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Original Article

# Evaluation of Oral and Dental Health Findings in Children with Hemophilia

## *Hemofili Hastası Çocuklarda Ağız ve Diş Sağlığı Bulgularının Değerlendirilmesi*

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## ABSTRACT

**Objective:** This study aims to compare the dental caries, plaque accumulation, gingival health, and oral hygiene habits of children with hemophilia to those of systemically healthy children of the same age group.

**Method:** Our study consists of a study group of 54 male children with hemophilia, with a mean age of 7.80±3.18 years, and a control group of 55 healthy male children, with a mean age of 8.20±2.63 years. A case report form was used to assess patients' demographics, medical and dental history, and oral hygiene habits. In addition to an intraoral examination, data related to oral hygiene, including caries indices [decayed, missing, filled teeth (DMFT/dmft) gingival index (GI), and plaque index (PI), were recorded in the case report form.

**Results:** The dmft score of the control group was found to be significantly higher than that of the study group. However, the DMFT score did not show a significant difference between the groups. The GI score did not exhibit a significant difference between the groups either. The PI score of the control group was found to be significantly higher than that of the study group. No significant difference was observed between the two groups in terms of oral hygiene habits.

**Conclusion:** Children with hemophilia are a special patient group at risk in terms of oral and dental health. Awareness should be raised among patients and their parents to ensure preventive dental treatments and regular dental check-ups from an early age.

**Keywords:** Hemophilia, oral hygiene, DMFT/dmft index, gingival index, plaque index

## ÖZ

**Amaç:** Bu çalışmada, hemofili hastası çocukların diş çürüğü, plak birikimi, diş eti sağlığı ve oral hijyen alışkanlıklarının yaş grubu ile uyumlu sistemik olarak sağlıklı çocuklarla karşılaştırılması hedeflenmiştir.

**Yöntem:** Çalışmamız, yaş ortalaması 7,80±3,18 olan 54 hemofili hastası erkek çocuk hastadan oluşan çalışma grubu ve yaş ortalaması 8,20±2,63 olan sağlıklı 55 erkek çocuktan oluşan kontrol grubundan oluşmaktadır. Hastaların demografik özelliklerini, tıbbi ve dental geçmişlerini ve ağız hijyeni alışkanlıklarını değerlendirmek için bir olgu rapor formu kullanılmıştır. Ağız içi muayene ile birlikte olgu rapor formunda yer alan ağız hijyeni ile ilişkili çürük indeksleri [daimi dişlerde çürük, eksik ve dolgulu diş sayısı (DMFT/dmft)], dişeti indeksi (GI) ve plak indeksi (PI) verileri kaydedilmiştir.

**Bulgular:** Kontrol grubu dmft skoru, çalışma grubuna göre anlamlı düzeyde yüksek bulunmuştur. DMFT skorunun ise gruplara göre anlamlı farklılık göstermediği tespit edilmiştir. GI skoru gruplara göre istatistiksel olarak anlamlı farklılık göstermemiştir. Kontrol grubu PI skoru, çalışma grubu PI skoruna göre anlamlı düzeyde daha yüksek tespit edilmiştir. Oral hijyen alışkanlıkları açısından iki grup arasında anlamlı bir fark bulunmamıştır.

**Sonuç:** Hemofili hastalığına sahip çocuklar ağız ve diş sağlığı açısından risk altında olan özel hasta grubundadır. Erken yaşlardan itibaren koruyucu diş tedavileri ve düzenli diş hekimi kontrollerinin sağlanması amacıyla hastalar ve ebeveynlerinde farkındalık oluşturulmalıdır.

