ORIGINAL ARTICLE *Clinical haemophilia*

Oral health in children and adolescents with haemophilia

L. M. EVANGELISTA,* C. C. B. LIMA,* R. C. L. IDALINO,† M. D. M. LIMA‡ and
L. F. A. D. MOURA‡

Federal University of Piauí; †Informatic and Statistic Department, Federal University of Piauí; and ‡Postgraduate Program in Dentistry and Postgraduate Program in Health Sciences, Federal University of Piauí, Teresina, Piauí, Brazil

Introduction: Bleeding disorders are certainly one of the most difficult health conditions among health professionals. The haemophilia requires special care in dentistry mainly due to highly vascularized oral cavity. **Aim:** The objective of this study was to determine oral health conditions of children and adolescents with haemophilia at a haematology centre. **Methods:** The sample was composed of 40 children and adolescents aged 1–18 years. Data collection occurred by means of a questionnaire about social and economical conditions, and an examination of the oral cavity. The decayed, missing and filled deciduous teeth (dmft), decayed, missing and filled permanent teeth (DMFT) and gingival bleeding (allows the determination of gingival bleeding) epidemiological indexes were evaluated. **Results:** When evaluating caries experience, the mean value for the dmft index was 3.4 for the deciduous dentition, DMFT score of 0.9 for mixed-dentition, and 2.9 for permanent dentition. The mean value for the gingival bleeding index was similar in all three phases of development, showing a mean value of 1.74. **Conclusion:** The majority of the children and adolescents demonstrated unsatisfactory oral health.

Keywords: epidemiology, oral health, paediatric dentistry

Introduction

Haemophilia is an X-linked recessive hereditary haemorrhage disorder that affects mainly individuals of the male gender. The alteration is characterized by a deficiency in the blood coagulation mechanism that promotes a greater predisposition to haemorrhage [1,2]. Haemophilia is classified according to deficiencies and percentages of blood coagulation factors, being considered type A when factor VIII is affected and type B when FIX is affected. It is designated as severe when the individual possesses a coagulation factor of <1%, moderate between 1% and 5% and mild if it maintains between 5% and 30% [3].

The prevalence of haemophilia is estimated at 1:10 000 live births [4]. There are around 257 182 people with haemophilia in the world and Brazil occupies the third position in the number of patients with the alteration ($n = 10\ 065$), after the United States (17.485) and India (13 993) [5].

Due to the fact that the oral cavity is highly vascularized and that the execution of procedures can initiate bleeding, it is important for dental surgeons to have knowledge about haemophilia and the problems that dental treatments, if not properly conducted, can cause in patients that possess this alteration [6–9].

The literature lacks publications that approach questions in reference to the oral heath of patients with haemophilia. The present study has the objective of determining the oral conditions of children and adolescents patients with haemophilia that frequent a haematology centre.

Methods

The Research Ethics Committee of the Federal University of Piauí (UFPI) approved the present study under the reference number CAAE 0325.0.045.045-11. The legal guardians of the children and adolescents signed an Informed Consent Term, obeying the Declaration of Helsinki guidelines and norms that regulate research in human beings in the National Health Council Resolution no. 196/96 [10].

This study is of transversal observational nature, whose sample was composed of children and adolescents with haemophilia, aged 1–18 years, that frequented the Hematology and Hemotherapy Center of

Correspondence: Lidiane de Morais Evangelista, MSc, Federal University of Piauí, Av. Frei Serafim, 2280, Centro, Teresina, Piauí, CEP 64001-020, Brazil.
Tel.: +(86) 9927 4601; fax: + (86) 3215-1148;
e-mail: lidianemorais25@yahoo.com.br

Piauí (HEMOPI) consecutively from October, 2011, to June, 2012. Children/adolescents that were considered eligible for the study were those who were registered in the 'Hemovida Web Coagulopatia' data bank (National Registry of Coagulopathy patients), registered in the state of Piauí. Of the 66 patients registered, 40 were included in the study, by attending the HEMOPI during the period of the research.

The collection of the data coincided with the individuals' ambulatory visits and occurred at two moments: (i) application of the questionnaire with the guardians and (ii) clinical examination of the oral cavity. The questionnaire was elaborated specifically for the study, based on scientific literature [2,3,5,6,8,11] and adapting issues already assessed in these studies, and contained questions about social, economical and demographical aspects, as well as questions about oral health habits (daily brushing frequency, ingestion of fermentable carbohydrates, time and reason for the last dental visit, among others).

In order to make the necessary adjustments for better understanding of the issues addressed by parents/guardians, the questionnaire was previously tested with 10 parents/guardians whose children did not participate in the study.

