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ORIGINAL ARTICLE

The state of oral health in patients with haemophilia in the Netherlands

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Abstract

Introduction: Previous studies show contradictory outcomes regarding dental, gingival and periodontal status in persons with haemophilia (PWH) compared to healthy controls. PWH may experience disease-specific barriers to access dental care due to their bleeding tendency, which may lead to delays in oral care and severe dental problems.

Aim: To determine the current subjective and objective oral health status in adult PWH.

Methods: Randomly selected PWH of the Erasmus MC Haemophilia Treatment Center (HTC), Rotterdam, the Netherlands, were invited to participate. Data was collected using the Oral Health Impact Profile (OHIP-14NL) and personal interviews. A dentist used the DMFT index, the Dutch Periodontal Screening index (DPSI), plaque and bleeding index to score the dental status.

Results: Forty-eight adult PWH were included in this study, 20 mild, 15 moderate-severe and 13 severe haemophilia with a mean age of 44.7. PWH scored low on the OHIP-14 questionnaire (median total score 1.0; IQR .0–3.0), indicating a high self-rating oral health status. The number of bleeding events, bleeding- and plaque index score was not statistically significant between patients with mild, moderate or severe haemophilia. The mean number of decayed, missing, and filled teeth (DMFT-score) was significantly lower in the group of patients with severe haemophilia (median 2.0) compared to mild haemophilia (median 16.0) ($p = .04$). Twenty-five patients (52.1%) reported to have encountered bleeding problems during or after dental interventions during their lifetime.

Conclusion: Dutch adult PWH A/B have good dental status and oral health status.

KEYWORDS
bleeding, dental examination, haemophilia, oral health, oral health impact profile (OHIP)

1 | INTRODUCTION

Haemophilia is a rare x-linked bleeding disorder mainly affecting males. The bleeding disorder is caused by a deficiency of coagulation factor VIII (FVIII) in patients with haemophilia (PWH) A or factor IX (FIX) in PWH B. PWH A or B are classified according to the residual factor activity in their blood into mild (5%–40%), moderate-severe (1%–5%) and severe (<1%) haemophilia.[1] Patients with mild haemophilia generally only suffer from bleeding after serious injury, trauma or surgery (including dental interventions), whereas patients with severe haemophilia may have spontaneous bleeding episodes. Therefore most of these patients regularly receive prophylactic treatment.[2]

In the past, dental care for PWH has often been neglected or even refused due to their (severe) bleeding tendency. In the long term lack of regular dental care may result in serious dental problems. Dental procedures were often performed under general anaesthetics, with tooth extraction as the only option for treatment.[3] With the introduction of factor concentrates dental treatments can now be performed at a general dental office.[4] However, PWH still face disease-specific barriers in accessing primary dental care. The most frequently mentioned disease-specific barrier was the dentists' unwillingness to provide treatment for PWH.[5–8] PWH still have concerns or anxiety about dental treatments, based on complications in the past or lack of knowledge about haemophilia by the dentist.[6–8] These access barriers could lead to delay or avoidance of dental check-ups and treatments. In turn, this may lead to the aggravation of dental problems and the need for even more dental interventions. Studies comparing the oral health status in children, adolescents and adults PWH with aged-matched healthy controls reported contradicting outcomes. These studies reported that PWH might have a better, similar or worse dental condition compared to the healthy controls.[8–22] Despite these contradicting outcomes, more recent studies found no difference in Oral Health-Related Quality of Life (OHR-QoL) between children and adolescents PWH and healthy controls.[13,17,21] However, adult PWH reported a lower OHR-QoL score than healthy controls.[8,17] It is unknown what the current dental status is of PWH in the Netherlands, a country with a well-organized healthcare system with dedicated HTCs. Therefore, the aim of our study was to assess the current dental care and dental status in adult PWH and to identify potential oral health problems in these patients.

2 | MATERIALS AND METHODS

2.1 | Study population

All adult patients (18 years and older) with haemophilia A or B treated at the HTC at Erasmus University Medical Center, Rotterdam, the Netherlands, (n = 311) were eligible for this study.

2.2 | Sample size

Because no data on the prevalence of dental problems in PWH in the Netherlands is available yet we assumed, based on findings of previous international studies, that the prevalence of any dental problem is 50% in PWH.[17] Based on this percentage, a sample size of 100 PWH was chosen with a 10% deviation in outcome measurements, to make a comparison with the general population. A 70% inclusion rate was expected. Therefore 141 PWH were randomly selected and invited to participate in this study. Our hospital, in the southwest of the Netherlands has a large and diverse adherence area of around 4.5 million inhabitants and has a regional function for the care of PWH. With more than 500 patients registered at our HTC, this is a representative population sample for the Netherlands.

2.3 | Recruitment and consent

PWH received the questionnaire by email. Completion of the questionnaire meant consent. After a week the nurse practitioner (GM) called the participants to make an appointment for the dental exam at the Erasmus University Medical Center. The study was approved by the Ethics Committee (2018-1211).

2.4 | Questionnaires

The patients filled in a questionnaire on baseline characteristics. (Appendix 1). Socio-economic parameters about highest education level, comorbidity, intoxication, insurance, dental care and hygiene were recorded (Appendix 1). Patients received the short Dutch version of the Oral Health Impact Profile questionnaire using 14 questions (OHIP-NL14) via a hospital portal with their own personal subject number (Appendix 2). This OHIP-14 instrument consists questions about the effects of oral conditions on daily functioning. It summarises various aspects of perceived oral health in a composite score and is the current standard to measure the impact of oral disease and dental interventions. The comprehensive questionnaire has been developed based on recommendations of the World Health Organization.[23,24] The psychometric properties, reliability and validity proved to be excellent for both short and comprehensive questionnaires and well suited for determining the OHR-QoL. The OHIP is hierarchical ranked in seven domains; functional limitation, physical discomfort (pain), psychological discomfort, physical disability, psychological disability, social deficiency and handicap. The questions within the domains are rated on a 5-point scale (never = 0, hardly ever = 1, occasionally = 2, often = 3 and very often = 4).