**Anahtar kelimeler:** Hemofili, oral hijyen, DMFT/dmft indeksi, gingival indeks, plak indeksi

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## INTRODUCTION

Hemophilia is a congenital bleeding disorder caused by a deficiency of coagulation Factor VIII or Factor IX, and follows an X-linked recessive inheritance pattern. The deficiency results from mutations in the genes encoding the respective coagulation factors[1]. Individuals affected by hemophilia exhibit a lifelong predisposition to bleeding due to the failure of secondary hemostasis, with the severity of the disorder correlating with the extent of deficiency in the specific coagulation factor[2]. A family history of bleeding is identified in approximately two-thirds of all patients. Hemophilia A is significantly more common than hemophilia B, with estimates suggesting that hemophilia A accounts for 80-85%, while hemophilia B constitutes approximately 15-20% of all hemophilia cases[3].

Oral and dental health constitute integral part of overall health. Throughout both childhood and adulthood, oral and dental health can be directly affected by various systemic diseases that impact other bodily systems and may also negatively influence general health state[4]. Since the oral cavity is one of the highly vascularized regions prone to frequent bleeding in individuals with hemophilia, it is crucial for dentists to have adequate knowledge in oral and dental care in order to play a role in early diagnosis, administer appropriate dental treatments, and prevent potential complications [5,6]. Patients with bleeding disorders may neglect oral and dental care due to their low levels of education, difficulties in affording for factor concentrates or fear of bleeding during dental procedures or tooth brushing[7].

Although individuals with hemophilia do not constitute a large proportion of the population, their dental treatments are often challenging, very risky, and costly, as they may require factor supplementation prior to procedures[8]. Therefore, after the diagnosis of hemophilia is established, it is crucial to educate families about regular dental check-ups, oral hygiene practices, and preventive dental treatments. Early-stage awareness and preventive approaches play a critical role in reducing the need for more invasive and complex treatments in the future, thereby enhancing the patients' quality of life and preventing high treatment costs[1,9-11].

The aim of our study is to compare the oral health findings of children with hemophilia to those of systemically healthy children of the same age group. Additionally, the study aims to raise an awareness about the importance of establishing the foundations of healthy and proper oral hygiene at an early age for children with hemophilia, as is the case for all children with special medical conditions.

## MATERIALS and METHODS

The study group of our research consisted of children aged 2-13 years diagnosed,and followed up with hemophilia at the Ege University Department of Pediatrics, Division of Pediatric Hematology and Oncology, Faculty of Medicine, as well as children with hemophilia aged 2-13 years who applied for a general examination at the Ege University Department of Pedodontics, Faculty of Dentistry, located in İzmir and surrounding provinces. The control group consisted of healthy children aged 2-13 years without any systemic disease who applied to the Ege University Department of Pedodontics, Faculty of Dentistry for a general dental examination.

Our study was ethically approved by the Ege University Medical Research Ethics Committee with the decision dated April 25, 2024, and numbered 24-4.1T/62.

### Selection Criteria of the Subjects

Patients aged between 2-13 years with the established diagnosis of hemophilia, whose caregivers had given their consent for the participation of the children in the study constituted the study group. Patients older than 13 years, without proven diagnosis of hemophilia, and refused to participate were excluded from the study. The control group included children aged 2-13 years who were healthy without any systemic disease, physical or mental disabilities, and applied for a routine examination to the Department of Pedodontics, and Faculty of Dentistry of Ege University.

### Study Design and Procedures

In this study, a case report form was used as a data collection tool. The form contained various questions to assess the patient's demographic characteristics, medical and dental history, and oral hygiene habits. In the study group, information acquired from the parents about the child's age, gender, type of hemophilia, any accompanying systemic diseases, medication use, and the presence of any physical/mental disabilities were recorded. To evaluate oral hygiene habits, questions about the presence and frequency of tooth brushing, type of toothbrush, whether bleeding occurs while brushing, interproximal tooth care and its frequency, presence of foul breath, frequency of dental visits, and the timing of the last dental visit were inquired. The form also inquired about whether bleeding problems

occurred during or after dental treatment, and whether premedication and consultation requests were made before dental procedures. In the control group, the parents provided information about the child's age, gender, tooth brushing habits and its frequency, type of toothbrush used, presence of bleeding during brushing, interproximal tooth care and frequency, presence of foul breath, frequency of dental visits, and the timing of the last visit.

