

SHORT THESIS FOR THE DEGREE OF DOCTOR OF PHILOSOPHY (PhD)

Dental care for patients with haemophilia - experiences and attitudes from the perspective of patients and dentists

by Dr. Kunkáné Dr. Kitti Sipos

Supervisor: Prof. Dr. Csongor Kiss
Prof. Dr. Attila Csaba Nagy



UNIVERSITY OF DEBRECEN
DOCTORAL SCHOOL OF MEDICAL SCIENCES

DEBRECEN, 2026

Dental care for patients with haemophilia - experiences and attitudes from the perspective of patients and dentists

By Dr. Kunkáné Dr. Kitti Sipos, DMD

Supervisor: Prof. Dr. Csongor Kiss, MD, PhD, DSc
Prof. Dr. Attila Csaba Nagy, MD, PhD

Doctoral School of Medical Sciences, University of Debrecen

Head of the **Defense Committee:**

Prof. Dr. Miklós Udvardy, MD, DSc

Reviewers:

Dr. Imelda Marton, MD, PhD

Dr. Zsolt Csaba Oláh, MD, PhD

Members of the Defense Committee:

Dr. Ferenc Vincze, PhD

Dr. Botond Barna Simon, DMD, PhD

The PhD Defense takes place at the Lecture Hall of Bldg. A, Department of Internal Medicine, Faculty of Medicine, University of Debrecen, at 1:30 p.m. on September 2, 2026.

1. Introduction

Coagulation factor deficiencies represent the most prevalent hereditary, congenital forms of bleeding disorders, with haemophilia A, haemophilia B, and von Willebrand disease accounting for the vast majority of cases (95-97%). The daily lives of patients with haemophilia (PWH) are characterised by numerous difficulties, among which access to dental care deserves special attention. As this is a rare disorder, dentists rarely encounter this patient group, which results in a lack of confidence in their knowledge and treatment experience. Consequently, the provision of dental care for these patients is frequently compromised. This issue is further compounded by patients' fear of dentists and dental procedures, as well as the fear of bleeding during treatment. Consequently, some patients only seek dental care when they experience a complaint, often when invasive treatment has already become imperative. This issue is further complicated by the high rejection rate experienced in the care of PWH, a finding that is also confirmed by the results of our study. The consequences of delayed care, discrimination and rejection due to underlying conditions and possible infectious diseases can be significant, potentially leading to a marked deterioration in patients' quality of life and causing psychological distress. Maintaining optimal oral hygiene and health is of paramount importance for patients with bleeding disorders, as it can prevent the necessity for invasive dental procedures that are associated with bleeding risk. This type of care extends beyond the scope of primary care and can only be provided in a specialist facility after appropriate precautions. Nevertheless, the significance of primary care must be emphasised, as dentists working at this level of care are the first and usually the fastest point of contact for patients. Within this framework, general dental practices (GDPs) financed by the National Health Insurance Fund of Hungary are the only providers of free primary care in the Hungarian insurance system. Despite the fact that patients suffering from bleeding disorders are regarded as high-risk in the context of dental care, the majority of routine, non-surgical dental

procedures can be conducted in primary dental care, provided that professional protocols are adhered to and a haematologist is consulted. This ensures that patients have expeditious and convenient access to dental services.

2. Objectives

The objective of the study was to conduct a questionnaire survey exploring the situation of dental care for PWH, which was analysed from the perspective of both patients and dentists:

1. **The experiences and opinions of PWH regarding dental care (henceforth referred to as the Patient Questionnaire)** - The objective of the present study was to explore the experiences of PWH regarding their access to dental care. Moreover, data was collated concerning the impact of the coronavirus type-2 (CoV-2) disease (COVID-19) on access to dental treatment in this particular patient group.
2. **Dental care for PWH from the perspective of dentists (henceforth referred to as the Dentist Questionnaire)** - In view of the fact that the majority of previous studies concentrated on the patients' perspective, the aim of this research was to ascertain the opinions, experiences, and treatment practices of Hungarian dentists.

The objective of this study was to obtain a comprehensive understanding of the dental care situation for PWH by comparing data from the perspectives of patients and dentists, and to identify possible directions for improving and optimising care.

In both studies, particular emphasis was placed on primary care dentistry, as many procedures do not require a higher level of care following appropriate protocols and consultation with a haematologist.

