

## Tooth loss and associated factors in patients with coagulopathies in the State of Paraíba, Brazil

Larycia Vicente Rodrigues  
 Mayara dos Santos Camêlo Moreira  
 Carla Ramos de Oliveira  
 Julia Julliêta de Medeiros  
 Eufrásio de Andrade Lima Neto  
 Ana Maria Gondim Valença

Universidade Federal da Paraíba - UFPB,  
 João Pessoa, PB, Brazil

**Background:** The most common and best known hereditary coagulopathies are hemophilia A and B followed by von Willebrand Disease.

**Objective:** This study aimed to estimate the prevalence of tooth loss and investigate its association with demographic and socioeconomic data, as well as to discuss self-reported oral morbidity and use of health services by patients with coagulopathies treated in blood centers in Paraíba, Brazil.

**Methods:** This was a quantitative cross-sectional epidemiological survey. Data was collected in the period from October 2011 to July 2012 by clinical examination and by assessing interviews using a semi-structured questionnaire. The findings were analyzed by descriptive and inferential statistics with the level of significance ( $\alpha$ ) being set at 10%.

**Results:** One hundred and six, predominantly male (88.8%), patients with coagulopathies were evaluated. The ages ranged from one to 59 years. Most patients were of mixed race (61.3%), most reported family incomes between R\$ 501.00 and R\$ 1500.00 (49.1%), and most had not completed elementary school (37.1%). Hemophilia A was found in 76.4% of the cases. The prevalence of dental caries among individuals was 50.0% predominantly in the 13- to 19-year-old age range (66.7%). As regards to tooth loss, teeth were missing in 35.1% of the study participants.

**Conclusion:** Tooth loss is high in this population. Males with severe hemophilia A, those who use fluoride and have a good or very good perception about their last dental appointment have a reduced chance of losing their teeth.

**Keywords:** Tooth loss; Tooth diseases; Blood coagulation disorders; Hemophilia A; Hemophilia B; von Willebrand diseases; Oral health; Logistic models

### Introduction

The most common and best known hereditary coagulopathies are hemophilia A and B. These can be classified as mild (6-50 IU/dL), moderate (1-5 IU/dL) or severe (< 1 IU/dL) depending on the presence or absence of factors VIII and IX<sup>(1)</sup>. Following these diseases is von Willebrand Disease, which is caused by a deficiency of the vWF protein which assists the transport of factor VIII and platelet aggregation<sup>(2,3)</sup>. These bleeding disorders are hemorrhagic conditions resulting from quantitative and/or qualitative deficiencies of one or more coagulation factor. These disorders are present in all ethnic groups and geographic regions but higher prevalences of certain deficiencies are reported in specific ethnic groups<sup>(4,5)</sup>.

According to studies investigating the dental status of patients with coagulopathies, these individuals tend to neglect their oral health conditions due to fear of severe bleeding which requires hospitalization, either caused by flossing and brushing their teeth at home or by invasive procedures at the dentist's office<sup>(1,6)</sup>. Some diseases as a consequence of not adopting these preventive measures, including dental caries, result in tooth loss. This affects a biological condition – chewing – and a psychological condition – self-esteem – which disturb the daily life of individuals and may cause difficulties in establishing social relationships<sup>(7)</sup>.

The occurrence of tooth loss was observed in a specific population of patients with hereditary bleeding disorders including hemophilia and von Willebrand Disease in the state of Paraíba, Brazil. Tooth loss should be minimized in this population because, even though extraction is a simple dental procedure, complications can occur when proper care is not taken that may cause death by extensive bleeding. Thus, the present study aimed to estimate the prevalence of tooth loss and investigate its association with demographic and socioeconomic data, as well as to assess self-reported oral morbidity and the use of health services by patients with coagulopathies treated in the state of Paraíba, Brazil.

### Methods

This was a cross-sectional, quantitative, epidemiological study of a descriptive and inferential nature<sup>(8)</sup>.