2.5 | Dental examination and interview

The nurse practitioner (GM) interviewed the patients about their experience with oral health and dental care related to haemophilia (Appendix 4). Before the dental examination, it was recorded if the patient had taken prophylactic clotting factor concentrate. The subsequent dental examination consisted of scoring the soft and hard oral tissues.

2.6 | Statistical analysis

The statistical analyses were only performed on patients who both filled in the questionnaire and were examined by the dentist (n = 48). The variables were checked for normal distribution (Kolmogorov–Smirnov and Shaprio–Wilk test). Based on these outcomes, parametric or non-parametric statistical analyses were performed. The data were analyzed using the Statistical Package for Social Sciences (SPSS)[26] version 21.0 (SPSS Inc., Chicago, IL) taking the cut-off level for statistical significance at .05.

3 | RESULTS

For this study, 141 patients were invited to participate and received the questionnaires (OHIP-NL14 and questions related to age, education, general health, oral status and behaviour). Sixty-one patients (43.3%) agreed to participate and were invited for the dental examination. A total of 13 patients did not undergo a dental examination because they were not able to visit the hospital due to various reasons, including COVID restrictions. Forty-eight PWH (78.7%) were eventually seen for dental examination.

3.1 | Patient characteristics

The majority of patients had haemophilia A (n = 40, 83.3%). The severity of the haemophilia was: 20 mild (41.7%), 15 moderate-severe (31.2%) and 13 severe (27.1%) PWH. The mean age was 44.7 years (SD 16.8, range 19–76). There was a significant difference in age between the different haemophilia severity groups. Patients with severe haemophilia were younger (34 ± 15.3; mean ± SD) than patients with mild haemophilia (53.5 ± 15) (p = .002). The patients reported various levels of education based on the International Standard Classification of Education[27]; lower (n = 7, 14.6%), medium (n = 14, 29.1%) and higher (n = 27, 56.3%). We found no difference in education between the different severities of haemophilia (p = .229). Other health conditions, besides haemophilia, were reported by 16 patients (33.3 %). Seven of these patients had a condition (e.g., diabetes, von Willebrand disease, Crohn's disease) or medication (e.g., antidepressants, anticonvulsive drugs) that may be associated with a higher bleeding tendency or possible influence on oral health (e.g., parodontitis, dry mouth and gingival hyperplasia) (Table 1). Information concerning health insurance is given in Appendix 6.

3.2 | Preventive dental behaviour

Patients were asked about their daily oral routine. The self-reported outcome shows that 31 (72.1%) of the dentate patients brush their teeth ≥2 min per day and 33 (76.7%) brush ≥2 min per cleaning session. At least 33 patients (76.7%) use fluoride toothpaste and 18 (41.9%) use daily interdental cleaning products (e.g., sticks, brushes or floss). Use of mouthwash was reported by 12 patients (27.9%), because of bad breath, caries prevention and dental plaque reduction.

3.3 | Oro-facial problems

Nine dentate patients (20%) reported problems with the teeth related to caries, fractures, sensitivity or pain, mobility and dental calculus. Two edentate patients reported problems related to lack of retention and insufficient chewing ability with their dentures. In both groups, dentate and edentate, mucosal problems were described with sensitivity or pain, gum bleeding and mucosal recessions (n = 5, 10.4%). Only one patient (2.1%) reported joint sounds as a problem related to the temporomandibular joint. The dentate patients reported that their problems are not addressed by a dental care giver.

3.4 | Bleeding events encountered

In total, 25 patients (52.1%) reported to have encountered a total of 29 bleeding events during or after dental procedures. Bleeding incidents occurred in 11 out of 20 patients (55%) with mild PWH, 10 out of 15 patients (67%) with moderate and 4 out of 13 (30%) with severe haemophilia. There is no significant difference in number of bleeding events between the severities of haemophilia. Bleeding events occurred after interventions by the maxillofacial surgeon (n = 20), dentist (n = 7) and dental hygienist (n = 2). Multiple answers were possible. Unfortunately, no information is available about the use of haemostatic treatments including desmopressin (DDAVP), coagulation factor concentrates (CFC) or the use of tranexamic acid (TXA) before or after the procedures. While brushing 16 of the dentate patients (37.2%) reported regular gum bleeding. Patients that reported bleeding gums during brushing and/or interdental cleaning have a higher bleeding score than patients who do not experience gum bleeding during cleaning (p = .004). However, there is no significant statistical difference between the severity of haemophilia and reported gum bleeding (p = .903).

3.5 | Oral-health-related quality of life

Table 2 shows the results of the individual items of the OHIP Questionnaire, which assesses the quality of oral health. The median of all items separately and the total score of the questionnaire for the severity levels of haemophilia are given in Table 2. No difference in the total OHIP-score was observed for the haemophilia severities (p = .526), education (p = .225) and dental status (p = .437). An extend report of