It is hoped that the findings of this research will contribute to the simplification of dental care for PWH in the future, and that it will provide guidance for the development of safe and collaborative dental treatments. The dissertation is based on publications which address an absence in both the region and in the literature.

3. Methods

3.1. Patient Questionnaire

3.1.1. Study design

The study was performed between July and December 2022 in four major Hungarian haemophilia treatment centres (HTCs):

- **Clinical Centre of the University of Debrecen** - Department of Internal Medicine, Division of Haematology (Internal Medicine Clinic, Building B) and Department of Paediatrics, Paediatric Haematology, Debrecen
- **Central Hospital of Northern Pest** - Medical Centre of the Hungarian Defence Forces, National Haemophilia Centre and Haemostasis Department, Budapest
- **Heim Pal Children's Hospital**, Budapest
- **University of Pécs Clinical Centre, Mohács Hospital**, Mohács

We used a self-administered anonymous questionnaire developed for this study based on literature review and expert opinion of the senior dentist and haematologist (Prof. Dr. Ildikó Márton and Prof. Dr. Csongor Kiss). A pilot test of the questionnaire was carried out with 10 PWH (eight adults, two children) from the Debrecen centre. Children under 16 years of age completed the questionnaire with the help of their guardians.

The inclusion criteria for this study are as follows: patients diagnosed with haemophilia A or B, of any severity, and female subjects with low FVIII/IX levels (FVIII/IX activity levels of less than 50%). The following were considered to be exclusion criteria: the presence of an additional haemostatic disorder, the presence of severe cognitive impairment, the absence of signed written informed consent and the participation of the subject in the pilot study for the development of the study questionnaire. Participation in the study was voluntary. Ethical

approval was obtained (Regional and Institutional Ethics Committee, Clinical Centre, University of Debrecen; No. DE RKEB/IKEB: 6087-2022).

3.1.2. Sample population and data collection

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 0.05. During data collection period, 80 patients were invited, and 68 patients enrolled (response rate: 85%). The questionnaires were completed in HTC (n=63; 93%) or online via a Microsoft Forms link (n=5; 7%).

3.1.3. Variable specification

The questionnaire contained 30 questions and was divided into four sections. With regard to the level of dental care, the statistical analysis was based on the Hungarian healthcare system, and three age groups were delineated (ages 0-18, ages 19-44 and ages >45). Haemophilia severity defined conventionally by factor VIII and IX levels. Patients with severe or moderate haemophilia and those with mild haemophilia were assessed separately. Inhibitors were categorized conventionally by Bethesda titer (BU/mL).

3.1.4. 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 (machine learning 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. A p-value <0.05 was considered significant.

3.2. Dentist Questionnaire

3.2.1. Study design

The study was conducted between September and December 2022 among dentists working in the primary care sector in Hungary. The selection criteria were specifically designed to include only dentists working at primary care level in Hungary. This category includes dentists working in GDPs registered with the National Health Insurance Fund of Hungary and private practices.

Data were collected via an anonymous self-administered questionnaire based on literature data and expert opinion provided by the senior dentist and a haematologist co-authors (Prof. Dr. Ildikó Márton and Prof. Dr. Csongor Kiss). A pilot test of the questionnaire was conducted with a sample of 30 dentists employed at the University of Debrecen, Faculty of Dentistry, who were responsible for providing advanced level care. The present study excluded dentists who exclusively worked at a higher level of care (i.e. in a specialist institution or care centre); those who participated in the pilot study to develop the survey questionnaire; and dentists who did not answer the question on informed consent. Ethical approval was obtained (Regional and Institutional Ethics Committee, Clinical Centre, University of Debrecen; No. DE RKEB/IKEB: 6087-2022).

3.2.2. Sample population and data collection

Participants were selected from a list of GDPs from the website of the National Health Insurance Fund of Hungary in September 2022.

Priori sample size calculation indicated that a minimum of 158 subjects were required for the study, with a power level of 80% and an alpha level of 0.05 using proportions (25.81% and 49.07%).

Selected dentists were invited to participate in the survey via email. All participants provided informed consent by affirmative response given to the first question of the questionnaire. Participation was voluntary.

Of the 163 randomly selected dentists 47 returned the questionnaire (response rate: 28%). In consideration of the low rate of participation, an invitation to contribute was extended to private practice dentists (n=115) who were affiliated with the University of Debrecen, specifically those engaged in the practical graduate dental student training programs and/or regularly participating in the postgraduate training programs organized by the University of Debrecen. Questionnaires were sent to dentists who had replied positively to the invitation.