The clinical exams were performed in conventional dental clinics at the HEMOPI by one trained and calibrated examiner. The calibration was performed in two stages. The first was theoretical in which epidemiological indexes were presented and discussed, followed by study of the clinical form and exam routine. The second stage was practical with clinical exams in patients of the paediatric clinic of the UFPI that were of the same age group as that of the sample. Individual protection equipment, such as masks, safety glasses, bouffant caps and gloves, were used during the clinical procedures. Oral health was determined by dental and periodontal exams, since none of the patients examined showed other clinically diagnosed alterations.

Before beginning the clinical exams, the patients and guardians received orientation about the health/disease process and methods for prevention and and/or control of biofilm-induced oral conditions. Each subject received fluoridated toothpaste, a toothbrush and dental floss and the teeth were brushed under the supervision of the examiner. The exams were performed in conventional dental clinics with the aid of a dental mirror, a blunt-tip dental probe, dental light and dry field.

To guarantee the reliability and reproducibility of the data and to validate the diagnostic criteria for dental caries, the exams were repeated in 10% of the sample evaluated 20 days after the first exam. The intra-examiner diagnostic agreement obtained a kappa value of 0.89.

To evaluate the periodontal conditions, the gingiva was slightly dried with compressed air and a World Health Organization (WHO) recommended periodontal probe was used. The probe was passed along the entire gingival margin (vestibular, lingual, mesial and distal), at a mean depth of 1 mm with the probe positioned at 60° to the long axis of the tooth, with the intention of avoiding mechanical trauma to the gingival tissue. The presence or absence of gingival bleeding was evaluated 30 s after inspection of the area [11].

The scores used to determine the presence of bleeding were: 0 – absence of gingival bleeding; 1 – presence of gingival bleeding. According to the quantity of gingival bleeding points, the individuals were categorized as: (i) Gingival bleeding index (GBI) = 0: absence of gingival bleeding; (ii) Mild GBI: when 1–4 points were present; (iii) Moderate GBI: when 5–9 points were present and (iv) Severe GBI: 10 or more points present [11].

To determine the dental health conditions, the epidemiological indexes dmft (decayed, missing and filled teeth) and DMFT (decayed, missing and filled teeth) were applied, which made quantitative expressions of caries prevalence in the deciduous and permanent dentitions, respectively.

The data were tabulated in the Microsoft Excel (2010) to facilitate organization of the collected information and analyses were performed using the 'R' statistic software, version 2.15.1 [12]. To verify the correlations among the variables the chi-squared test and Fisher's exact test were used to relate qualitative variables and the Mann–Whitney U test for the quantitative variables. A 5% level of significance was used for all of the tests.

Results

In the sample evaluated, 85% of the individuals had type A haemophilia and 15% had type B. In relation to the severity, 42.5% of the patients had mild haemophilia, 37.5% had severe, 10% had moderate and 10% still had not performed the disease severity test.

The information on social, economical, demographical and oral hygiene habits is described in Tables 1 and 2. Tables 3 and 4 depict caries experience and the GBI. The association among the variables of the study can be observed in Table 5. It is important to note that due to the young age of two of the subjects, it was not possible to perform the test with these children in the 1–5 year age group.

Discussion

The HEMOPI group offers interdisciplinary treatment among the diverse health sciences fields to patients with coagulation disorders. The implementation of the 'Hemovida Web' coagulopathies system represents a great advancement in the management of coagulopathy

Table 1. Social and demographical characteristics of children and adolescents with haemophilia (*n* = 40).

	n	%
Age group		
1–5	09	22.5
6–10	16	40.0
11–18	15	37.7
Total	40	100.0
Mother's education (years of formal study)		
<3	02	5.0
4–7	18	45.0
8–11	18	45.0
University	02	5.0
Total	40	100.0
Family income (Brazilian minimum wages)		
<1	13	32.5
1–2	25	62.5
3–5	02	5.0
Total	40	100.0

Table 2. Oral hygiene habits of children and adolescents with haemophilia (*n* = 40).

	n	%
Daily brushing frequency		
>1	30	75.0
1	10	25.0
Does he/she ingest sweets between meals?		
Yes	27	67.5
No	07	17.5
Sometimes	06	15.0
Has he/she ever had a toothache?		
Yes	27	67.5
No	13	32.5
Do his/her gums bleed when brushing?		
Yes	07	17.5
No	22	55.0
Sometimes	11	27.5
Last visit to the dentist (months)		
>6	11	27.5
<6	23	57.5
Never	06	15.0
Purpose of last dental visit		
Preventative procedures	12	30.0
Restorative procedures	08	20.0
Urgency	08	20.0
Other	06	15.0
Never been to the dentist	06	15.0
Presence of dental caries (self-evaluation)		
Yes	21	52.5
No	19	47.5
How would you grade his/her oral health?		
Good	17	42.5
Poor	11	27.5
Fair	12	30.0

patients in Brazil, since the absence of a standardized data bank is one of the factors that contributed to the sub-notification of haemophilia cases in the whole world [13].