The collection of data on the oral health status followed the criteria established by the 2010 Brazilian survey (SB Brazil)<sup>(9)</sup> and was carried out in two blood centers responsible for the dental

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#### Corresponding author:

Larycia Vicente Rodrigues  
 Programa de Pós Graduação em Modelos de  
 Decisão e Saúde  
 Universidade Federal da Paraíba  
 CCEN - Departamento de Estatística  
 Cidade Universitária s/n  
 58051-900 João Pessoa, PB, Brazil  
 larycia\_rodrigues@yahoo.com.br

www.rbhh.org or www.scielo.br/rbhh

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care of patients with coagulopathies in Paraíba, located in the cities of João Pessoa and Campina Grande. Data were collected in the period between October 2011 and July 2012 by a previously trained examiner (inter-examiner Kappa coefficient = 0.96; intra-examiner Kappa coefficient = 0.88) and by two volunteer note takers.

A total of 106 patients with factor-deficiency coagulopathies were selected for convenience from a population of 254 individuals whom looked for dental care. Sample selection was performed regardless of age and gender. All participants signed written informed consent forms after the objectives and the purpose of the study had been duly explained by the researchers. The interviews, recorded on an audio device, were undertaken using a semi-structured questionnaire to investigate demographic and socioeconomic issues, as well as self-reported oral morbidity and the use of healthcare services. In order to assess the oral health status, clinical examinations were performed following biosafety standards using a dental mirror and a World Health Organization (WHO) probe in dental offices in the blood centers.

Data were tabulated and organized in an Excel 2010 worksheet to facilitate analysis. The variable of interest (outcome), tolerable tooth loss (TTL), was dichotomized as follows: zero was attributed to individuals aged 1-18 years with no tooth loss and also to over 19-year-old individuals who had lost up to two teeth, i.e., 10% of the 20 minimum functional dental elements of an adult population<sup>(10)</sup>. On the other hand, those who did not meet this criterion were attributed one, indicating the presence of tooth loss over the limit tolerated for the corresponding age.

Then a simple logistic regression model was adjusted, with a significance level ( $\alpha$ ) of 10%, for the TTL outcome and each of the independent variables (socioeconomic, demographic, self-reported oral morbidity and use of health services). After analyzing the variables that showed statistically significant differences ( $p$ -value  $\leq 0.10$ ), the respective odds ratio (OR) and confidence interval at a level of 90% were established based on the estimation of the angular coefficient ( $\beta$ ) of the model. The use of OR is very important for epidemiological studies because it allows a calculation of the probability of a particular disease to occur when individuals are exposed to determining factors. Hence, it was possible to identify whether the association between the outcome under study and the independent variables had occurred as protective or risk factor, in addition to quantify the intensity of this relationship. Finally, a multivariate analysis was performed using a logistic regression model containing the outcome and the statistically significant variables. The statistical program R 2.15.1 was used for the inferential analysis of data<sup>(11)</sup>.

All ethical and legal issues was taken into account following the 196/96 Brazilian Resolution that regulates how to conduct research involving human beings. Thus, this study was submitted to and approved by the Research Ethics Committee of the Hospital Universitário Lauro Wanderley (UFPB) under the number 261/11.

## Results

A total of 88.8% ( $n = 94$ ) of the 106 individuals with coagulopathies were male. The ages ranged between one and 59 years (mean: 23.34). Most participants were of mixed race

Table 1 - Distribution of patients treated in blood centers in the state of Paraíba according to demographic and socioeconomic variables and the type of coagulopathy