Both groups of patients underwent dental examinations using a mirror and probe under a reflected light during the appointment. The caries status of the patients was recorded according to the decayed, missing, filled teeth (DMFT/dmft) index. The DMFT index is calculated by summing the number of decayed, missing, and filled permanent teeth, while the dmft index is determined by the total number of decayed and filled primary teeth. This index was first introduced by Klein and Palmer in 1938 and is considered a key measure in dental epidemiology for assessing the oral health status of populations[12].

Dental surfaces were examined using a mirror and a blunt-ended probe under reflector light. A single score was assigned for each tooth, and the total score was used to determine the index value. In the calculation of the DMFT or dmft indices, congenitally missing teeth, unerupted teeth, supernumerary teeth, teeth lost or restored due to reasons other than caries, and physiologically exfoliated primary teeth were excluded from the evaluation. Information about the type of dentition (primary, permanent, or mixed dentition) was also recorded during the intraoral examination. The gingival health status was assessed using the gingival index (GI) proposed by Silness and Loe[13] (1963) during the oral examination. The GI is a scoring system that grades different stages of gingival inflammation based on clinical features. Six representative teeth, reflecting the entire mouth, are examined for the severity of inflammatory changes in the gingiva, and each site is assigned a score ranging from 0 to 3 points. The scores for each tooth are summed and divided by four to obtain the GI value for that tooth. The indices of the teeth representing each region are then summed and divided by six to calculate the patient's overall GI. The GI value represents an average GI score of the examined gingival areas of the patient[13].

Plaque presence and amount on the dental surfaces were recorded according to the Plaque Index (PI) proposed by Silness and Loe[13] (1964). The PI is an index used to determine the amount of dental plaque in contact with the gingiva. Six teeth representing the entire mouth are selected for examination. Each of the four surfaces of the selected teeth—buccal, lingual, mesial, and distal—is scored on a scale from 0 to 3 points. The scores for each tooth are summed and divided by four to obtain the PI value for that tooth. The indices of the teeth representing each region are then summed and divided by six to calculate the patient's overall PI. The PI value represents the average score of the examined gingival sites for the patient[14].

To ensure standardization, all oral examinations and evaluations of patients in both the study and control groups were performed by a single researcher (M.D.).

### Statistical Analysis

The data were analyzed using the SPSS 21.0 software. The frequency and percentage tables of the evaluation questions in the case report form were compared between groups using the chi-square test. Since our groups did not show a normal distribution, the comparison of the DMFT, GI, and PI scores between two groups was performed using the Mann-Whitney U test, while for comparisons involving more than two groups, the Kruskal-Wallis H test was used. To analyze the correlation between DMFT, GI, and PI scores, the Spearman's Rho correlation test was applied. The confidence interval was set at 95%, and the significance level was determined as $p<0.05$.

## RESULTS

The study group consisted of 54 male hemophiliac children with a mean age of 7.80±3.18 years and a control group of 55 healthy male children with a mean age of 8.20±2.63 years. Among the cases in the study group, 83.3% (n=45) were diagnosed with hemophilia A, and 16.7% (n=9) of them with hemophilia B (Table 1). Fifty-four hemophiliac patients in the study group, had either mild (n=14: 25.9%), moderate (n=9: 16.6%) or severe (n=31: 57.4%) hemophilia.

In the study group, 87% and in the control group, 85.5% of the cases were found to have the habit of tooth brushing. There was no statistically significant difference

**Table 1. Distribution of the study group according to hemophilia type**

| Hemophilia type | n | % |
|---|---|---|
| A | 45 | 83.3 |
| B | 9 | 16.7 |

between groups in terms of the daily frequency of tooth brushing. In the patient group, 23.4% and in the control group, 12.8% of the cases brushed their teeth twice a day. Teeth brushing frequency appeared consistent across the groups, without any significant differences between both groups. Gum bleeding during tooth brushing was observed in 14.8% of the cases in the study and 16.4% of the cases in the control group. Frequency of gum bleeding during tooth brushing did not differ significantly between both groups. Foul breath was reported in 27.8% of the cases in the study and 47.3% of the cases in the control group. The control group demonstrated a significantly higher frequency of foul breath compared to the study group (Table 2).