The prevalence of type A haemophilia in the subjects evaluated collaborates with the researched literature, indicating that a deficiency of coagulation FVIII is prevalent in relation to deficiency of coagulation FIX [4,14,15].

Low family income and low maternal education level exhibit direct influence on dietary patterns, proportioning poor dietary habits that provoke increase in caries experience [16]. In this study, a low number of mothers with an education level >11 years of study was observed (Table 1). This fact could be attributed to the abandonment of studies for the constant care required by their children, apart from the reduced family income. However, information that reinforces this hypothesis was not investigated by other studies with haemophilia patients.

The motivation of patients in relation to oral hygiene habits and control of fermentable carbohydrate consumption is a basic procedure and essential in oral health promotion. In patients with haemophilia, this motivation becomes crucial and must be accompanied by professional monitoring seeking to prevent the installation of gingivitis or other dental problems that could cause bleeding [9,14,17].

The fact that a low percentage of children and adolescents had never been to the dentist (Table 2) leads to the reflection on the importance of oral health habits for these individuals and their families. Patients that have systemic disorders, like haemophilia, are generally spared by their family members from having to perform certain procedures that are not to their liking, and oral hygiene often falls into this category [18,19].

When evaluating caries experience, the majority of children in the 1–5 year age group did not have caries, although the dmft index was higher than that of the mean dmft of healthy Brazilian children and North-eastern-Brazilian children [20]. Some of the individuals evaluated showed multiple caries lesions in the deciduous dentition (Table 4), although not characterizing polarization of the disease. In this manner, this study does not confirm a lower caries experience in the deciduous dentition of haemophilia patients in comparison to the general population [21].

A reduced number of children and adolescents in the deciduous and mixed dentitions were free of caries (Table 4). This differs from studies with haemophilia patients in which significant values of patients without caries were observed [3,22]. Children in the first decade of life show a reduced number of permanent teeth, a condition that can justify low DMFT values [23]. In this sense, an increase in the values of the index in the 11–18 year age group can be noticed, remaining above the goal set by the WHO for 2010 (DMFT <1.0 at age 12 years).

In this study, the decayed component of the dmft and DMFT indexes predominated (Table 4). This is similar to results from studies with haemophilia patients [24–26] and higher than those encountered in patients without haemophilia [23,27,28]. This situation verifies the association between dental pain and caries experience (Table 5) in which multiple

Table 3. Gingival bleeding index (GBI) of children and adolescents with haemophilia (*n* = 40).

Age group	Frequency N	Frequency %	GBI Mean ± SD	Difference P < 0.05	GBI = 0 N	GBI = 0 %	Mild GBI N	Mild GBI %	Moderate GBI N	Moderate GBI %
1–5	07	18.4	1.14 ± 0.37	b	06	46.2	01	4.5	–	–
6–10	16	42.1	1.87 ± 0.61	a	04	30.8	10	45.5	02	66.7
11–18	15	39.5	1.86 ± 0.51	a	03	23.0	11	50.0	01	33.3
Total	38	100.0	1.74 ± 0.60	–	13	100	22	100	03	100

Mann–Whitney *U* test. SD, standard deviation.
a and b – Same letters have no statistical difference.

Table 4. The dmft and DMFT indexes of children and adolescents with haemophilia (*n* = 40).

Age group	N	dmft = 0 N	dmft = 0 %*	dmft Mean ± SD	d	m	f	DMFT = 0 N	DMFT = 0 %*	dmft Mean ± SD	D	M	F
1–5	09	05	55.5	3.4 ± 5.57	29	–	02	–	–	–	–	–	–
6–10	16	02	12.5	5.8 ± 3.78	79	08	06	08	61.5	0.9 ± 1.50	11	–	01
11–18	15	–	–	–	–	–	–	04	26.7	2.9 ± 2.65	20	05	19
Total	40	07	17.5	4.77	108	08	08	12	30	2.0	31	05	20

SD, standard deviation.
*% in relation to the number of children examined according to age group.

Table 5. Association between caries experience and gingival bleeding and demographic and behavioural variables.