TABLE 1 Patients characteristics

Patients with haemophilia			Severity			
		Total n (%)	Mild n (%)	Moderate n (%)	Severe n (%)	*p*-value
General						
AAge (mean, SD)		44.7 (16.8)	53.5 (15.0)	42.4 (14.3)	34.0 (15.3)	
EEducation	**Low**	7 (14.6)	4 (8.3)	3 (6.3)	0 (.0)	
	Medium	14 (29.2)	6 (12.5)	2 (4.2)	6 (12.5)	
	High	27 (56.3)	10 (20.8)	10 (20.8)	7 (14.6)	
						.229**
Health						
Type	**A**	40 (83.3)	18 (37.5)	13 (27.1)	9 (18.8)	
	B	8 (16.7)	2 (4.2)	2 (4.2)	4 (8.3)	
Comorbidity		16 (33.3)	9 (18.8)	5 (10.4)	2 (4.2)	
Medication (not clotting factor)		16 (33.3)	10 (20.8)	5 (10.4)	1 (2.1)	
Allergies		14 (29.2)	6 (12.5)	4 (8.3)	4 (8.3)	
Smoking		8 (16.7)	4 (8.3)	2 (4.2)	2 (4.2)	
Alcohol		43 (89.6)	19 (39.6)	14 (29.2)	10 (20.8)	
Drugs		3 (6.3)	0 (.0)	1 (2.1)	2 (4.2)	
Dental care						
Dentist	**No**	3 (6.3)	3 (6.3)	0 (0)	0 (0)	
	Yes	45 (93.8)	17 (35.4)	15 (31.3)	13 (27.1)	
Dental check-up (per year)	**Only with problems**	6 (12.5)	3 (6.3)	2 (4.2)	1 (2.1)	
	1×	10 (20.8)	4 (8.3)	4 (8.3)	2 (4.2)	
	2×	27 (56.3)	11 (22.9)	8 (16.7)	8 (16.7)	
	>2	5 (10.4)	2 (4.2)	1 (2.1)	2 (4.2)	
Last dental appointment (months)	**<6**	35 (72.9)	15 (31.3)	11 (22.9)	9 (18.8)	
	6–12	6 (12.5)	1 (2.1)	2 (4.2)	3 (6.3)	
	>12	7 (14.6)	4 (8.3)	2 (4.2)	1 (2.1)	
Dental bleeding incident*	**yes**	25 (52.1)	11 (22.9)	10 (20.8)	4 (8.3)	
	no	23 (47.9)	9 (18.8)	5 (10.4)	9 (18.8)	
						.156***
Dental Hygiene (n = 43, excl. 3 edendate and 2 missing)						
Tooth brushing (daily)	**Not daily**	1 (2.1)	0 (.0)	1 (2.3)	0 (.0)	
	1×	11 (25.6)	4 (9.3)	3 (7.0)	4 (9.3)	
	2×	30 (69.8)	13 (30.2)	10 (23.3)	7 (16.3)	
	>2×	1 (2.3)	0 (.0)	0 (.0)	1 (2.3)	
Tooth brushing session (minutes)	**<2**	10 (23.3)	7 (16.3)	2 (4.7)	1 (2.3)	
	2	21 (48.8)	7 (16.3)	6 (14.0)	8 (18.6)	
	>2	12 (27.9)	3 (7.0)	6 (14.0)	3 (7.0)	
I Interdental cleaning	**Never**	8 (18.6)	0 (.0)	1 (2.3)	7 (16.3)	
	Daily	18 (41.9)	7 (16.3)	8 (18.6)	3 (7.0)	
	Weekly	12 (27.9)	9 (20.9)	3 (7.0)	0 (.0)	
	Monthly	5 (11.6)	1 (2.3)	2 (4.7)	2 (4.7)	

(Continues)

TABLE 1 (Continued)

Patients with haemophilia			Severity			
		Total n (%)	Mild n (%)	Moderate n (%)	Severe n (%)	*p*-value
Bleeding gums*	No	27 (52.8)	10 (23.3)	9 (20.9)	8 (18.6)	
	Yes	16 (37.2)	7 (16.3)	5 (11.6)	4 (9.3)	
						.903****
iIntra oral (n = 48/*n = 45, excl. 3 edentate)						
Dentate	without partial denture	42 (87.5)	16 (33.3)	14 (29.2)	12 (25.0)	
	with partial denture	1 (2.1)	1 (2.1)	0 (.0)	0 (.0)	
Dentate vs. edentatejaw with denture		2 (4.2)	1 (2.1)	0 (.0)	1 (2.1)	
Edentate	Denture without implant	3 (6.3)	2 (4.2)	1 (2.1)	0 (0)	
	Denture with implant	0 (.0)	0 (.0)	0 (.0)	0 (.0)	
DMFT-index (med, IQR)	D (decay)	.0 (.0, .8)	.0 (.0, .0)	.0 (.0, 2.0)	.0 (.0, .0)	
	M (missing)	.0 (.0, 2.0)	1.0 (.0, 7.0)	1.0 (.0, 2.0)	.0 (.0, .0)	
	F (filled)	4.5 (.0, 12.0)	10,0 (2.5, 15.5)	5.0 (.0, 9.0)	1.0 (0.0, 10.0)	
	T (total)	11.5 (2.3, 16.8)	16.0 (7.0, 20.0)	10.0 (2.0, 13.0)	2.0 (.0, 14.0)	
						.010*****
DPSI-category*	A (score 0,1 & 2)	30 (66.7)	10 (22.2)	9 (20.0)	11 (24.4)	
	B (score 3-)	11 (24.4)	5 (11.1)	4 (8.9)	2 (4.4)	
	C (score 3+ & 4)	4 (8.9)	3 (6.7)	1 (2.2)	0 (.0)	
						.404******
Bleeding index score % (med, IQR)*		30.0 (23.0, 42.0)	30.0 (16.0, 48.0)	23.5 (18.0, 33.0)	33.0 (26.0, 43.0)	
						.567*******
Plaque index score % (med, IQR)*		30.0 (22.0, 45.0)	33.0 (20.0, 69.0)	22.5 (21.0, 36.0)	32.0 (37.0, 40.0)	
						.364********

*Life time events.
**No difference in education between the different severities of haemophilia (X2(4) = 5.622, $p = .229$).
***The highest number of bleeding incidents occurred in patients with mild haemophilia (n = 11, 22.9%) and the lowest within patients with severe haemophilia (n = 4, 8.3%), but no statistically significant difference was found between the level of severity (X2(2) = 3.713, $p = .156$).
****No significant statistical difference between the severity of haemophilia and reported gum bleeding (X2(2) = .205, $p = .903$).
*****For the DMFT-score no difference was found between severity groups (H(2) = 9.280, $p = .010$).
******Between the different levels of severity there was no difference for and DSPI-category (X2(4) = 4.018, $p = .404$).
*******Between the different levels of severity there was no difference for the bleeding index score (F(2,43) = .576, $p = .567$).
********Between the different levels of severity there was no difference for the plaque score (H(2) = 2.002, = .364).

the dental examination, including DMFT, DPSI and bleeding and plaque index (Appendix 5).

