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Original Article

Factors Associated with Toothache in Patients Affected by Hereditary Coagulopathies

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Abstract

Objective: To determine a probabilistic model that represents the likelihood of the event Dental Pain to occur in Brazilians patients affected by hereditary coagulopathies. Material and Methods: In this cross-sectional study, information on use and access to dental services, oral morbidity, self-perceived oral health and behavioral health habits were obtained through semi-structured questionnaire and analyzed by means of logistic regression. Results: High prevalence of dental caries was observed for children aged 1-5 years (50%) and for adolescents aged 13-19 years (66%). Cumulative pattern of dental caries in the age groups of 20-35 years and 36-59 years was observed. Most patients did not report pain in the last six months prior to the survey (24.5%). It was observed that searching for dental care for the last time in case of localized pain (OR=26.7), for feeling uncomfortable when brushing teeth (OR=7.3) and difficulty searching for health professionals, not only the dentist (OR 3.7) was considered risk factors for the determination of Dental Pain diagnosis. However, if the patient has mild hemophilia A (OR=0.21), this likelihood decreases, being thus a protective factor. Conclusion: Patients affected by hereditary coagulopathies have high prevalence of caries, indicating the cumulative nature of this disease, which suggests the symptomatic effect of Dental

Keywords: Health Services Research; Oral Health; Blood Coagulation Disorders.



Introduction

Dental pain, one of the most important discomfort in the context of dentistry, is considered a phenomenon that goes beyond the subjective and reaches various dimensions, as it is influenced by individual, cultural, social, psychological, environmental and physical factors [1,2]. Most of the Brazilian population seek health services in order to solve it, especially when it appears acutely, showing the presence of tissue injury by interfering with human physical integrity. Therefore, pain generates disability, reduces quality of life and brings immeasurable psychosocial and economic repercussions [2-6]. Nowadays, there has been a growing concern about dental pain, making it a major public health problem worldwide [7]. Authors have attributed this to the existence of a limited access to dental health services and to a mutilating practice, which favors the development of other health problems such as tooth loss [8,9].

Nevertheless, it is observed that studies on this topic are still scarce, especially when it comes to epidemiological studies and their social determinants [10-12]. In this perspective, the knowledge of factors about the development of oral health diseases for various populations is imperative, including individuals with specific health conditions such as the case of patients with hereditary bleeding disorders by factor deficiency (HBD).

One aspect to be observed is the fact that patients with hereditary bleeding disorders tend to neglect their oral health, perhaps due to the fear of bleeding provided by dental care, suggesting that various diseases may be more severe in this population [13,14].

In this context, the present study aimed to examine the association between socioeconomic and demographic factors, use and access to health services, self-perceived oral health and oral morbidity in patients suffering from HBD and evidence of dental pain by application of logistic regression.

The diagnosis of dental caries disease has become essential to understand the dental pain outcome in this population, because in addition to the fact that oral health issue is rarely addressed today in studies, it denotes the need to make a relevance study to help managers in decision making and in the formulation of programs to promote health and disease prevention to patients affected by bleeding disorders.

Material and Methods

Study Design

This cross-sectional study of descriptive and inferential nature with quantitative approach was conducted from October / 2011 to July / 2012 in two Blood Centers of the state of Paraiba (cities of João Pessoa and Campina Grande), located in the northeastern region Brazil. Blood Centers are references in the multidisciplinary care to individuals with bleeding disorders, among them those affected by hereditary bleeding disorders.

Sample and Data Collection



Of a total population of 165 active patients in blood centers, sampling error of 5.75 percentage points and 95% confidence level were adopted, resulting in a sample size of 116 individuals.

The selection of patients who participated in the sample met the following inclusion criteria: those who sought dental service for medical or dental care regardless of age and gender and who agreed to sign the informed consent form. Based on these criteria, 106 patients participated in the study, representing 91.5% of the estimated sample.

The collection of data on the oral health status followed criteria established by the most recent national epidemiological survey - SB Brazil 2010 [15] and was performed by an appropriately trained and calibrated examiner (kappa > 0.85), and two volunteer annotators. Interviews were conducted using a semi-structured questionnaire on demographic, socioeconomic conditions, self-reported oral morbidity, use of health services and self-perceived oral health. To evaluate oral health conditions, clinical examination was performed in the dental office of blood centers, according to biosecurity standards, using dental mirror and WHO millimeter probe. Information was tabulated and organized in an Excel 2010 worksheet to facilitate analysis.

The outcome variable Dental Pain was measured by the following question: "Did you have toothache in the last 6 months preceding the survey?" The answer was yes or no. Therefore, it was dichotomized at Y = 1 (one) if it was present and Y = 0 (zero) when absent.

