% (3)
Do not know what to do	4.41% (3)
Patient's choice of level of care	
Private practice	61.76% (42)
Specialised care facility or centre	23.52% (16)
Primary care, general dentist	14.70% (10)

<sup>a</sup>Other complaints include: ongoing root canal treatment (*n* = 1); ongoing restorative and prosthetic treatments (*n* = 2).

<sup>b</sup>For example, filling, root canal treatment.

<sup>c</sup>For example, denture, crown, bridge.

<sup>d</sup>Other dental surgical intervention was: deciduous teeth removal (*n* = 1); unknown (*n* = 1).

<sup>e</sup>Other dental surgical intervention was: circumcision of the wisdom tooth.

was higher among older individuals similar to Frusca do Monte et al.<sup>8</sup> As adults undergo a greater number of dental examinations over their lifespan than children and adolescents they are more likely to experience refusal. Moreover, the complexity of dental treatment needs and medical comorbidities are greater among older individuals, which presents a challenge in providing appropriate care. Our findings corroborate other studies that having demonstrated a negative impact of current or previous infectious diseases on access to dental treatments for PWH.<sup>3,8</sup> Notably, 4% of the respondents concealed their infectious disease for fear of rejection. Comorbidities also increased hesitancy of dentists to treat PWH as well as previously noticed bleeding signs following dental procedures. Present data confirmed observations of an US survey, suggesting that finding care providers willing to treat patients with BD is a major hurdle.<sup>5</sup> Participants who had a permanent dentist had higher odds of visiting a dental practice, than those who did not. Patients may find difficult to access dental service because many dentists have limited confidence in treating PWH. Furthermore, there are notable discrepancies in the equipment and facilities utilized in dental practices.

Twenty-four (77%) of all the participants who did not attend a dentist in the previous year denied any dental problems. Other difficulties, identified as barriers, have also been reported previously (Table 5).<sup>3-7</sup> The outbreak of COVID-19 and its impact on the health sector has created new difficulties. In our survey, despite the challenges encountered, 84% of patients reporting obstacles still visited their dentist at least once between 2021 and 2022. However, 16% of respondents went for only the most necessary treatments, even though avoiding dental problems should have been a priority for this group of patients. A large proportion of respondents (32%) were unable to assess the impact of COVID-19 on dental care, probably because 86% had not visited a dentist in the preceding year.

Fifty percent of participants said dentists were aware of haemophilia, but their knowledge was incomplete. Similarly, Frusca do Monte et al. observed that 53% of patients with BD lacked confidence in the GDP's dental care ability.<sup>8</sup> In contrast, Fiske et al. and Kalsi et al. observed lower rates.<sup>3,4</sup> It is of the utmost importance to emphasize the significance of dental care for PWH and to provide assistance to dentists in this regard. It is recommended that the curriculum at the undergraduate level should place an accent on the care of patients with BD, and that this topic should continue to be addressed in postgraduate training. Furthermore, a comprehensive guide should be made readily available to healthcare professionals in their native language. The work of Hungarian dentists is supported by contents in Hungarian covering all fields of dentistry.<sup>20,21</sup> Furthermore, consultation channels need to be established with HTC's where dentists can easily access information. According to our data, in case of dental pain 31% of the participants would consult a haematologist for help, which highlights the importance of close cooperation between the two professions.

Supporting dental care and prevention for PWH can be of paramount importance, as our data show that those who have ever

**TABLE 9** Patients' knowledge and satisfaction with the dental care options available to PWH.

Patient's knowledge on dental care options	Fully informed <i>n</i> = 9	Mostly informed <i>n</i> = 45	Uninformed <i>n</i> = 14	<i>p</i> -value
Satisfied patients	100.00% (9)	86.49% (32)	75.00% (6)	.305
Dissatisfied patients	.00% (0)	13.51% (5)	25.00% (2)	
NE	0	8	6	

Abbreviation: NE, not evaluable answer (Patients do not know as they have never had dental treatment).

attended an oral hygiene consultation are more likely to visit a dental office. Forty-one percent of participants indicated that they had received such advice, a low proportion compared to the findings from a previous study (76% and 85% of adults and children, respectively).<sup>3</sup> In accordance with the recommendation of the World Federation of Haemophilia (WFH), preventative care should commence at an early age, ideally with the eruption of the first tooth.<sup>2</sup> Our survey showed that 56% of parents of children with haemophilia did not know about this guideline. Otherwise, majority of respondents felt that they were mostly informed about dental care options for PWH and completely or mostly satisfied with the dental care options. The importance of prevention is also shown by the fact that participants who had ever attended an oral hygiene consultation had higher odds to being satisfied with their dental care.

Seventy-nine percent of the participants had dental procedure and the majority of these were restorative treatments. Most dental procedures (65%) were not associated with bleeding complications, which were more commonly reported after surgical procedures. Therefore, dentists can play a key role, even in the early detection of haemophilia. According to the study by Sonis and Musselman 13.6% of the 132 participants with haemophilia were diagnosed because of a persisting oral bleeding.<sup>22</sup> We observed a rate of 7.3%.

The study emphasized the significance of patients visiting the dentist on a regular basis and receiving appropriate oral hygiene advice during dental visits. The most direct access dental treatment is through primary dental care. However, our survey indicated that this was the least preferred option for patients, presumably due to the high refusal rate. Nevertheless, we believe that dentists working in primary care have an important role to play and can help alleviate the dental care difficulties of PWH.

The strength of the survey is that it was collected in the largest HTC in the country, and 93% were completed in the dental office. The small number of patients is a limitation of the study. An international study of larger sample size comparing findings across countries would give a more accurate picture of factors influencing patients' access to dental care services. Secondly, a limitation of the questionnaire was the absence of open text domains, which compelled participants to select from predefined options. This may have resulted in a lack of comprehensive representation of their full range of thoughts and experiences. Furthermore, the questionnaire was based on self-report, which can lead to distortions of social desirability and errors in recall. In addition, patients invited to participate in the study were randomly selected from the total haemophilia population of the HTCs. The voluntary par-

ticipation of these patients can lead to the selection of a high-quality cohort.

## 5 | CONCLUSION

This study identified obstacles in the oral care of PWH, evaluating the COVID-19 epidemic as a new barrier. In conclusion, supporting dental care for PWH is of paramount importance, with particular emphasis on primary care and importance of a permanent dentist so as a fast and simple access to dental care can be available. However, our survey shows that in most cases, PWH are rejected by GDPs. In addition, COVID-19 pandemic has exaggerated the difficulties of PWH in accessing dental care.

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## CONFLICT OF INTEREST STATEMENT

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

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request due to privacy/ethical restrictions.

## ETHICS STATEMENT

This paper was approved by the Regional and Institutional Ethics Committee, Clinical Center, University of Debrecen, Hungary (registration No DE RKEB/IKEB: 6087-2022). The study has been conducted in full compliance with the Declaration of Helsinki.

## PATIENT CONSENT STATEMENT

Written informed consent was signed by participating patients and/or by their legal guardians.

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## SUPPORTING INFORMATION

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

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