3.6 | Personal interviews of patients

The nurse practitioner (G.M.) interviewed the patients before dental examination about their experiences with oral health and (preventive) dental care related to haemophilia (Appendix 4). A few older patients had poor experiences in the past with dental care and interventions. Since the use of prophylactic treatment of clotting factor concentrate PWH can safely undergo treatment for dental disease with much less risk of bleeding, this has led to improvement in patient adherence to regular dental check-ups and a better oral QOL.

4 | DISCUSSION

The most important finding of our study is that dental and oral health status in PWH in the Netherlands is in general very good. Almost all patients included in this study have a dentist and are routinely screened for dental care once or two times a year. No patient has been refused treatment by a dentist due to disease-specific reasons and fear of bleeding complications. The majority of the patients obtain a den-

TABLE 2 Results of the OHIP questionnaire

Response (0-4) n (%)						Statistics med (Q1, Q3)			
OHIP-14 questions and domains	Never= 0	Seldom= 1	Sometimes= 2	Often= 3	Always= 4	Total	Mild	Moderate	Severe
Functional limitations									
1. Have you had trouble pronouncing any words because of problems with your teeth, mouth or denture?	45 (93.8)	3 (6.3)	0 (.0)	0 (.0)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, .0)
2. Have you felt that your sense of taste has worsened because of problems with your teeth, mouth or denture?	46 (95.8)	2 (4.2)	0 (.0)	0 (.0)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, .0)
Physical pain									
3. Have you had a painful aching in your mouth?[a]	27 (56.3)	17 (35.4)	1 (2.1)	2 (4.2)	1 (2.1)	.0 (.0, 1.0)	1.0 (.0, 1.0)	.0 (.0, 1.0)	.0 (.0, 1.0)
4. Have you found it uncomfortable to eat any foods because of problems with your teeth, mouth or denture?[b]	38 (79.2)	7 (14.6)	2 (4.2)	1 (2.1)	0 (.0)	.0 (.0, 1.0)	.0 (.0, 1.0)	.0 (.0, 1.0)	.0 (.0, .0)
Psychological discomfort									
5. Have you been self-conscious because of your teeth, mouth or denture?	38 (79.2)	7 (14.6)	1 (2.1)	1 (2.1)	1 (2.1)	.0 (.0, .5)	.0 (.0, .0)	.00 (.0, 1.0)	.0 (.0, .0)
6. Have you felt tense because of problems with your teeth, mouth or denture?	38 (79.2)	8 (16.7)	0 (.0)	2 (4.2)	0 (.0)	.0 (.0, 1.0)	.0 (.0, .0)	.0 (.0, 1.0)	.0. (.0, .5)
Physical disability									
7. Has your diet been unsatisfactory because of problems with your teeth, mouth or denture?	41 (85.4)	5 (10.4)	2 (4.2)	0 (.0)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, 1.0)	.0 (.0, .0)
8. Have you had to interrupt meals because of problems with your teeth, mouth or denture?[c]	40 (83.3)	6 (12.5)	0 (.0)	2 (4.2)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.00 (.0, 1.0)	.0 (.0, .0)
Psychological disability									
9. Have you found it difficult to relax because of problems with your teeth, mouth or denture?[d]	41 (85.4)	6 (12.5)	0 (.0)	1 (2.1)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, 1.0)	.0 (.0, .0)
10. Have you been a bit embarrassed because of problems with your teeth, mouth or denture?[e]	40 (83.3)	7 (14.6)	0 (.0)	1 (2.1)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, 1.0)	.0 (.0, .0)
Social disability									
11. Have you been a bit irritable with other people because of problems with your teeth, mouth or denture?	40 (83.3)	7 (14.6)	0 (.0)	1 (2.1)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, 1.0)	.00 (.0, .0)
12. Have you had difficulty doing your usual jobs because of problems with your teeth, mouth or denture?	43 89.6)	4 (8.3)	1 (2.1)	0 (.0)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, 1.0)	.0 (.0, .0)

(Continues)

TABLE 2 (Continued)

Response (0-4) n (%)						Statistics med (Q1, Q3)			
OHIP-14 questions and domains	**Never= 0**	**Seldom= 1**	**Sometimes= 2**	**Often= 3**	**Always= 4**	**Total**	**Mild**	**Moderate**	**Severe**
Social handicap									
13. Have you felt that life in general was less satisfying because of problems with your teeth, mouth or denture?	44 (91.7)	2 (4.2)	1 (2.1)	1 (2.1)	0 (.0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, .0)	.0 (.0, .0)
14. Have you been totally unable to function because of problems with your teeth, mouth or denture?	45 (93.8)	3 (6.3)	0 (.0)	0 (.0)	0 (.0)	.0 (.0, .0)	.0(.0, .0)	.0 (.0, .0)	.0 (.0, .0)
	Total OHIP- scores (0–56) n (%)					**Statistics med (Q1, Q3)**			
	0–5	**6–10**	**11–15**	**16–21**	**25–30**	**Total**	**Mild**	**Moderate**	**Severe**
	40 (83.3)	3 (6.3)	2 (4.2)	2 (4.2)	1 (2.1)	1.0 (.0, 3.0)	1.0 (.0, 3.0)	1.0 (.0, 7.0)	.5 (.0, 1.5)

[a]Including three patients with total/partial denture, none of them experienced pain (score).
[b]Including three patients with total/partial denture, one of them experienced discomfort (score 1).
[c]Including three patients with total/partial denture, none of them experienced interruption of meals (score 1).
[d]Including three patients with total/partial denture, none of them experienced difficulties to relax (score 0).
[e]Including three patients with total/partial denture, none of them experienced embarrassment (score 0).

tal hygiene regimen that is in line with the national dental hygiene guideline[31]: brushing ≥2 per day, brush ≥2 min per cleaning session, use fluoride toothpaste. PWH scored low on the experienced OHIP-14 questionnaire, which indicates that they barely experience dental limitations, discomfort, disabilities or handicap. The bleeding events, bleeding- and plaque index scores did not differ significantly among patients with different severity of haemophilia. The DMFT score was significantly lower in patients with severe haemophilia, compared to the patients with mild haemophilia, indicating a better dental status in severe haemophilia.