It was revealed that more than half of the cases in the study (55.6%) and in the control group (60%) applied to their dentists regularly. The groups did not differ significantly in terms of their regular dental visit patterns.Some (35.2%) cases in the study, and control (38.2%) groups visited their dentists more than two years previously. The frequency of dental visits did not differ statistically significantly between both groups. The most recent dental visits in the study, and control groups were made by 44.4% of the cases 6 months-2 years previously, and realized by 47.3% of the cases in the control group 6 months previously. The study group significantly delayed their dental visits compared to the control group (p=0.007) (Table 3).

The dmft values of the cases in the study, and control groups were 4.06±4.29, and 5.79±2.87, respectively. The control group exhibited a significantly greater dmft score compared to the study group (p=0.006) (Table 4). The DMFT values in the study, and control groups were 1.46±2.16, and 2.46±3.05, respectively. The DMFT index appeared greater in the control group; without any statistically significant intergroup difference. The GI values in the study and the control groups were 1.44±0.54, and 1.54±0.54, respectively. Both groups exhibited similar GI values, without any statistically significant intergroup difference. The PI values in the study, and the control groups were 1.80±0.79, and 2.20±0.75, respectively. The PI was significantly elevated in the control group relative to the study group (p=0.006) (Table 4).

**Table 2. Distribution of oral and dental care behaviors according to groups**

| | **Study group** | | **Control group** | | | |
|---|---|---|---|---|---|---|
| | **n** | **%** | **n** | **%** | **$X^2$** | **p** |
| **Tooth brushing** | | | | | 0.057 | 0.810 |
| Yes | 47 | 87.0 | 47 | 85.5 | | |
| No | 7 | 13.0 | 8 | 14.5 | | |
| **Tooth brushing frequency** | | | | | 2.146 | 0.342 |
| Rarely | 20 | 42.6 | 20 | 42.6 | | |
| Once a day | 16 | 34.0 | 21 | 44.7 | | |
| Twice a day | 11 | 23.4 | 6 | 12.8 | | |
| **Toothbrush type** | | | | | 2.063 | 0.356 |
| Manual | 40 | 74.1 | 45 | 81.8 | | |
| Electric | 4 | 7.4 | 5 | 9.1 | | |
| Manual ve electric | 10 | 18.5 | 5 | 9.1 | | |
| **Gum bleeding while brushing teeth** | | | | | 0.050 | 0.824 |
| Yes | 8 | 14.8 | 9 | 16.4 | | |
| No | 46 | 85.2 | 46 | 83.6 | | |
| **Interdental care** | | | | | 1.945 | 0.163 |
| Yes (irregular) | 4 | 7.4 | 1 | 1.8 | | |
| No | 50 | 92.6 | 54 | 98.2 | | |
| **Oral malodor** | | | | | 4.413 | **0.036*** |
| Yes | 15 | 27.8 | 26 | 47.3 | | |
| No | 39 | 72.2 | 29 | 52.7 | | |

*: p<0.05

| Table 3. Distribution and comparison of behaviors related to dental check-ups and examinations according to groups | | | | | | |
|---|---|---|---|---|---|---|
| | Study group | | Control group | | | |
| | n | % | n | % | $X^2$ | p |
| **Regular dental visit** | | | | | 0.221 | 0.639 |
| Yes | 30 | 55.6 | 33 | 60.0 | | |
| No | 24 | 44.4 | 22 | 40.0 | | |
| **Frequency of dental visits** | | | | | 1.025 | 0.795 |
| Every 6 months | 6 | 11.1 | 9 | 16.4 | | |
| Once a year | 17 | 31.5 | 14 | 25.5 | | |
| Once every two years | 12 | 22.2 | 11 | 20.0 | | |
| Less than once every two years | 19 | 35.2 | 21 | 38.2 | | |
| **Last dental visit** | | | | | 12.214 | **0.007*** |
| 0-6 months | 9 | 16.7 | 26 | 47.3 | | |
| 6 months-2 years | 24 | 44.4 | 14 | 25.5 | | |
| More than 2 years | 6 | 11.1 | 3 | 5.5 | | |
| Never | 15 | 27.8 | 12 | 21.8 | | |
| *: p<0.05 | | | | | | |