Variable	n	%
<b>Racial background</b>		
White	31	29.24
Black	9	8.50
Oriental		
mixed race	65	61.30
Indian	1	0.94
<b>Family income*</b>		
Up to R\$ 250.00	1	0.94
> R\$251 ≤ R\$500	6	5.66
> R\$501 ≤ R\$1500	52	49.06
> R\$ 1501 ≤ R\$ 2500	16	15.10
> R\$ 2501 ≤ R\$ 4000	7	6.60
> R\$4001 ≤ R\$ 9500	13	12.26
> R\$ 9500	8	7.55
No information	3	2.83
<b>Schooling</b>		
Illiterate	16	15.10
Incomplete elementary school	34	32.07
Complete elementary school	4	3.77
Incomplete high school	9	8.49
Complete high school	24	22.64
Complete higher education	16	15.10
No information	3	2.83
<b>Coagulopathy</b>		
Hemophilia A		
Mild	36	34.00
Moderate	17	16.03
Severe	26	24.52
With inhibitor	1	0.94
Unclassified	1	0.94
Hemophilia B		
Mild	3	2.83
Moderate	2	1.88
Severe	4	3.77
Von Willebrand disease	15	14.15
Factor VII deficiency	1	0.94
<b>Total</b>	<b>106</b>	

\*Values expressed in Brazilian reais

(61.32%;  $n = 65$ ), most had family incomes between R\$ 501.00 and R\$ 1500.00 (49.06%), and most individuals (37.08%) had not finished elementary school. With regard to the hereditary bleeding disorders, hemophilia A was found in 76.42% ( $n = 81$ ) of the patients, with 34% ( $n = 36$ ) of them classified as mild type A (Table 1).

The prevalence of dental caries among all individuals was 50.00% ( $n = 53$ ) with the 13- to 19-year-old age range predominating (66.66%;  $n = 16$ ) (Table 2).

Regarding the indices used to assess dental caries, there was a mean number of decayed, missing and filled primary (milk) teeth (dmft) of 2.45 and a mean number of decayed, missing and filled permanent teeth (DMFT) of 10.56.

Table 2 - Prevalence of dental caries among patients with coagulopathies treated in the blood centers of João Pessoa and Campina Grande, Paraíba

Age	Total			Male			Female		
	n	With caries	Prevalence (%)	n	With caries	Prevalence (%)	n	With caries	Prevalence (%)
1-5	7	4	57.14	7	4	57.14	0	-	-
6-12	20	10	50.00	19	9	47.37	1	1	100.00
13-19	24	16	66.66	23	15	65.21	1	1	100.00
20-35	34	13	38.23	27	11	40.74	7	2	28.57
36-59	21	10	47.62	18	10	55.55	3	0	0.00
Total	106	53	50.00	94	49	52.13	12	4	33.33

Table 3 - Distribution of means and standard deviation of the dmft and DMFT indices of patients with coagulopathies treated in blood centers of João Pessoa and Campina Grande, Paraíba

dmft							
Age	Mean dmft	Decayed teeth		Missing teeth		Filled teeth	
		Mean	SD	Mean	SD	Mean	SD
1-5	6.00	4.5	3.10	0.00	0.00	1.5	3.00
6-12	1.71	1.14	1.51	0.07	0.26	1.57	1.39
13-19	0.5	1.00	1.41	0.00	0.00	3.00	2.82
20-35	-	-	-	-	-	-	-
36-59	-	-	-	-	-	-	-

  

DMFT							
Age	Mean DMFT	Decayed teeth		Missing teeth		Filled teeth	
		Mean	SD	Mean	SD	Mean	SD
1-5	-	-	-	-	-	-	-
6-12	1.2	1.12	1.45	0.07	0.25	1.6	1.35
13-19	6.36	1.47	1.50	0.26	0.93	4.68	3.91
20-35	11.5	1.14	2.07	3.97	7.12	6.41	4.01
36-59	20.04	2.09	3.01	11.57	8.57	6.38	6.35

Mean number of decayed, missing and filled primary (milk) teeth: dmft; Mean number of decayed, missing and filled permanent teeth: DMFT; SD: Standard deviation

Table 4 - Statistically significant variables in the simple logistic regression for tolerable tooth loss and associated factors