4.1 | Bleeding events

PWH have a bleeding tendency which could lead to bleeding during or after dental procedure. In this study, 52.1% of the PWH reported a bleeding during or after dental procedures. Most events (n = 20) occurred at the maxillofacial surgeon. In the Netherlands, PWH are referred to the maxillofacial surgeon when surgical procedures (e.g., extraction, apex resection or implant placements) are carried out. Although the procedures are carried in consultation with a HTC and patients receive haemostatic treatment (e.g., administration of factor concentration, DDAVP and/or tranexamic acid) and local additional measures (e.g., absorbable haemostatic agents), the procedure in itself carriers a high bleeding risk and leads to a larger wound bed than procedures performed at the regular dentist office. We observed no difference in the number of bleeding events between the various severities of haemophilia. Remarkably more bleeding events are reported by patients with mild to moderate haemophilia than with patients with severe haemophilia. One explanation might be that patients with severe haemophilia always use additional measures with any dental intervention, for instance use their regular prophylaxis just before the intervention and patients with mild haemophilia do not.

4.2 | Preventive dental behaviour

In the Netherlands, the general advice is to clean the teeth twice per day with a toothbrush and fluoride toothpaste for at least 2 min per session and once per day additional interdental cleaning.[31]The results of our study shows similar results in PWH as the general Dutch population. A study in the Netherlands show that the use of daily interdental cleaning was high in the Dutch population reporting an average 37% using dental picks and 22% using floss.[28] The oral hygiene regime of the Dutch PWH is much better than in other countries. A study in New Zealand reported 45% of the patients brushing more than one per day with fluoride toothpaste and no adult patients reported daily interdental cleaning.[8] In India a lower number is reported with 23% of the patients brushing more than once per day.[18] In Turkey brushing once or more per day was already categorised as regular oral care, which was found in 60.5%. During brushing and/or with the use of interdental cleaning products 37% of the dentate patients report gum bleeding.[17] Compared to the Dutch population, this was in line with people with a higher education reporting 38% and distinguishably lower than people with a lower education reporting 53%.[28] In the report of Schuller[28] people with a lower education level had a poor oral health and had limited access to professional oral care. Respondents with a high level of education had the best oral health and the most favourable dental preventive behaviour. This is also observed in our cohort. There are no studies reporting gum bleeding during daily

oral care in adult PWH. In our study, no difference between the severity of haemophilia and reported gum bleeding was found. We observed a positive correlation between the plaque index score and the bleeding index score. This suggests that the accumulation of plaque due to insufficient dental cleaning may result in more gum bleeds. The median plaque index score was around 30% is considered insufficient. A healthy plaque index score should be at least below 20%, preferably below 10%.[25] Lower scores of gingival inflammation were reported in New Zealand with 25.9%[8] and in India with 28%.[18] However in the United Kingdom 46.7% of PWH were diagnosed with chronic periodontitis (gum disease).[29] Professional and personal guidance with regard to oral hygiene should be advised to lower the plaque index score in Dutch PWH. Therefore, patients with a higher plaque- and bleeding index score received oral hygiene instructions at the dental screening in our hospital or were referred to a dental hygienist. This is essential to prevent periodontal disease and caries in these patients. Dental hygiene should be improved to prevent gum disease and caries leading to tooth loss. Promotion of oral health is extremely important to prevent dental diseases and resulting in less dental treatments, which are accompanied by the risk of bleeding.

4.3 | Oral health related- quality of life

In this study PWH scored low (median 1.0) on the experienced OHIP-14 questionnaire which means that they hardly experience dental limitations, discomfort, disabilities or handicap. The Dutch PWH even have a higher OHR-QoL than the general Dutch population.[32,34,35] The opposite finding was reported in a study from Turkey and the United Kingdom where PWH perceived their OHR-QoL to be poorer than those without haemophilia.[8,17] The Dutch PWH scored lower than patients in other countries with a median total score of 3.0 in New Zealand[8] and mean score of 12.7 in Turkey.[17]

4.4 | Dental examination

All dental examinations has been carried by the same dentist in all patients (HV). During the dental examination, the DMFT index, the DPSI, the plaque- and bleeding index score were scored (Appendix 3). An increase in the sum of the number of Decayed, Missing due to caries, and Filled Teeth in the permanent teeth (DMFT) is observed with age and the lowest DMFT values are seen among younger people.[33] The total number of permanent human teeth is 32; thus, the maximum DMFT index is 32, and the minimum value is 0. We observed a difference in DMFT-score between the different levels of haemophilia severity. The patients with severe haemophilia had a lower DMFT-score than patients with mild haemophilia, which indicate less decayed, missing or filled teeth. We found a positive correlation between age and DMFT score. The variation in age between the different levels of haemophilia severity might explain the difference in DMFT score between the levels of haemophilia severity. The same variation are seen in the Dutch population.[28] In India, no DMFT difference was observed between the adult PWH and control group.[17] In New Zealand, an even lower caries experience was seen in PWH than in the New Zealand population.[8] Unfortunately no comparison could be made between the Dutch PWH and the general population for the DMFT index.