A total of 162 dentists provided consent by completing the questionnaire, which was either submitted online via a Microsoft Forms link (n=114; 70%) or returned by post (n=48; 30%).

3.2.3. Variable specifications

The questionnaire contained 21 questions and was divided into four domains.

The statistical analysis considered four academic titles, i.e. Doctor of Dental Medicine degree (DMD), Doctor of Dental Surgery degree (DDS), Doctor of Philosophy degree (PhD), and Doctor of Science of the Hungarian Academy of Sciences degree (DSc). According to the Hungarian Specialist Training Program, three groups of professional experience were defined (<2 years: Resident Doctor; 2-5 years: Specialist Candidate and Young Specialist; >5 years:

Senior Specialist). This study investigated three major regions of Hungary (Great Plains and North Hungary; Central Hungary (the capital city of Budapest + Pest County) and Transdanubia).

3.2.4. Variable selection and statistical analysis

Categorical variables were compared using Pearson's Chi-squared test and Fisher's exact test. Multiple logistic regression model was built to investigate the factors influencing the dentists' self-reported confidence in providing dental care to PWH. Findings from the logistic regression analyses were represented as ORs and 95% CIs. Statistical evaluations were executed using STATA IC Version 18.0 software. A p-value <0.05 was considered significant.

4. Novel scientific results

4.1. The present study is the first to provide comprehensive data on dental care for

PWH in the continental European context.

To date, the extant literature concerning the experiences of dental care for PWH and the current state of care is limited. The paucity of research conducted on this topic in Europe is evident, with only two studies published in the United Kingdom in 2000 and 2011 specifically addressing it. Consequently, a comprehensive study was not conducted for a considerable period, and thus the two publications that form the basis of this dissertation offer novel insights regarding the continent.

4.2. From a dental perspective, the research also constitutes the first publication to

provide data on the treatment of PWH on the continent.

Previously, there was only one publication in the literature among dentists that did not focus on knowledge assessment. Rather, it examined the practical situation of patient care, involving dentists' perspectives in the evaluation of the care of patients with congenital bleeding disorders. The present study exclusively focused on patients diagnosed with haemophilia, thereby providing data that addresses a significant gap in the literature on this subject.

4.3. The present study found that the frequency of visits to the dentist's office is

significantly related to the severity of haemophilia and age of patients.

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 ($p=0.026$). Furthermore, age had a significant effect on the

frequency of dental visits ($p=0.033$). The data revealed a markedly higher frequency of dental visits among individuals within the 0-18 age group (51.35%).

4.4. The findings of this study indicate that regular dental attendance and participation in oral hygiene counselling are significant factors determining the frequency of dental visits.

The findings of the multiple logistic regression model based on LASSO selection indicate that participants having a permanent dentist had higher odds of visiting a dental practice (OR: 9.95, 95% CI: 2.86-34.62). Furthermore, patients who had previously attended an oral hygiene consultation had higher odds of visiting a dental office (OR: 3.84, 95% CI: 1.09-13.58).

4.5. In the course of our research, we identified and analysed the barriers to dental care access experienced by PWH. Furthermore, our investigation was the first to assess the impact of the COVID-19 pandemic on dental care for this cohort of patients.

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. The findings of this study indicate that a similar factor hindering care may also significantly influence the availability and quality of dental care for PWH in the future.

4.6. The study revealed a high prevalence of dental care refusal (21%), particularly in primary care (86% in primary dental care financed by the National Health Insurance Fund of Hungary).

The present study is distinguished from previous research by its focus on this level of care, a factor that had not been given significant consideration in previous studies. A higher refusal rate was observed among older patients ($p=0.020$) and in the presence of comorbidities ($p=0.006$).

4.7. The findings of this study indicate that infectious diseases, bleeding, or swelling following dental procedures are associated with an increased risk of patient rejection.

The findings of the multiple logistic regression model based on LASSO selection indicate that those patients who have or ever had an infectious disease had higher odds for refusal (OR: 4.48, 95% CI: 1.14-17.69). Patients who ever experienced bleeding or swelling after dental procedures were also more likely to experience refusal (OR: 4.23; 95% CI: 1.10-16.27).

4.8. A significant discrepancy was identified between the assessments of satisfied and dissatisfied patients with regard to the dental care options available to PWH.