Variable	Estimator	p-value	OR	90% CI
Number of missing teeth	2.0372	0.00	7.67	3.00- 19.3
Age	0.11798	0.00	1.13	1.07-1.16
Male	-1.1037	0.08	0.33	0.12-0.94
Number of people in household	0.16633	0.09	1.18	1.00-1.38
Reason for the last appointment				
Extraction	2.7081	0.01	15.00	2.29-97.51*
Treatment	0.8473	0.07	2.33	1.07-5.05
Did you feel any teeth discomfort while eating hot or cold food (Difficulty eating)?	0.7785	0.07	2.18	1.05-4.48
Discomfort when brushing your teeth	1.3318	0.00	3.78	1.7-8.33
Getting nervous about the teeth	1.1015	0.02	3.01	1.36-6.55
Being ashamed of smiling	1.4601	0.00	4.31	1.82-10.07
With severe Hemophilia A	-1.0300	0.08	0.35	0.13-0.97
Having difficulty with dentists	1.2246	0.00	3.40	1.55-7.38
Using fluoride	-1.6759	0.00	0.18	0.06-0.54
Thinking his/her quality of life is poor	2.3716	0.03	10.71	1.69-67.35*
Thinking his/her last appointment at the dentist's was				
Very good	-1.2835	0.06	0.27	0.09-0.87
Good	-1.4351	0.04	0.23	0.07-0.79

90% CI: 90% confidence interval. Due to lack of information the data of only 94 patients were used for statistical analysis. level of significance set for  $\alpha$  error = 10%. \* The amplitude of this confidence interval is owing to the standard error of the estimation, so it should be analyzed with caution.

The 1- to 5-year-old age range had a mean dmft of 6.00, with the highest mean being for decayed teeth (4.5). The highest mean DMFT values (20.04) were observed among 36- to 59-year-old individuals with missing teeth responsible for the highest value (Table 3). With regard to the variable 'outcome', 35.10% (n = 33) had lost more than the TTL. Of the 57 adults, 31 (54.38%) had lost more than two teeth, while of the 37 children and adolescents (up to 18 years), two (5.4%) had already lost at least one tooth. In addition, seven (7.45%) patients had less than 20 functional dental elements, and of these one (1.06%) was edentulous.

The simple logistic regression model showed that being male, having severe hemophilia A, using fluoride and having a good or very good perception about the last dental appointment proved to be protective factors. In contrast, the other covariates were found to be risk factors (Table 4).

## Discussion

The study population was mostly male, presenting with Hemophilia A. This predominance of the disease in males is due to its sex-linked recessive pattern (X chromosome) and to the fact that the type A of the disease has an incidence of 1:10,000 live births against between 1:40,000 and 1:50,000 for type B<sup>(12)</sup>. Von Willebrand disease affects up to 1% of the world population; studies have shown that it is more prevalent but it is underreported<sup>(5,13)</sup>.

The prevalence of mixed race among patients with coagulopathies in Paraíba can be explained by the significant number of the population that states it has this ethnic background (82.2 million). With respect to schooling, in spite of the study population not having completed elementary school, according to the Brazilian Institute of Geography and Statistics (IBGE) there was an increase in access to education among the over 10-year-old population. The percentage of people with no schooling or elementary education decreased from 65.1% to 50.2% and that of people with higher education increased from 4.4% to 7.9%. The average family income among the patients of our study was below the national average but consistent with the reality of the northeastern region. In that same group of over 10-year-old individuals, Brazil had an average family income of R\$1202.00. The Northeast has a lower income than the other regions of the country<sup>(14)</sup>.

When one aims to assess tooth loss in a given population, it is necessary to identify the oral health conditions using indices that measure the experience and severity of dental caries. Therefore the dmft and DMFT indices were used, which allow an understanding of the distribution of events in these individuals as well as establishing coping measures to be taken in order to improve their oral health conditions.