4.5 | Limitations

Despite the fact that this is the first and most extensive study in the Netherlands focusing on subjective dental experiences and objective clinical dental status in Dutch PWH, the study has some limitations. We did not include a control group in our study and comparisons between the PWH group and Dutch population are based on existing literature. There was a lower inclusion rate than estimated, due to the COVID-19 pandemic it was not possible to include additional patients. Patients invited for this study were randomly selected from the total haemophilia population in our HTC, the voluntarily participation may lead to selection of patients with a high standard over dental care. A larger cohort, a control group and equal groups of different severities of haemophilia would be advised for further studies.

5 | CONCLUSION

According to our findings, PWH in the Netherlands have a good oral health and vitality. The results of the OHIP questionnaires showed that PWH have a high self-rating oral health quality of life.

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CONFLICT OF INTEREST

G.M. has received speaker's fees from CSL Behring and consultancy fees from NovoNordisk outside the submitted work. F.L. reports unrestricted grants from CSL Behring, Takeda, uniQure and the Dutch Hemophilia Foundation (Stichting Hemophilia) and is a consultant for UniQure, CSL Behing, Takeda and Biomarin for which fees go to the Erasmus MC. All other authors have declared no conflicting interests.

DATA AVAILABILITY STATEMENT

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

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APPENDIX 1: ADDITIONAL QUESTIONNAIRE

General

1. What is your age?
2. What is the highest level of education you have completed?

Medical

3. Which type of haemophilia do you have?
4. How severe is your haemophilia?
5. In addition to haemophilia, do you have any other conditions/illnesses?
6. Do you use other medicines besides coagulation factors?
7. Have you had surgery in the past?
8. Do you have allergies?

9. If yes, which allergies?
10. Do you smoke or did you smoke in the past?
11. If not, are you a passive smoker?
12. If yes,
    - At which age did you start smoking and/or when did you stop?
    - What do or did you smoke and how much?
13. Do you drink alcohol or have you drunk in the past?
14. If yes,
    - At which age did you start drinking and/or when did you stop?
    - What do or did you drink and how much?
15. Do you use drugs or did you use them in the past?
16. If yes,
    - At which age did you start doing drugs and/or when did you stop?
    - What kind of drugs do or did you use and how much?

Dental care

1. Which dental situation is applicable to you?
2. Dentatemaxilla and mandible without partial removable dentures;
3. Partial dentate maxilla and/or mandible with partial removable dentures;
4. Dentate jaw (with or without partial removable dentures) opposed by a edentate jaw (with removable denture);
5. Edentate maxilla and mandible with removable denture without implant support;
6. Edentate maxilla and mandible with removable denture with implant support.
7. Do you have dental insurance?
8. What is the reason you have or do not have dental insurance?
9. Do you have a dentist?
10. If yes,
    - Who is your dentist and where is the practice located?
    - How often do you visit your dentist for a check-up every year ?
11. If not, what is the reason for you not to have a dentist?
12. When was your last visit to the dentist office?
13. Are you being treated or have you been treated by a periodontist, dental hygienist or a (paro)-prevention assistant?
14. Are you being treated or have you been treated by a dental surgeon?
15. Have you ever had a bleed during or after dental treatment?
16. When were the last X-rays taken?

Dental hygiene

1. How often do you brush your teeth?
2. How long do you brush your teeth per cleaning session?
3. What kind of toothbrush do you use (manual, electric or both)?
4. What kind of toothpaste do you use (with or without fluoride)?
5. Do you use interdental cleaners?
6. If yes,
    - What kind of interdental cleaners?
    - How often do you use interdental cleaners?
7. Do your gums bleed while brushing or using interdental cleaners?
8. Do you use a mouthwash?
9. If yes,
    - Why do you use a mouthwash?
    - How often do you use a mouthwash?

Oral complaints

1. Do you have complaints about your teeth?
2. If yes,
    - What are those problems?
    - Are these problems addressed by a dental healthcare professional?
3. Do you have complaints about your gums or oral mucosa?
4. If yes,
    - What are those problems?
    - Are these problems addressed by a dental healthcare professional?
5. Do you have complaints about your chewing muscles or your jaw joints?
6. If yes,
    - What are those problems?
    - Are these problems addressed by a dental healthcare professional?

APPENDIX 2: OHIP-14 QUESTIONNAIRE

Please answer the questions below, and score how often you have suffered from these complaints during the past month. Circle the answer that is most applicable for your situation?

Questions	Response				
Functional limitations	**Never**	**Seldom**	**Sometimes**	**Often**	**Always**
1. Have you had trouble pronouncing any words because of problems with your teeth, mouth or denture?	0	1	2	3	4
2. Have you felt that your sense of taste has worsened because of problems with your teeth, mouth or denture?	0	1	2	3	4
Physical pain					
3. Have you had a painful aching in your mouth?	0	1	2	3	4
4. Have you found it uncomfortable to eat any foods because of problems with your teeth, mouth or denture?	0	1	2	3	4

(Continues)

Questions	Response				
Functional limitations	**Never**	**Seldom**	**Sometimes**	**Often**	**Always**
Psychological discomfort					
5. Have you been self-conscious because of your teeth, mouth or denture?	0	1	2	3	4
6. Have you felt tense because of problems with your teeth, mouth or denture?	0	1	2	3	4
Physical disability					
7. Has your diet been unsatisfactory because of problems with your teeth, mouth or denture?	0	1	2	3	4
8. Have you had to interrupt meals because of problems with your teeth, mouth or denture?	0	1	2	3	4
Psychological disability					
9. Have you found it difficult to relax because of problems with your teeth, mouth or denture?	0	1	2	3	4
10. Have you been a bit embarrassed because of problems with your teeth, mouth or denture?	0	1	2	3	4
Social disability					
11. Have you been a bit irritable with other people because of problems with your teeth, mouth or denture?	0	1	2	3	4
12. Have you had difficulty doing your usual jobs because of problems with your teeth, mouth or denture?	0	1	2	3	4
Social handicap					
13. Have you felt that life in general was less satisfying because of problems with your teeth, mouth or denture?	0	1	2	3	4
14. Have you been totally unable to function because of problems with your teeth, mouth or denture?	0	1	2	3	4

APPENDIX 3: DENTAL EXAMINATION

During the dental examination, the dental status was recorded first;

a. Dentatemaxilla and mandible without partial removable dentures;
   Partial dentate maxilla and/or mandible with partial removable dentures;
   Dentatejaw (with or without partial removable dentures) opposed by a edentatejaw
   Edentate maxilla and mandible with removable denture without implant support;
   Edentate maxilla and mandible with removable denture with implant support.