Statistical Analysis

Initially, a preliminary analysis among independent variables (socioeconomic, demographic, self-reported oral morbidity, self-perceived oral health and use of health services) and tooth pain was performed using the chi-square test considering p-value ≤ 0.30 , to determine which of these would be more likely to explain the variable of interest, or, only those that obtain level $\alpha = 30\%$ would be entered in the multivariate logistic regression model.

Subsequently, all significant variables by the chi-square test were entered for adjustment of the multiple logistic regression model, where through the Backward selection method at significance level $\alpha = 5\%$, non-significant variables were removed one by one until only the determining factors for Dental Pain were defined.

With the final model, parameters Beta (β) , standard deviation, odds ratio (OR) and their confidence intervals with 95% reliability could be established for each of the variables. OR has become a reference in epidemiological studies, since it allows determining whether a factor has protective or risk behavior for the development of diseases when individuals are exposed to them and to quantify the intensity of this relationship. The R statistical software version 2.15.1 was used for inferential assessment of data [16].

Ethical Aspects



The study was submitted to and approved by the Ethics Research Committee of the Lauro Wanderley University Hospital, João Pessoa, PB, Brazil.

Results

Most patients were male (88.80%), have brown skin color (61.32%), mean age of 23.34 years, average income of R\$ 1.000,50 (49.06%) and incomplete primary education (50.94%). Hemophilia A occurred in 76.42% of cases, and 34.00% of mild type.

The prevalence of toothache in the last 6 months preceding the survey was 24.50% (n=26). By visual analog scale (VAS), toothache intensity was observed according to report from patients (Table 1).

Table 1. Visual Analog Scale (VAS) of dental pain reported by patients suffering from hereditary bleeding disorders in the state of Paraiba, Brazil.

VAS	N (%)		
0	77 (72.60)		
1	2 (1.90)		
3	2 (1.90)		
4	1 (0.90)		
5	3(2.80)		
6	4 (3.80)		
7	2 (1.90)		
8	3(2.80)		
10	9 (8.50)		
No information	3(2.80)		

Among individuals who reported having dental pain, 84.60% (n=22) already experienced dental caries at the interview. The prevalence of dental caries among individuals was 50.00% (n=53), and the age group in which lesions were more prevalent was 13-19 years, with 66.66% (n=16) - Table 2.

Table 2. Prevalence of dental caries in patients with coagulopathies assisted in blood centers of Paraiba, Brazil.

Age groups	Total			Male			Female		
	With				With			With	
	n	dental caries	Prevalence	n	dental caries	Prevalence	n	dental caries	Prevalence
1-5	07	04	57.14%	07	04	57.14%	0	-	-
6-12	20	10	50.00%	19	09	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%

Preliminary analysis has shown that variables distance (p-value = 0.09), sex (p-value = 0.00), number of persons residing in the same house (p-value = 0.21), perceiving the need for dental treatment (p-value = 0.19), seeking the dentist for pain reasons (p-value = 0.16), seeking the dentist



for treatment (p-value = 0.16), satisfaction with dental status (p-value = 0.13), mild hemophilia A (p-value = 0.06), severe hemophilia A (p-value = 0.12), Von Willebrand disease (p-value = 0.04), difficulty seeking other health professionals, not the dentist (p-value = 0.01), difficulty seeking the dentist (p-value = 0.03), receiving fluoride treatment (p-value = 0.12); Regarding the impact on oral health: Consuming hot or cold foods / drinks (p-value = 0.04), feeling uncomfortable while brushing (p-value = 0.00), getting nervous (p-value = 0.00); going to parties / having fun (p-value = 0.02), practicing sports (p-value = 0.06), talking (p-value = 0.02), studying / working (p-value = 0.11) and sleeping (p-value = 0.00), were associated with the Dental Pain outcome.

In adjusting the logistic regression model, 8 patients were excluded because 7 of them had never been to a dental office and one did complete the questionnaire.

With the use of Multiple Logistic Regression, it was possible to assess who best explains the Dental Pain outcome in the population affected for hereditary coagulopathy (Table 3).

Table 3. Logistic model for predicting dental pain in patients suffering from hereditary bleeding disorders in Paraiba, Brazil.