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 ($p<0.001$) in the refusal by dental care-providers between satisfied ($n=47$) and dissatisfied patients ($n=7$). All dissatisfied patients were refused.

4.9. The results of the study demonstrated that participation in oral hygiene consultations was associated with an increased level of satisfaction with dental care options among PWH.

The findings of the multiple logistic regression model based on LASSO selection indicate that 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: 0.71-55.88). 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: 0.60, 95% CI: 0.04-8.94, OR: 0.07, 95% CI: 0.01-0.89 and OR: 0.13, 95% CI: 0.02-1.03, respectively).

4.10. The present study found that the majority of dentists do not consider themselves to be confident or adequately informed about providing primary care to PWH.

The majority (n=113; 70%) of surveyed dentists indicated that they lacked confidence in treating PWH due to a perceived lack of knowledge.

4.11. The study found that male gender, experience in treating PWH and additional qualifications (PhD or DSc) beyond a basic dental degree (DMD/DDS) significantly influenced confidence.

A multiple logistic regression model was developed to examine the effect of factors on self-reported confidence in providing dental care to PWH. The model identified three significant factors among investigated variables used in Pearson's Chi-squared test analysis. Male dentists (OR: 3.43, 95% CI: 1.49-7.90), respondents with treatment experience (OR: 4.06, 95% CI: 1.73-9.52), and those with additional qualifications

beyond basic dental degrees (OR: 9.40, 95% CI: 1.10-80.48) were more likely to report confidence in providing dental care to PWH.

4.12. The results of the study indicated that dentists with a minimum of five years of experience were more likely to refer PWH to a higher level of care.

Fisher's exact test was employed to examine the correlation between the years of professional experience and the referral to advanced dental care.

The findings indicated a tendency towards a higher rate of referrals for each intervention among those practicing for over five years (n=113, 70%), when compared to younger colleagues (<2 years, and 2-5 years professional experience groups, n=49, 30%). Among various interventions, the context of the preparation and impression taking for fixed dentures was identified as a significant factor, with 19% of respondents in practice for >5 years indicating that they would refer their patients for higher-level care (p=0.040).

4.13. The findings of this study indicate that dentists with less than two years of professional experience are significantly more likely to rely on their previous undergraduate studies and books, and to seek professional help from senior dentists or oral surgeons, compared to colleagues with more than five years of experience.

Correlation between years of professional experience and orientation sources of dentists was examined by Pearson's Chi-squared test. We found a statistically significant correlation between professional experience and the utilization of previous undergraduate studies and books (p<0.001). Professionals with >5 years of experience were significantly less likely to rely on these resources, whereas those with <2 years of experience were more likely to utilize them. Moreover, a statistically significant correlation exists between

professional experience and the probability of obtaining assistance from dental and oral surgery colleagues. Specifically, professionals with <2 years of experience were more likely to seek assistance, whereas those with >5 years of experience were less likely to accept help ($p=0.015$).

5. Summary

PWH frequently encounter difficulties in accessing dental care. Consequently, their access to dental treatment merits particular consideration and attention. It is of particular significance to emphasise the importance of prevention and regular dental check-ups in this patient group, highlighting the active role of dentists working in primary care.

The findings of the present survey demonstrate that primary dental care performed by GDPs is currently the least preferred option among patients. In this context, it is essential to emphasise the role of undergraduate and postgraduate training, and to establish professional guidelines. These measures have the potential to enhance the confidence and preparedness of dentists.

In the context of the barriers affecting care, this study was the first to assess the impact of the COVID-19 pandemic on access to dental care for PWH, which significantly increased the difficulties experienced by this patient group. This finding indicates that similar future events may have a significant impact on the already complex care processes for this patient group.

The English-language publications of the dissertation aim to draw international attention to the importance of dental care in this cohort of patients, while also contributing to the increase in the relatively small number of publications on the subject to date. The Hungarian-language literature review can provide useful guidance for Hungarian dentists and professionals involved in the care of PWH, helping to make care more effective.

It is expected that, in addition to adhering to professional guidelines and protocols, the enhanced involvement of primary care dentists in the dental care of PWH will contribute to the enhancement of the quality of care provided to patients and to the alleviation of the current treatment difficulties.