The dental examination consisted of scoring the soft and hard oral tissues. The visual and tactual exploration of the soft tissues includes lips, upper and lower alveolar ridges, vestibulum oris, tongue, floor of the mouth, retromolar space, hard and soft palate and pharyngeal arcs. If there was an abnormality (e.g., ulceration, abscess, fistula) it was marked as abnormal and was given a description.

The DMFT-index is used on the permanent dentition and is expressed as the total number of teeth (T) that are decayed (D), missing (M), or filled (F) in an individual. The scores can range from 0 to 28 (excluded third molars). The DMFT is calculated as follows. Only visible permanent teeth are counted and listed as T. Primary and secondary caries are marked as D. When a tooth is extracted due to caries, it is listed as M. When a permanent or temporary, direct or indirect restoration is present, this is counted as F. Also the presence and type of removable dentures were recorded for the maxilla and mandible. The number of implants and the type of supra-structure were scored.[25]

The Dutch Periodontal Screening index (DPSI) was used to score the periodontal status of a patient. The highest score per sextant was recorded, ranging from a score 0 to 4. In addition a bleeding index was scored at six different surface locations (mesio-, mid- and disto-buccal and lingual). Per location was scored; 1 (bleeding within 30 s after probing) or 0 (no bleeding). Based on the score, patients were divided into DPSI-category. The Dutch Periodontal Screening Index (DPSI) is the instrument to detect progression, periodic screening of a periodontal disorder. In the mouth; per sextant DPSI score is detected and recorded by mirror and a pocked probe.[25]

A plaque index was scored in the same way as the bleeding index. Recorded per tooth and per surface, with scores 1 (plaque) or 0 (no plaque). For both bleeding- and plaque-index the score is calculated by dividing the total score by the total number of surfaces and given in percentage.

Scoring 0%–10% good, 10%–20% sufficient and > 20% insufficient. Dental x-rays were only indicated when there were problems which needed acute treatment and there were no recent dental x-rays at their regular dentist. If needed, a referral to the appropriate healthcare professional was made. The plaque index is used to evaluate the level and rate of plaque formation on tooth surfaces and to test the efficacy of oral care products for removal and prevention of plaque deposits from these surfaces.[25]

The dental bleeding index is the most common bleeding gingival index used on teeth, the index scores gingival inflammation from 0 to 3 on the facial, lingual and mesial surfaces of all teeth. The symptom of bleeding comprises a score of at least 2.[25] All the different indices were scored by the dentist. Dental x-rays were only indicated when there were dental problems that needed acute treatment and no recent dental x-rays were available. If needed, a referral to the appropriate healthcare professional was made.

APPENDIX 4: PATIENT INTERVIEWS

The nurse practitioner interviewed the patients before dental examination about experience with oral health and (preventive) dental care related to haemophilia.

Historical dimension: refers to patients' dental history and their dental caries experience

HA 2% (63 y) "*I just went to the dentist when I was 18 years old and met my girl-friend. Before that age I never went; my mother did not allow it; a nephew with haemophilia died after dental surgery in the hospital. she was too scared to lose me.*"

HA < .1 (53 y) "*I have dropped dentists in the past. I think that how they were able to relate to me as a person with haemophilia was probably the biggest indicator of whether I felt comfortable with what they were doing.*"

HA severe (76 y) "*All my teeth were removed when I was 21 years old, in a hospital. With one week of treatment with cryo's, I was still bleeding. I was sad to have lost all my teeth. But at that time, it was frequently done with all severe haemophilia patients.*"

Psychosocial dimension: refers to the psychological and social aspects of patients' oral health, including patients' emotional suffering due to dental issues

HA 3% (35 y) "*As long as I can remember, I was and am afraid of the dentist because of bleeding risk and pain. Still I find it important to keep my own teeth and not have false teeth. So I will go to my regular appointments and participate in this study. However, I take medication to relax and need a shower after these examinations, because it is giving me a lot of stress.*"

HA 8% (40 y) "*The knowledge of dentists about bleeding disorders is lacking but it is getting better. As a person with haemophilia you must be aware at all times that we are at risk. We have to educate other health care providers because this is a rare disease*"

Habitual dimension: refers to customary activities related to or consequences of dental caries

HA < .1% (28 y) "*For an appointment with the dentist, I take a prophylactic treatment. Like, with your hematologist, also with your dentist; it is a two-way relationship. So it is the gentleness, it is the trust, it is the respect, it is actually the transparency that has been able to build up a relationship where you can trust your dentist to give you a very open and honest answer about any treatment.*"

HA moderate (35 y) "*My worst habit is probably not cleaning my teeth regularly before I go to bed, well; I reckon I am a bit lazy, I should know better, because I had a lot of bleeding because of periodontitis, and it never stopped bleeding.*"

APPENDIX 5: SPECIFIC RESULTS ON THE DENTAL EXAMINATION

Several abnormalities were found at dental examination. Abnormalities of the soft tissues were related to stomatitis of the palate, a fistula and mouth ulcers (n = 3, 6.3%). In other cases signs of parafunctions were observed (e.g., dental imprints of the buccal mucosa, tongue and/or lips) (n = 4, 8.3%). In one case (2.1%) there was frothy saliva, an indication for a dry mouth.

