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Original Article

Evaluation of gum health status in hemophilia patients in Birjand (a case-control study)

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Abstract: Background: Gum bleeding in the absence of trauma or injury in adults with haemophilia is often reported. So hemophilic patients require special care and attention in various dentistry practices. The purpose of this study was to evaluate the gingival health status in patients with hemophilia in Birjand in 2018-2019. Methods: Sampling was used in this case-control study to recruit cases (89 patients with hemophilia) and 89 individuals as control group recruited randomly from the general population, which were matched with cases based on gender, age and place of residence. Clinical examinations included dental health and salivary assessments. After taking informed consent from cases who had the inclusion criteria for the study, information including demographic characterizes, family history, viral infections, coagulation history, and the time of onset treatment was recorded. Oral examination was done by catheter and dental mirror. MGI (Modified gingival index) and Periodontal Index (PI) were determined for each subject. Results: In this study, 84.3% of the hemophilia patients were males and 76.4% of them had a family history of hemophilia. The results show that difference between the mean of MGI and PI in hemophilic patients and healthy subjects is not statistically significant ($P>0.05$) while, it is higher in hemophilic patients than healthy subjects, but this difference is not statistically significant ($P>0.05$). Also, with increasing age, MGI and PI showed a significant increase ($P<0.05$). Conclusion: Although the higher MGI and PI index in hemophilia patients, attention to oral health and frequent control in hemophiliac is necessary. Also, to prevent dental problems and further bleeding play an important role.

Keywords: Hemophilia, gingival inflammation index (MGI), periodontal index (PI)

Introduction

The World Health Organization views oral health as a necessity and as an important part of public health throughout life and states that poor oral health and untreated oral diseases can have a profound effect on the quality of life [1]. There are several thousand people with periodontal disease in the world; the incidence of these diseases is high and exceeds the prevalence of dental caries [2]. Periodontal diseases are the leading cause of permanent tooth loss, inflammation, hemorrhage, gingival resorption, bone resorption, mobility and tooth movement, pain, etc, are the complications of periodontal disease [3]. Periodontal diseases can lead to bad breath and problems with speech, chewing, tooth eruption, and pain. On the other hand, gingival health plays an important role in physical and mental health [4]. Gum disease interferes with beauty, causing anxiety, depression, and worry in individuals [5]. Various studies have shown that there are various factors, including demographic factors such as age, sex, socioeconomic status, inflammatory factors, systemic conditions, medications, and many other agents that can affect periodontal disease [5-8].

One of the systemic conditions affecting periodontal tissue is hemophilia. Hemophilia is a coagulopathy disorder; so that, patients with

hemophilia require special attention and care due to the importance and the critical role of coagulation in different dental practices [9]. In the different varieties of hemophilia, there are disorders in the blood coagulation process that results from a defect in a coagulation precursor [10]. The most common types of hemophilia are types A and B. Oral manifestations of hemophilia occur in different forms. Bleeding from different parts of the oral cavity and gingival bleeding can be very costly and prolonged. Even the physiological stages of tooth eruption can be associated with prolonged bleeding [11]. Hemophilic pseudotumor in the mandible is a complication that occurs due to repeated sub-periosteal bleeding. Other oral findings, such as high rates of caries and extensive periodontal disease, appear to be more due to poor health and diet than hemophilia [12]. Comprehensive hemophilia care includes all medical services including dentistry. Dentists are responsible for providing dental care (preventive, restorative, prosthetic, orthodontic, etc.), cooperation with hematologists to prevent bleeding during dental practice, educating the patient and their families about oral care at home. On the other hand, periodontal tissue health is essential to prevent bleeding and tooth loss [13]. The risk of bleeding due to periodontal treatment in hemophilia patients can be problematic. Considering the possibility of these patients being consulted and referred by physicians to dentists and the high prevalence of these diseases in societies where consanguine marriages are common, so studying about these problems is of great importance [14].

This study aimed to evaluate the gingival and periodontal health of patients with hemophilia, so that, in this paper, we determined the health status of gingival in patients with hemophilia and compare them with healthy individuals in Birjand, during 2018-2019.