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Explanatory Variable	Estimator	SD	p-value	OR	CI for OR
Intercept	- 2.3145	0.5381	1.7e-05	-	-
Last time seeking the dentist due to pain (x_1)	3.2837	1.0250	0.00136	26.7	3.58; 198.88*
Feeling uncomfortable when brushing teeth (x2)	1.9886	0.6152	0.00123	7.3	2.18; 24.39
Mild Hemophilia A (x3)	- 1.5285	0.7358	0.03777	0.21	0.05; 0.91
Difficulty seeking other health professionals, not the dentist (x_4)	1.3120	0.5901	0.02618	3.7	1.16; 11.80

SD = Standard deviation; OR = Odds Ratio; C.I. = confidence interval (95%); Residual deviance: 76.005 on 93 degrees of freedom; Significance level $\alpha = 5\%$. *The interval was wide due to the standard deviation estimated for this variable, however, maintaining its statistical significance.

It was observed that variable last time seeking the dentist due to pain, feeling uncomfortable when brushing teeth and having difficulty seeking other health professionals, not the dentist, were considered risk factors for the Dental Pain outcome, increasing the chances of its occurrence in this population, respectively, 26.7, 7.3 and 3.7 times. If the patient is affected by mild hemophilia A, this makes the chances to decrease 4.6 times, which is a protective factor.

Discussion

This research examined the relationship between socioeconomic and demographic factors, use and access to health services, self-perceived health and oral morbidity and self-reported dental pain in patients suffering from hereditary bleeding disorders in a city in northeastern Brazil, identifying risk and protective factors to this disease.

This study is one of the few in literature that proposed to investigate such association and emphasizes that aspects even more preliminary on such patients are infrequent, specifically regarding dental pain in patients suffering from coagulopathies, studies are even less frequent, making the findings of this work, at some times of the discussion, to be compared to surveys covering dental pain in patients without coagulation disorders, as evidenced by authors when observing the lack of



epidemiological studies on the oral health condition of hemophilic patients. The authors found that the available results are controversial and dependent on the countries where studies were developed.

[17]

One aspect to be highlighted is the caution that should be taken in generalizing the results of this study, since it was developed in two regional blood centers of a single Brazilian state. However, we emphasize that the regional blood centers are reference centers for patients with coagulopathies, and therefore we understand that the identification of factors associated with dental pain in these individuals would make it possible to approach the reality of people affected by such condition.

As previously pointed out, due to the organization of users at health care points and the strategic role that regional blood centers play in providing care to individuals with bleeding disorders, these health facilities were selected as study setting, thus justifying the convenience sampling.

One limitation of this study is the small number of observations (n=106). However, this fact does not compromise its internal validity because the sampling error and the assumed reliability level provide accuracy of estimates.

In Brazil, there are around 82.2 million of brown skin color individuals, especially in the northern and northeastern regions, according to the 2010 census [18]. Brown skin color was present in 61.32% of respondents. The clinical importance of this factor against dental pain may be explained by the high incidence of black and brown individuals in the region where the research was developed.

The average income of patients with HBD ranged from U\$ 284.66 to U\$ 852.27 (average of U\$ 568.46), i.e., 49.06% have income below the national average, which is U\$ 682.95. This can be explained by the fact that the northeastern region of Brazil has had the lowest income in the last 10 years when compared to other regions of the country, reaching only U\$ 457.95 against U\$ 807.95 in the Midwestern region, for example [18]. Studies confirm that the experience of pain is present in people with low socioeconomic status, mainly because in the poor population there is a greater prevalence of the oral disease and there is delay in the search for the treatment [19].

Of the individuals who composed the sample, 50.94% had not finished elementary school. Corroborating this finding, a study conducted in Brazil found that among individuals aged 9-50 years with hemophilia, 69.9% of them were students, but with predominance of incomplete primary school in 45.5% of cases [20]. According to international studies, school absenteeism has played an important role in the lives of individuals with hemophilia, therefore determining low educational levels in this population [20]. In this situation, the low level of schooling suggests a predisposition to dental pain and other problems in buccal health due to the little information on the subject [21].

About dental caries experience, patients analyzed here had dental caries at a fairly high proportion and from an early age, with higher proportions among the age groups of 13-19 years followed by 1-5 years. Corroborating these findings, a study that evaluated various oral health diseases in order to identify disparities by race / color among adolescents in each one of the Brazilian



macro-regions, showed that the northern and northeastern regions had the worst oral health indices, and that this finding was not surprising, since these are the regions with the poorest socio-economic profiles of the country. In addition, the finding of high prevalence of dental caries is consistent [4].

Dental pain is a condition that is strongly associated with oral health care. The literature shows the complexity of factors that may be related to this situation. It is believed that social, demographics, genetic, behavioral, and also cognitive aspects are among the causes [22-24]. Suggest that the dental surgeon may express the feeling of not being adequately trained to assist patients with HBD, and consequently the individual may have a negative experience and only return to the service when he really needs help, as in the case of dental pain that doesn't give way [25].