DMFT score

A total of 11.5 point was given for the total group of PWH for the DMFT index score. For the DMFT-score no different was found between severity groups ($p = .010$). The DMFT was significantly lower in the group with patients with severe haemophilia (median 2.0) compared to mild haemophilia (median 16.0) ($p = .04$). Total DMFT-score and Missing-score correlated with age ($p = .000$) ($p = .000$). The DMFT-score was also different between the different education levels ($p = .040$). The DMFT-score was lower in the group of patients with a higher education (median 18.0) compared to the group of patients with a lower education (median 9.0) ($p = .007$).

DPSI

No difference for brushing time ($p = .291$) type of brush ($p = .896$), brushing frequency ($p = .896$), interdental cleaning ($p = .746$), the use of mouth rinse ($p = .344$) and DPSI ($p = .178$) in comparison to the total bleeding index score. However, there is a trend towards a higher bleeding score with a higher DPSI-category (cat-A 29.6% bleeding, cat-B 34.6% bleeding and cat-C 44.0% bleeding).

Bleeding and Plaque index

Five patients (11.1%) scored 10%–20% (sufficient) and 40 patients (88.9%) scored > 20% (insufficient) on the plaque index. A positive correlation between the plaque and bleeding index was observed $p = .000$), after excluding of two outliers (1 = plaque 72%/bleeding 2%, 2 = plaque 18%/bleeding 61%). Between the different levels of severity there was no difference for the dentals status (X2 (4) = 2.394, $p = .664$), bleeding score ($p = .567$) plaque score (H(2) = 2.002, = .364) and DSPI-category($p = .404$). There was a difference in plaque index the use of different types of brushes (H(2) = 9.317, $p = .009$). The plaque score was lower in patients using a manual brush than compared to an electrical brush (U = 76.500, $p = .011$) or using both (U = 20.500, $p = .006$). We found no association between the duration of brushing time and a lower plaque index (H(2) = .488, $p = .784$), but there is a trend when brushing longer there is more plaque reduction (>2 min 32.5% plaque, 2 min 32.0% plaque and >2 min 26.0% plaque). No difference was observed between brushing frequency (H(3) = 1.718, $p = .633$), interdental cleaning (U = 136.500, $p = .913$) and the use of mouth rinse (U = 157.00, $p = .432$) in comparison to the total plaque index there is no difference for the plaque score compared to the DPSI-index) = 3.736, $p = .154$). However there is a trend, with a higher plaque score the DPSI-category is also higher (cat-A 31.5%, cat-B 41.8% and cat-C 62.0%).

APPENDIX 6: DENTAL INSURANCE AND CARE

Thirty patients had dental insurance (62.5%), 14 patients did not have dental insurance (29.2%) and of four patients it was unknown (8.3%). Of the 30 patients with insurance 29 were dentate and one edentate. The reasons for having dental insurance are very diverse; unforeseen problems (n = 2, 6.7%), just in case (n = 4, 13.3%), problems in the past (n = 7, 23.3%), costs of the insurance is lower than self-payment of treatment (n = 6, 20.0%), importance of good dental care (n = 2, 6.7%) and others (n = 9, 30.0%), which includes habit of having dental insurance, total insurance package including dental insurance, spreading of costs and dental insurance with included coverage for children. There are various reasons for not having dental insurance; costs of the insurance are higher than self-payment of treatment (n = 5, 35.7%), unnecessary due to lack of dental problems (n = 3, 21.4%), financial reasons (n = 3, 21.4%) and others (n = 3, 21.4%, including edentolousness, not eligible for insurance and the fact that surgical procedures carried by a maxillofacial surgeon are included in regular medical insurance). As is recommended in national dental guidelines[31] 93.8% of PWH visit yearly their own dentist. None of the patients reported that a dental caregiver refused treatment because of haemophilia. Forty-one PWH (85.4%) were seen for a dental screening or treatment in the past year.

In the Netherlands, dental insurance is not included in the basic insurance package for adults. People can choose whether they want to pay for additional insurance for dental care. The insurance policy conditions can be very diverse in terms of reimbursement and coverage of the different dental treatments by the different insurance companies. In this study, two-thirds of the dentatePWH have a dental insurance. Most of the reasons for having or not having dental insurance involve financial reasons.[28] None of the reasons are related to haemophilia. PWH in the Netherlands are receive routine dental care and treatment at the regular dentist office. In this study almost all patients have a dentist and a routinely dental check-up (≥1 per year). Before inclusion in the study, more than 80% of the patients were seen for a dental screening or treatment, which is comparable with the average attendance of 88% of the general Dutch population.[28] The attendance number of the haemophilia patients in the Netherlands is much higher in comparison to studies from other countries. A study from Turkey reported 54% of dental visits in the last year.[17] Similar results were reported in New Zealand with a 52% attendance rate in the previous year, however nearly half of the patients only attended when having dental problems.[8] Numbers in the United Kingdom were even lower, with only 13% of haemophilia patients being enrolled with a regular dentist, and most patients only attend a dentist when in pain.[29] In our study none of the patients reported that a dental caregiver refused treatment because of their haemophilia. A study from New Zealand shows that 20% of the patients did experience a dentist refusing care because of their bleeding disorder. In the United States 41% of the haemophilia treatment centers (HTCs) reported that there is a lack of dentists willing to treat patients with a bleeding disorder, causing a significant barrier to access dental care.[7] Guidelines of the World Federation of Haemophilia (WFH) recommend a dentist should be part of the multidisciplinary haemophilia team, and this expertise would contribute to accurate risk assessment. Based on the expert opinion, it seems in the Netherlands that regular dental care could be provided by a regular dentist practice, with the exception of surgical procedures such as dental extractions.[30] PWH are well educated and know when additional measures need to be taken for certain dental procedures. HTCs are easily approachable for consultation by a patient or dentist. However, a tool to assess the invasiveness of the dental procedure against the severity of haemophilia, such as the Dental Bleeding Risk Assessment and Treatment Tool (DeBRATT), could help in a risk-based approach.[29]