Materials and methods

This study was a case-control study.

Sample size and sampling method

The sample size was based on the comparison of two means and the results related to the MGI index in the study of Othman [8], which in each group, 89 patients were estimated. The case group was selected by a non-probability sampling method from the patients with hemophilia referred to the hemophilia center of South Khorasan. Also, patients referred to the dental clinic who were matched for age, sex to the case group and had not been referred for dental services, were selected as the control group.

Inclusion criteria: Patients with type A and B hemophilia were matched by age and sex with healthy controls.

Exclusion criteria: Patients with severe crowding, history of periodontal surgery, use of narcotics or tobacco products, use of drugs affecting the histological features of the gums such as *Nefidipine* and *Corticosteroids* - in addition to other systemic diseases such as leukemia, diabetes, white blood cell deficiency, severe anemia and -AIDS, failure to complete the consent form.

After obtaining informed consent from people and evaluating their criteria to enter the study, their information was recorded in a questionnaire form consisting of demographic information, family history, viral infections (such as HCV, HBV, HIV), history of coagulation problems and time of treatment. Onset examination was performed by a dental catheter and mirror. The patients were seated in a chair in front of a flashlight and an oral examination was performed by a trained examiner under the supervision of periodontitis.

Modified gingival index (MGI) and Periodontal Index (PI) were used to measure gingivitis. The MGI index was especially used when probing the gum was not possible. All subjects were examined by a specialist and the above criteria were recorded.

Statistical analysis

The quantitative and qualitative data were analyzed using SPSS, ver. 22 (Chicago, IL, U.S.). For qualitative data, the normal distribution was assessed using the K-S test. For data with a normal distribution, the means ± standard deviation (SD) was computed, and independent T-tests and the Mann-Whitney test were performed. Qualitative data were described and analyzed using frequencies and percent, in addition to the chi-squared test. A P value less than 0.05 was considered statistically significant.

Table 1. Frequency distribution of clinical information in the study groups

Study group		Hemophilia patients N (%)	healthy people N (%)	Total N (%)
sex	male	75 (84.3)	75 (84.3)	150 (84.3)
	female	14 (15.7)	14 (15.7)	28 (15.7)
Type of hemophilia	A	73 (82)	-	
	B	12 (13.5)	-	
	other	4 (4.5)	-	
Family history	Yes	68 (76.4)	-	-
	No	21 (23.6)	-	-

Ethical considerations

All of the participants were provided free written consent to participate in the study before entering the study and were examined and guided. All information obtained was confidential. Patients could also drop out of the study at any stage of the study if they did not willing to participate.

Results

Characteristics of the study participants

This study was performed on 89 hemophilic patients and 89 healthy individuals with a mean age of 26.6±14.8 and 27.5±15.1 years in hemophilia patients and healthy subjects respectively; which was not statistically significant (P>0.05). The frequency distribution of sex was not observed statistically significant in the two groups of hemophilic patients and healthy subjects.

Most of the patients had hemophilia type A (82%) and 76.4% had a family history of hemophilia and none of them had hepatitis B or C (**Table 1**).

Comparing behaviors associated with gum health in two groups

There was no statistically significant difference in referral to the dentists between the two study groups. There was also no significant difference in the cause of the referral to the dentists in the two study groups (**Table 2**).

The use of mouthwash and tooth brushing in healthy subjects was significantly better than hemophilia patients. As only 10.1% of hemophilia patients used mouthwash and 31.5% did not brush at all, this condition was more favorable in healthy subjects (**Table 2**).

The worst MGI and PI index was found in people who never brushed, and the best was found in those who brushed once a day, the difference between the groups who never brushed with those who brushed once a day or brushed 2 to 3 times a week showed a significant difference (P<0.05) (**Table 2**).

Although the mean of MGI and PI index in hemophilia patients were higher than healthy individuals, this difference was not statistically significant (**Table 3**).

There was a significant increase in MGI and PI index with age; so that, Mann-Whitney test showed a statistically significant difference between two age groups of below 10 years and 30 years and over (P<0.05) (**Table 5**).