	Caries experience Negative	Caries experience Positive	P-value	Gingival bleeding Negative	Gingival bleeding Positive	P-value
Mother's education						
<3	–	02 (6.8%)	0.112[†]	–	02 (8.0%)	0.072[†]
4–7	05 (45.4%)	13 (44.9%)		04 (30.8%)	12 (48%)	
8–11	04 (36.4%)	14 (48.3%)		08 (61.5%)	10 (40%)	
University	02 (18.2%)	–		01 (7.7%)	01 (4.0%)	
Family income (number of minimum wage salaries)						
<1	02 (18.2%)	11 (38%)	0.511[†]	03 (23.1%)	09 (36%)	0.322[†]
1–2	08 (72.7%)	17 (58.6%)		08 (61.5%)	16 (64%)	
>3	01 (9.1%)	01 (3.4%)		02 (16.4%)	–	
Daily brushing frequency						
>1	06 (54.5%)	24 (82.8%)	0.065*	12 (92.3%)	17 (68%)	0.112[†]
1	05 (45.5%)	05 (17.2%)		01 (77%)	08 (32%)	
Do they eat sweets in between meals?						
Yes	06 (54.5%)	21 (72.4%)	0.155[†]	08 (61.5%)	19 (76%)	0.203[†]
No	02 (18.2%)	05 (17.2%)		03 (23.1%)	03 (12%)	
Sometimes	03 (27.3%)	03 (10.3%)		02 (15.4%)	03 (12%)	
Have they ever had a toothache?						
Yes	03 (27.3%)	24 (82.8%)	0.041[†]	08 (61.5%)	19 (76%)	0.122[†]
No	08 (72.7%)	05 (17.2%)		05 (38.5%)	06 (24%)	

*Chi-squared test.
[†]Fisher's exact test.

non-treated carious lesions were observed (Table 5). This fact reflects negatively on the quality of life of the individuals [29].

Individuals with haemophilia with low family income showed high caries prevalence (Table 5), similar to results encountered with patients without haemophilia [30,31]. However, no association was observed between family income and caries experience, correlating to results obtained in patients without haemophilia [32].

Although dental health services are available at HEMOPI, caries prevalence was high in the deciduous dentition, according to WHO standards, and it was possible to observe, during data collection, that the majority of patients did not spontaneously seek dental care. This relates to the habits acquired by the parents during their life experiences with dentistry, which are used as a base for their decisions on their children's dental care [33]. The literature demonstrates that regular dental visits of children with haemophilia are proportional to those of their family [34].

Periodontal diseases, traumatic brushing and consumption of dry, hard or spicy foods contribute to gingival bleeding in patients with or without haemophilia. However, predisposition for bleeding in haemophilia patients requires greater care to avoid excessive bleeding [14,15,34]. Thus, it should be emphasized that having haemophilia is not considered a determinant factor for gingival bleeding, seeing that the aetiological agent of gingivitis is the presence of organized dental biofilm [35,36].

Hormonal alterations during puberty can alter the gingival inflammatory response, provoking bleeding episodes [37–39], thus the need for clarifying parents of adolescent patients with haemophilia for effective monitoring of oral hygiene. The results of the present study demonstrated that high values of GBI were encountered in adolescent patients (Table 3). The lower levels of GBI found in the children aged 1–5 years were similar to studies with patients without haemophilia [35].

The main limitations of this study are related to potential biases associated with data obtained in the questionnaire, mainly memory bias. Another limitation is the difficulty in accessing the patients because of their great geographic dispersion, as well as their tendency to miss routine appointments at the haematology centre. The heterogeneity of the age group made it difficult to obtain a control group of healthy individuals with similar social-demographical characteristics. Furthermore, a control group constituted by a reduced number of children and adolescents with an age so heterogeneous would not be representative of a healthy population and it would not bring a confusion bias to analysis of this study.

This study obtained knowledge about the oral health of children and adolescents with haemophilia, enabling planning of future interventions directed towards this group of patients, such as implementing educational/preventative dental programmes with the intent of motivating the patients/guardians to adopt daily habits to maintain or improve oral health. Since the literature reveals that there are no differences between the types of oral problems found among patients with and without haemophilia [40], the difference is in knowing the characteristics of haemophilia for planning dental treatment and haemostatic management, as well as establishing interdisciplinary relations with other professionals seeking integral health of patients with haemophilia [4,18].

Conclusion

Patients with haemophilia in this study showed unsatisfactory oral health, with the need for restorative dental treatment. However, they displayed good periodontal health, due to reduced gingival bleeding.

Author contributions

LME participated in the design, research design, data collection, data tabulation, data interpretation and writing of the manuscript. CCBL was involved in collecting, interpreting data and writing the manuscript. RCLIL contributed to data analysis and writing of the manuscript. MDML performed data interpretation and writing of the manuscript. LFADM participated inconception, design, analysis, data interpretation and writing the manuscript.

Disclosures

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

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