| Table 4. Comparison of DMFT, dmft, gingival index, and plaque index values according to groups | | | | |
|---|---|---|---|---|
| | Study group | Control group | Total | |
| **Variable** | $\overline{X}$±SD | $\overline{X}$±SD | $\overline{X}$±SD | p |
| dmft | 4.06±4.29 | 5.79±2.87 | 4.90±3.75 | **0.006*** |
| DMFT | 1.46±2.16 | 2.46±3.05 | 1.90±2.69 | 0.124 |
| GI | 1.44±0.54 | 1.54±0.54 | 1.49±0.54 | 0.305 |
| PI | 1.80±0.79 | 2.20±0.75 | 2.00±0.79 | **0.006*** |
| *: p<0.05, DMFT/dmft: Decayed, missing, filled teeth, SD: Standard deviation | | | | |

## DISCUSSION

The results of studies on the oral and dental health of children with hemophilia show differences between countries. Some studies have found that the oral and dental health status of hemophiliac patients are better compared to healthy patients[10,15-19], while others have reported poorer results[8,20-24]. There are also studies that report no difference between the groups[7,25].

In our study, the number of hemophilia A patients (45 patients, 83.3%) was five times higher than the number of hemophilia B patients (9 patients, 16.7%). When similar studies and literature on hemophilia were reviewed, it was reported that the prevalence of hemophilia A is higher than that of hemophilia B, which is consistent with the findings of our study[8,11,22,26-28].

Dental caries is recognized as one of the most prevalent chronic health problems affecting children worldwide[29]. In the study by Nagaveni et al.[18] compared to the control group, the study group exhibited decreased levels in both DMFT and dmft scores. When oral hygiene was compared, it was observed that children with hemophilia had better oral hygiene, which was in line with the decay status. In the studies by Sonbol et al.[16] the frequency of tooth decay was significantly lower in the hemophilia group. In a study conducted in Iran, the DMFT/S values were significantly lower in the hemophilia group[10]. In the studies by Zaliuniene et al.[30] better oral health, fewer number of primary tooth caries and lower need for dental treatment were observed in hemophilia patients. However, when permanent teeth were compared, no significant difference was found. In our research, consistent with previous findings, the study group exhibited a significantly lower dmft score compared to the control group. Although the control group showed a higher DMFT value, the difference between the groups did not reach statistical significance. In light of the data obtained in this study, it is thought that parents of children with hemophilia pay more

attention to their children's oral health care, as they are aware of the potential risks that may arise during dental treatment. Unlike our study, in the studies by Kabil et al.[20] the DMFT and dmft values were found to be significantly higher in hemophilia patients compared to the control group. In another study, the DMFT/dmft values were higher among children with hemophilia. They suggested that the higher rates of dental caries in hemophilic children could be due to high sugar consumption and inadequate oral hygiene habits[21].

In many studies, no meaningful differences between the groups with and without hemophilia regarding gum health has been found, similar to our study[16,19,31]. Our findings showed elevated GI values in the control group, though the difference was not statistically meaningful. In contrast to the present study, in the study by Alpkılıç et al.[32] the GI values of hemophilic patients were found to be statistically significantly greater than those of the control group.

Some previous research studies have indicated that the PI values did not differ significantly between groups[19,31,33]. However, our results showed a significantly increased PI value in the control group relative to the study group. In the study by Sonbol et al.[16] which included 38 hemophilic patients and 30 healthy children, similar to our study, the PI for permanent dentition was found to be significantly higher in the control group. There was no notable intergroup variation regarding gingivitis. Unlike our study, the study by Salem et al.[34] found that PI values were significantly higher in the hemophilia group. In the study by Babu et al.[21] statistical analysis revealed higher GI values in hemophilic children. No difference was found in PI values between both groups. Another study reported significantly elevated index scores for dental debris, calculus, and gingival inflammation in hemophilic patients. The same study also found that the oral hygiene status of hemophilia patients was lower compared to healthy individuals[35].