No significant difference was found in MGI and PI index in hemophilia patients by sex (P>0.05) (**Table 3**).

No significant difference was observed in MGI and PI index in hemophilia subtypes in the studied patients (**Table 4**).

Discussion

Hemophilia patients are in the category of patients with a coagulopathy disorder, who need special attention and care due to the importance and vital role of coagulation in different dental practices [15]. Due to the high prevalence of these diseases in societies where consanguine marriages are common; so, it is important to do studies about the problems of these patients. Most studies on hemophilia patients in Iran have focused on medical aspects such as drug dosage determination and different methods of gene therapy and surgery and no similar study has been performed on the determination of gingival markers in hemophilia patients [16, 17]. The purpose of this study was to compare the gingival health status of hemophilia patients with healthy individuals in Birjand.

According to the results of this study, most of the participants were male. In similar studies,

Table 2. Comparison of the status of dentist referral in the two study groups

Study group		Hemophilia patients N (%)	healthy people N (%)	Total N (%)	Test Result	*P*.value
Referral status	recently	12 (13.5)	10 (11.2)	22 (12.4)	λ^2=1.71	0.79
	6 months ago	8 (9)	7 (7.9)	15 (8.4)		
	One year ago	10 (11.2)	6 (6.7)	16 (9)		
	More than one year	50 (56.2)	57 (64)	107 (60.1)		
	No referral	9 (10.1)	9 (10.1)	18 (10.1)		
Reason for referral	Restoration	26 (32.5)	36 (45)	62 (38.8)	Fisher exact test =10.71	0.069
	pull	34 (42.5)	27 (33.8)	61 (38.1)		
	root canal	3 (3.8)	8 (10)	11 (6.9)		
	Gums Bleeding	2 (2.5)	0 (0)	2 (1.3)		
	Examination	12 (15)	4 (5)	16 (10)		
	Surgery	1 (1.3)	3 (3.8)	4 (2.5)		
	Crime	2 (2.5)	2 (2.5)	4 (2.5)		
Use mouthwash	Yes	9 (10.1)	28 (31.5)		λ^2=12.32	P<0.001
	No	80 (89.9)	61 (68.5)			
Brushing status	No brushing	28 (31.5)	9 (10.1)		λ^2=13.69	P<0.003
	Once a day	23 (25.8)	26 (29.2)			
	2 or 3 times in week	23 (25.8)	27 (30.3)			
	Once a week	15 (16.9)	27 (30.3)			

Table 3. Comparison of mean index - MGI and PI in the studies groups

Variable \ Study group	Hemophilia patients (N=89) Mean ± SD	healthy people (N=89) Mean ± SD	Mann-Whitney test	*P*.value
MGI	0.703±0.88	0.505±0.68	Z=1.16	0.25
PI	0.48±0.68	0.43±0.58	Z=0.14	0.89

Table 4. Comparison of mean index - MGI and PI according to hemophilia type and gender in hemophilia patients

variable \ index		MGI (N) Mean ± SD	PI Mean ± SD
sex	(n)		
	Male (75)	0.74±0.89	0.49±0.67
	Female (14)	0.53±0.85	0.37±0.73
	Mann-Whitney test	Z=1.22	Z=1.42
P.value		0.22	0.15
Hemophilia type	B (12)	0.75±0.84	0.48±0.76
	A (73)	0.69±0.89	0.46±0.66
	Other (4)	0.71±1.15	0.71±0.96
	Chi-Squre test	λ^2=0.39	λ^2=0.74
P.value		0.82	0.082

the majority of hemophilia patients were male [18]. The most common type of hemophilia in the present study and other similar studies was type A hemophilia. In Valizadeh's study, the prevalence of hemophilia A (82%) was higher than that of hemophilia B (13.5%). Hemophilia A is also more prevalent than hemophilia B in Iran [19].