In this study, the explanatory variable seeking the dentist for pain reasons, a risk factor for the outcome studied, was maintained to reinforce its importance due to the high prevalence of this disease in the population of northeastern Brazil (12.2%), mainly in the age group 35-44 years, as evidenced in the latest national epidemiological survey [15].

About that fact that variable feeling uncomfortable while brushing increases the likelihood of toothache by 7.3 times (OR = 7.3), it can be attributed to the intrinsic characteristic of this pathology, which is to present bleeding, especially during sudden brushing activities or flossing, for example, and because the oral cavity is a highly vascularized area [26]. This situation causes patients to restrict this kind of preventive activity, thus allowing the accumulation of dental plaque, caries formation and periodontal diseases, which will be strongly manifested with the presence of dental pain if the visit to the dentist is not regular.

Some authors have already reported that failure/negligence in tooth brushing by hemophilia patients led them to present worse DMFT index (number of decayed, missing and filled teeth in the permanent dentition) when compared to the control group without the disease [27]. Another study on oral diseases of children with chronic conditions mentioned hemophilia as a type of pathology related to high risk for the formation of gum disease due to poor brushing and presence of gingival bleeding [28], as well as greater negative impact among individuals who have never used dental floss in life compared to those who use it frequently [29].

However, a case-control study conducted in Lithuania found that better oral health condition (lower caries experience and need for dental treatment) was observed in children with hemophilia compared to healthy ones. This result was not found for permanent teeth. The authors suggest that parents of children with hemophilia are more careful with the oral health of their children possibly because they are more aware of the potential risk that problems of dental origin would lead to children with hemophilia [30].

These controversial findings could be attributed to the reality of each country where the study was developed, reinforcing the need for conducting local studies in order to identify the oral health condition of patients with congenital coagulopathies and the factors associated with it. This research meets this concern, revealing the demands of a Brazilian population with coagulopathy.



In relation to the report of respondents having difficulties searching for health professionals, not only the dentist, and this variable being up to 3.07 times more likely to present situations of pain is interpreted as appropriate and correct for the analysis, since a small number of professionals have knowledge about hereditary bleeding disorders, consecutively, would have difficulty in identifying the possible implications that can affect the quality of life and oral health of these individuals. Hemophilia, for example, is one of the most challenging medical conditions in the health field for all professionals, showing the need for dedication and union of this multidisciplinary body in the care of these individuals [26], thus explaining the presence of this risk in the logistic model for dental pain.

The fact that variable mild hemophilia A has behaved as a protective factor on dental pain, decreasing the chances of its occurrence in 4.6 times is due to the characteristic that the disease imposes on these individuals, which is the need for preventive and regular care. I.e., the absent coagulation factor must be replaced so that individuals with coagulopathy will have a normal life and avoid further damage to their quality of life. Thus, due to this ongoing and permanent care, patients with HBD can count on a multidisciplinary team with expertise in the area to assist them with anything they need, when it comes to reference centers. The dentist plays a key role in the control of oral diseases; therefore, it is known that simple procedures such as tooth extraction, if not well planned, can cause the patient to death due to severe bleeding.

In developed countries, hemophiliacs are advised to seek preventive and regular dental care as early as possible, since advanced oral health diseases and subsequent treatments appear to be more risky and complicated in this population [27]. Therefore, it is believed that this protective behavior occurs when caries is diagnosed and particularly when the individual received guidance so that new cases do not occur, or, the adoption of this new knowledge makes the search for dentist to be more efficient, periodic and important in their lives, therefore reducing the prevalence and severity of dental caries and consequently, dental pain.

Studies involving a probabilistic model may be important in this context. The results found on the association of dental pain and its influence on certain factors in the life of patients with coagulopathy, either of risk or protection, demonstrate that this is a condition that deserves greater attention in the field of public health, carrying out further studies on the continuous and permanent monitoring of the quality of dental care being offered to these individuals. A study conducted in Malaysia proved that the multidisciplinary team involved in the care of patients with hemophilia tends to have a similar oral condition, and even better, than people without the disease [31].

Conclusion

In this study, patients suffering from hereditary bleeding disorders have high prevalence of caries, showing the cumulative nature of this disease, which suggests the symptomatic effect of dental pain. The risk of development of dental pain was associated with seeking the dentist only when pain is already present, when feeling uncomfortable when brushing the teeth and reporting



difficulty in seeking other health professionals, not only the dentist. On the other hand, having mild hemophilia A provided a protection feature to dental pain.

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