In a study conducted in India, it was found that only 23% of patients in the hemophilia group, whereas 46% of the cases in the healthy group had the habit of brushing their teeth "twice a day," with a statistically significant difference between both groups[22]. In Güler's[36] study, where brushing habits were evaluated, 72% of the children with hemophilia, whereas only 42% of healthy children responded that they had brushed their teeth "more than once a day" or "once a day". It was concluded that children with hemophilia and their families paid more attention to brushing their teeth compared to the healthy group. In our study, 23.4% of patients in the hemophilia group whereas 12.8% of the cases in the control group brushed their teeth "twice a day". There was no statistically significant difference in terms of brushing frequencies between both groups. It has been reported that patients with bleeding disorders tend to avoid brushing their teeth due to the fear of gum bleeding[34]. Consistent with our findings, Czajkowska et al.[24] reported the incidence of bleeding while brushing teeth was greater among hemophilic patients, yet this difference was not statistically significant. Consistent with this data, our study observed no significant intergroup difference in frequencies of bleeding during brushing. Children with hemophilia and their parents should be educated that, with proper treatment tailored to the patient and in cooperation with medical professionals, the risk of bleeding can be minimized. In a study conducted in Italy, dental treatment data from three hemophilia centers over 10 years were examined, and it was found that bleeding complications were very low. The study concluded that dental treatments, under proper conditions, are highly effective and safe[37].

### Study Limitations

Our study was conducted with a limited number of hemophiliac patients in a specific age group. Subsequent researches should aim to expand the sample size, investigate different age groups, and evaluate oral and dental health findings by reaching a larger population of children with hemophilia. Oral and dental health assessments in this study were conducted through a single clinical examination and did not include follow-up evaluations. This is another limitation of our study in terms of assessing changes in the children's oral health over time or the effectiveness of oral hygiene education provided. Future research conducted over longer time periods and supported by periodic follow-up examinations would provide better insight into the improvement of the oral and dental health of children with hemophilia over time and allow for more comprehensive comparisons with healthy control groups.

## CONCLUSION

Our study population consisted of a study group of children with hemophilia and a control group of healthy children. In our study, the primary dentition dmft scores and PI values were found to be significantly higher in the control group compared to the study group. There was no notable difference in oral hygiene practices between the groups.

The preservation of dental health in patients with hemophilia is important, especially to prevent the risk of complications that may arise during comprehensive dental treatments and to avoid the costs of premedication taken before treatment. This approach should be our primary goal.

Studies can be conducted to increase the knowledge of dentists regarding the approach to patients with hemophilia. Additionally, pre-treatment consultations should be made in collaboration with hematology specialists, and patient management should be planned using a multidisciplinary approach.

Providing hemophiliac children and their families trustworthy information can help raise awareness about proper oral health practices, the necessity of regular dental check-ups, and the value of preventive treatments.

In conclusion, the management of oral and dental health in patients with hemophilia should be initiated at an early age and continued with regular follow-ups. This approach will help minimize the need for advanced dental interventions in individuals with hemophilia.

### Ethics

**Ethics Committee Approval:** Our study was ethically approved by the Ege University Medical Research Ethics Committee with the decision dated April 25, 2024, and numbered 24-4.1T/62.

**Informed Consent:** Informed consent was obtained from all individual participants included in the study.

### Footnotes

### Author Contributions

Surgical and Medical Practices: M.D., F.E., Concept: M.D., F.E., M.C.U., K.K., Design: M.D., F.E., M.C.U., K.K., Data Collection or Processing: M.D., M.C.U., K.K., Analysis or Interpretation: M.D., F.E., M.C.U., K.K., Literature Search: M.D., Writing: M.D., F.E.

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