It was found that 50% of the sample had dental caries in the present study. The mean dmft and DMFT indices were higher than those of the last Brazilian oral health survey (2010 SB Brazil) of under 5-year-old children (6.00) and for those between 13 and 19 years (6.36). The seriousness of these findings can be perceived when a comparison is made with the general Brazilian population, because the mean dmft of 5-year-old children in Brazil was 2.43 with predominance of decayed teeth; at the age of 12 and for 15- to 19-year-olds the mean DMFT were 2.07 and 4.25, respectively<sup>(15)</sup>.

Similar results to this study were reported by Sudhanshu & Shashikiran (2010) in children with hemophilia, in which 87.19% had tooth decay. In the 11- to 15-year age range the authors found a prevalence of 88.09% and a mean DMFT of 5.67. The similarity in the DMFT values can be attributed to the fact that these studies evaluated the prevalence of caries in a specific population<sup>(16)</sup>.

A study of 7- to 12-year-old hemophilic children in the city of Recife, Pernambuco, found a mean dmft of 2.00, with 50% being decayed teeth and 34.5% filled teeth. The mean DMFT was 0.67 (52.2% of decayed teeth and the rest filled teeth)<sup>(17)</sup>. In another study carried out in Northern Ireland with 38 hemophiliacs, the population of 2- to 10-year-old children had, on average, 0.13 decayed teeth and a mean dmft of 0.86; in the 7- to 15-year age group none had caries and the DMFT was 0.45. The authors attributed such good oral health conditions on the wealth of the country where the study was conducted, medical support received by the hemophilic patients and the centralization of dental services inside the hospital, so there was a strong emphasis on the prevention of oral cavity diseases<sup>(18)</sup>.

As for the 6- to 12-year age group, there was a mean dmft of 1.71 and a mean DMFT of 1.2, corroborating the results found in the 2010 SB Brazil<sup>(15)</sup>. In contrast, a survey conducted in Egypt that aimed to assess the oral health status of Egyptian children with hemophilia (6-12 years; n = 60) and to implement an oral health education program, showed that the DMFT values for the hemophilic population were significantly higher than those of the non-hemophilic population. In addition, these values were higher than those of hemophilic children in developed countries with the 'decayed' component representing the major part of the value. This difference between groups was attributed to the lack of an oral health education and prevention program emphasizing the importance of oral hygiene among hemophiliacs<sup>(19)</sup>.

Regarding tooth loss, the 'missing tooth' component was found to be the most expressive in the DMFT index for the 20- to 35-year (3.97) and 36- to 59-year age groups (11.57) with mean scores for the DMFT of 11.5 and 20.04, respectively. This highlights the cumulative pattern of dental caries among patients with coagulopathies in Paraíba. As such, this reality was also identified in a national epidemiological survey (2010 SB Brazil) in which it was observed that for individuals living in the Northeast aged from 35 to 44 years the missing component accounted for 8.92 of the mean DMFT of 16.62. In the older group analyzed by the SB Brazil (65-74 years), the index increased to 27.20 with missing teeth corresponding to 25.18. It is worth noting that the age groups are different in the two studies, as in the current study, adults were grouped as 20- to 35-year and 36- to 59-year categories, whereas in the national epidemiological survey, adults were grouped in the age range of 35 to 44 years<sup>(15)</sup>. This may explain the lower values recorded for missing teeth for the patients with coagulopathies treated in the referral centers of Paraíba.

A case-control study conducted in Germany investigated the oral health and periodontal bone loss in patients with coagulation disorders compared to healthy individuals. The authors selected patients aged between 18 and 60 years presenting with hemophilia A / B or von Willebrand disease (n = 15). They found no clinically relevant difference between oral health (DMFT) of patients with

coagulopathies and the control group. In spite of the better oral hygiene in patients with coagulopathies, the authors found a mean DMFT of 18<sup>(20)</sup>.

As for the simple logistic regression model, it was found that being male, presenting with severe hemophilia A, using fluoride and having a good or very good perception about the last dental appointment are protective factors. These variables decrease the chances of patients with coagulopathies having tooth loss by 3.01-fold, 2.8-fold, 5.34-fold, 3.6-fold and 4.2-fold, respectively.