6. List of publications



UNIVERSITY of
DEBRECEN

UNIVERSITY AND NATIONAL LIBRARY
UNIVERSITY OF DEBRECEN

H-4002 Egyetem tér 1, Debrecen

Phone: +3652/410-443, email: publikaciok@lib.unideb.hu

Registry number:
Subject:

DEENK/60/2026.PL
PhD Publication List

Candidate: Kitti Sipos

Doctoral School: Doctoral School of Medical Sciences

List of publications related to the dissertation

1. **Sipos, K.**, Márton, I., Móré, M., Nagy, A. C., Kiss, C.: Assessing the factors affecting the accessibility of primary dental care for people with haemophilia.
Haemophilia. 31 (1), 69-77, 2025.
DOI: <http://dx.doi.org/10.1111/hae.15124>
IF: 3 (2024)
2. **Sipos, K.**, Márton, I., Móré, M., Nagy, A. C., Kiss, C.: The attitudes and treatment practices of Hungarian primary care dentists regarding dental care for patients with haemophilia.
Sci Rep. 15 (1), 1-10, 2025.
DOI: <http://dx.doi.org/10.1038/s41598-025-11818-w>
IF: 3.9 (2024)





List of other publications

3. Ghanem, A. S., **Sipos, K.**, Tóth, Á., Nagy, A. C.: Inflammatory Biomarkers and Oral Health Disorders as Predictors of Head and Neck Cancer: a Retrospective Longitudinal Study.
Int. J. Mol. Sci. 26 (5), 1-20, 2025.
DOI: <http://dx.doi.org/10.3390/ijms26052279>
IF: 4.9 (2024)
4. **Sipos, K.**: Haemophiliás betegek fogorvosi ellátása.
Orv. hetil. 163 (24), 943-951, 2022.
DOI: <http://dx.doi.org/10.1556/650.2022.32491>
IF: 0.6

Total IF of journals (all publications): 12,4

Total IF of journals (publications related to the dissertation): 6,9

The Candidate's publication data submitted to the Tudóstér have been validated by DEENK on the basis of the Journal Citation Report (Impact Factor) database.

13 February, 2026



7. Acknowledgements

I would like to express my gratitude to my supervisors, Dr. Csongor Kiss and Dr. Attila Csaba Nagy, who provided professional assistance in data collection, work planning and technical implementation, evaluation of results, and preparation of the dissertation.

In addition to the provision of professional assistance, gratitude is extended for the kindness and wisdom demonstrated, which served as a consistent source of motivation during the preparation of work.

I would like to express my deepest gratitude to my former faculty and department head and my former supervisor, Professor Ildikó Márton, who provided invaluable support and guidance throughout my academic interests over many years, and who assisted me in preparing my publications and subsequently my dissertation. I would like to express my sincere gratitude to Professor Márton for her encouraging words, valuable comments, and constructive advice.

I would like to express gratitude to Dr. Kinga Ágnes Bágyi, Dean of the Faculty of Dentistry at the University of Debrecen, Clinic Director and Head of Department, for supporting my research efforts and providing me with the opportunity to continue my work and prepare my publications and dissertation.

I would like to express my gratitude to Dr. Ágota Schlamadinger and Dr. István Szegedi for the opportunity to participate in adult and paediatric haematology clinics. This provided insight into the care of PWH and the opportunity to personally complete questionnaires. I am grateful for the patience and guidance provided by them, who, despite my lack of experience in this field, provided invaluable advice that proved to be invaluable.

I would like to express my profound gratitude for their assistance in completing the questionnaires to the following contributors: Dr. László Nemes (MD - National Haemophilia Centre and Haemostasis Department, Medical Centre of the Hungarian Defence Forces,

Budapest), Dr. Marianna Zombori (MD - Heim Pal Children's Hospital, Budapest), Dr. Mária Kardos (MD, PhD - Mohács Hospital, Mohács), Prof. Dr. Katalin Nagy (DMD, PhD, DSc - University of Szeged, Faculty of Dentistry; Szeged), Prof. Dr. József Szalma (DMD, PhD, DSc - University of Pécs, Medical School, Department of Oral and Maxillofacial Surgery; Pécs), Dr. Péter Váradi (DMD, PhD; Hévíz) and Dr. Péter Rák (DMD; Sopron).

In addition, I would like to express my gratitude to Dr. Marianna Móri and my co-authors, who provided invaluable support and collaboration, thereby facilitating continuous development in my research.

I am appreciative and thankful to my family and friends for their love and support.