Considering family history, 76.4% of the subjects in the present study had a positive family history. In the Makarem's study, 48.6% of patients had a family history. The hemophilia defective gene is always found on the X chromosome and is a sex-linked disorder, but it has been estimated that 30% of cases of hemophilia have no family history of such disorder and it can occur as a result of gene mutation. In another study, 70-80% of hemophilia patients had a history of hemophilia in their family, relatives, and ancestors, which is similar to the results of the present study [15, 19].

The results of the present study show that the mean of MGI and PI index in hemophilia pati-

Table 5. Comparison of mean MGI and PI index by age groups in hemophilic patients

index / age	MGI (N) Mean ± SD	PI Mean ± SD	Mann-Whitney test (*p*.value)
<15	(27) 0.33±0.64**	0.51±0.25*	*P=0.0131
15-29	(22) 0.65±0.84	0.43±0.6	**P=0.0012
>29	(40) 0.98±0.96**	0.65±0.78*	
Test results	λ^2=10.73	λ^2=6.41	
P.value	0.005	0.041	

1-*p*.value of comparing 2 groups showed with one star. 2-*P*.value of comparing 2 groups showed with double star.

ents was higher than healthy individuals but this difference was not statistically significant. Higher levels of these two factors in hemophilia patients may be due to the lack of hygiene in fear of bleeding. In the Makarem, Ahmadi's study that reported DMFT and MGI indexes, these two indexes were also higher in hemophilic patients than in healthy patients, which is similar to the results of this study [16, 20].

Ziebolz reported a prevalence of moderate to severe periodontitis in 80% of the hemophilic population and 48% of healthy subjects [17]. The results of Othman's study showed that the MGI index was significantly lower in hemophilia than healthy controls [8], which is not in line with the results of the present study. In Nagaveni and Zaliuniene studies; the OHI index was better in hemophilic patients than healthy controls but there was no significant difference between two groups [9, 11]. Othman stated that the multifaceted care of the medical team and the presence of a dentist in the hemophilia team improve the oral health status of these patients.

In the present study, MGI and PI increased significantly with increasing age, and there was a significant difference between two age groups of below 15 years and 30 years to more. In Ahmadi and Makarem's study, as age increased, there was a significant increase in MGI, which was found the worst in people who never brushed and the best in those who brushed once daily, and the difference was statistically significant between the groups who never brushed and those who brushed once daily or 2-3 times a week [16].

It seems that lack of awareness about proper brushing and fear of bleeding causes poor oral hygiene in hemophilic patients. In the study of Ahmadi; there was a significant difference in the oral health indexes between those who regularly used toothbrushes compared to those who did not use a toothbrush and mouthwash [21]. Contrary to the results of the present study in the Macram's study, there was no significant difference between the MGI index and the frequency of tooth brushing in hemophilic patients [16]. In another study, parents' awareness of oral hygiene (use of a toothbrush, dental floss, and mouthwash) was very poor and 69.47% of parents did not have proper information about oral hygiene and the most important reason for not brushing in this patients was the fear of bleeding [22].

The present study did not find a significant difference in MGI and PI index in hemophilia patients by sex and type of hemophilia. No similar study was found to investigate these two variables. One of the advantages of the present study is the higher sample size in this study compared to similar studies [18]. In this study, in addition to the MGI index, the PI index was also measured. In such similar studies, only the MGI index was used to report periodontal status. One of the limitations of the present study was the lack of evaluation of the DMFT index in the study participants.

Conclusion

To prevent oral diseases, in addition to examining oral health status, similar studies should be conducted to determine the effect of education on the oral health status of these individuals. So that; better planning could be done to provide health services and raise the awareness of these patients to prevent gum disease it is suggested to design studies on parental awareness to develop effective measures to increase the oral health of these individuals.

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Dr. Parvayi and Dr. Ziaee designed and performed the research study analyzed the data and wrote the paper. Dr. Parvayi designed the research study; Mr. Sharifzadeh analyzed the data and wrote the paper. Dr. Osmani, designed the research study and analyzed the data and wrote the paper. Neda Shaygan contributed to the data collection and wrote the paper. All authors critically reviewed the manuscript and authorized the final version of the paper.

Disclosure of conflict of interest

None.

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