It is so inferred that the predominance of males is reflected by the biological condition of the disease. In a study by Sonbol investigating the prevalence of caries, plaque, gingivitis, enamel diseases and microflora in hemophiliacs, the author found that children with severe hemophilia have a significantly lower prevalence of dental caries in comparison with healthy children<sup>(21)</sup>. There have been no reports in the literature about severe hemophilia A behaving as a protective factor for tooth loss. The results of our study may be explained by the fact that individuals with this type of coagulopathy require greater attention, either medical or dental, which make them look for health care more often and are more likely to take care of their health.

Prevention is a key factor for good oral hygiene. Accordingly, fluoride was found to be a protective factor in our research; this is clear when one considers the established effect of exposure to fluoride products in the prevention of dental caries, as well as the reduced rate of progression in this disease<sup>(22)</sup>. It is important to point out that almost all municipalities in Paraíba do not have fluoridation of public water supplies, so the patients in this study were benefited primarily from topical fluoride products such as toothpastes. The use of fluoridated toothpaste is considered to be the main factor for the significant decline in the prevalence of caries in Brazil in recent decades<sup>(23)</sup>.

Regarding whether the last appointment had pleased the patient in a good or very good way, Oliveira et al. assessed the degree of users' satisfaction in relation to oral health in family health programs. The authors showed that 76.4% of patients attending the service in order to have their problems solved succeeded satisfactorily. Given this, the authors affirm that if their problem is solved, the user will be satisfied<sup>(24)</sup>.

An important issue to be discussed refers to the fact that the comprehensive analysis of the determinants for TTL (Table 4) was not statistically significant. The explanatory variables, when analyzed separately, were found to be associated with the outcome. However, when these variables were entered into a multivariate model, there was no statistical significance; this should be considered a limitation of this study.

It is essential to note that cross-sectional studies are those in which exposure and health statuses of individuals are determined at the same time (simultaneously). As the outcome and other factors are analyzed at the same time, one should consider the possibility of reverse causality and be cautious when interpreting the association between the variables under analysis<sup>(25)</sup>.

Furthermore, cross-sectional studies have limitations especially with respect to temporality: it is necessary to use the individuals' memory (subjective data) to unveil past facts by means of an interview, but it is difficult to standardize this data gathering process<sup>(26)</sup>.

Nevertheless, cross-sectional studies constitute a good method to detect disease frequency and risk factors. The current research evaluated the oral health status of hemophiliacs treated in blood centers of João Pessoa and Campina Grande, located in the state of Paraíba, identifying which variables were associated with tooth loss in this population.

It is also important to stress that there are few national studies assessing the prevalence of oral diseases in individuals with hemophilia. Accordingly, the present study is the first to be conducted for this purpose in the state of Paraíba.

## Conclusion

The prevalence of tooth loss in patients with coagulopathies is high. Males with severe hemophilia A, using fluoride and having a good or very good perception about the last dental appointment have less chances of losing their teeth. In addition, other factors favoring the patients with coagulopathies to lose their teeth are the number of missing teeth, aging, increased number of people in the household, extraction or treatment as the reasons for the last dental appointment, discomfort when eating cold or hot food or when brushing the teeth, getting nervous about the teeth, being ashamed of smiling, having difficulty with dentists, and thinking that their quality of life is poor.

Nowadays, educational and preventive measures in health care have become increasingly easier to plan. This fact is justified by the robust statistical analyzes applied, such as association and logistic regression tests, as these results provide a more thorough study of the disease process and help in taking relevant decisions as to the outcome of interest and associated factors.

Based on the above, it is suggested that blood centers should undergo restructuring and adopt effective strategies to promote oral health and disease prevention for people with bleeding disorders. This should happen with the active participation of patients to build a program that meets the principles of universality, integrality and equity advocated by the National Healthcare System, thus improving access to dental services and reducing dental caries and tooth loss.

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