Case-Control Study to Evaluate the Oral Hygiene Habits in 31 Children with Hemophilia in Bulgaria Using the Oral Health Impact Profile-14 (OHIP-14) Questionnaire

Background: For patients with hereditary coagulopathies such as hemophilia, the most common dental problem is spontaneous oral bleeding caused by daily oral hygiene habits like tooth brushing. Because of fear of bleeding, children with hemophilia often skip routine oral hygiene procedures, which can lead to dental caries and periodontal diseases. This case control study aimed to evaluate the oral hygiene awareness and hygiene practices of children with hemophilia from 6 to 18 years of age in Bulgaria.

Material/Methods: This study was conducted over 4 years and included 31 patients (children). A sociological approach was used, with direct individual questionnaires for self-assessment of dental habits. Statistical analyses were performed using SPSS.

Results: There was no significant difference between groups in methods of oral hygiene. The duration of oral hygiene procedures was 1 min (n=5, 16.13±6.61%) or 2 min (n=13, 41.94±8.86%), which was considered insufficient for proper oral hygiene. Children who had received oral hygiene advice by a dentist brushed their teeth significantly longer (n=9, 69.23±12.80%) than those who had not. There was a considerable difference between the reasons for the last dental visit between the groups of children (χ²=5.18, P<0.05).

Conclusions: Children with hemophilia have high awareness of oral hygiene methods; however, more attention should be focused on additional individual educational methods at the dental office. Frequent professional and self-assessment of the oral hygiene routine of children with hemophilia could identify the factors compromising dental status in this vulnerable population.

Keywords: Dental D • Hygiene • Dental Care for Children

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Background

Oral health deterioration is a significant problem. Oral health also has an effect on chronic diseases, such as hereditary coagulopathies. The compartmentalization of viewing dental diseases as separate from the general diseases must stop because oral health affects general health by causing considerable pain and suffering. Impaired dental health causes changes in people’s daily diet, speech, quality of life, and well-being [1-3].

At present, few studies have evaluated the relationship between the dental status of people with hemophilia and their chronic disease [4-10]. Limited social activity and compromised oral health has been shown to exist in cases of comorbidity [5,9]. For these patients, the most common dental problem is spontaneous oral bleeding caused by daily oral hygiene habits, such as tooth brushing [4,5]. Fear of inducing bleeding in the oral cavity is the reason many people underestimate their oral hygiene care [4,5]. Once a dental disease is present, dental care for patients with hemophilia can become more complicated and expensive. Chronic diseases discovered in early childhood, including hemophilia, have been reported as an additional risk factor for the development of dental diseases in children [7]. Controversy exists about whether there is better oral health in the deciduous dentition of children with hemophilia or in control groups because patients with hemophilia tend to be from families with lower social status, which is a well-known determinant of higher cariogenicity, especially in young children [8,10]. However, differences in oral status decrease and even become insignificant when comparing groups of adults [11]. Because dental caries and periodontitis are diseases in which health determinants accumulate over time, such as various bacteria and poor oral hygiene techniques, in order to clarify the differences in oral status in children and adults, future prospective studies with larger study groups are needed.

Self-assessment of the oral hygiene status of children with hemophilia could serve as a basis for the prevention of dental diseases, such as tooth decay and periodontal diseases, and can help prevent complications during dental treatment [12]. The habits of children copy those of their parents, so an evaluation of the family environment is essential. When the parents’ awareness is evaluated, it is easier to understand the oral hygiene practices of the children, too. Oral hygiene knowledge and practices directed toward children with coagulopathies through educational programs could be maintained through the whole life, starting from an early age. With basic guidance, motivation, and preventive care by primary dental professionals, it would be easier for the children to become autonomous in their daily activities [13].

Therefore, this case control study aimed to evaluate the oral hygiene awareness and habits of 31 children aged between 6 to 18 years with hemophilia in Bulgaria.

Material and Methods

Design

This case control study was conducted in Bulgaria over a 4-year period, from 2017 to 2020. Data was collected from 139 children, of which 31 had hemophilia and 108 did not. Children from the control group were matched with the those in the hemophilia group based on sex, age, and place of residence [8]. The socio-demographical information about children and parents was collected through self-administered questionnaires, which were developed in the local language, Bulgarian, for self-assessment of their dental hygiene habits. Data were collected from January 2017 to June 2020. The participants evaluated were 31 children with hemophilia, 108 children without hemophilia, and 33 parents of children with hemophilia.

The use of questionnaires provided information about the demographic characteristics of the participants, their knowledge of oral hygiene, oral hygiene habits, and self-assessment of their oral status. Some questions had options for multiple answers.

The study was conducted at the following: a summer camp for parents of children with hemophilia from 0 to 3 years old in Strelcha (Bulgaria), summer rehabilitation camp for children with hemophilia between 7 and 14 years old in Kranevo (Bulgaria), individual and group dental practices, and dental centers in Bulgaria. By means of statistical analysis, children were allocated to 4 age groups and parents to 5, based on age-dependent differences. The education level of the study participants was classified into 4 categories: pre-school, primary (grades 1-4), lower secondary (grades 5-7), and upper secondary (grades 8-12), with an additional undergraduate category for parents (with postgraduate diploma-bachelor or higher degree). The distribution of children with hemophilia by residence in Bulgaria was based on the classification of the Nomenclature of Territorial Units for Statistics, dividing the location of residences into 6 regions. The study protocol was approved by the Commission on Scientific Ethics at the Medical University of Plovdiv, Bulgaria (protocol no. 6/07.10.2021). The data presented for approval included the application form for consideration for ethical expert assessment of scientific research and projects including human subject research, questionnaire forms for the groups of children with and without hemophilia and parents of children with hemophilia, informed consent form for participation in the study from parents regarding their children, declaration of agreement to comply with the Declaration of Helsinki and its principles for medical research involving human subjects, and design and summary of the scientific research.

Statistical Analysis

Normally distributed variables are presented as mean±standard deviation. Statistical hypothesis tests for difference of means
were calculated using the t test and non-parametric Mann-Whitney test. All statistical analyses were performed using IBM SPSS Statistics v.25 (IBM Corp, Armonk, NY, USA). Values of $P<0.05$ were considered statistically significant. The Fisher’s exact test and chi-square test were used for the analyses.

**Results**

**Socio-Demographic Characteristics of the Groups**

The 31 surveyed children with hemophilia were between 7 and 18 years of age (mean 11.55±0.58) and, owing to the specificity of the disease, were only boys (Table 1). There were 30 children with hemophilia A and 1 with hemophilia B. The children in the control group without hemophilia were aged between 5 and 18 years (mean 12.30±0.30). The largest proportions of respondents were children with primary education ($n=13$, 41.90±2.95%), which was also influenced by the location of the surveys at the children’s summer camp.

| Characteristics          | Children with hemophilia | Control group | Parents of children with hemophilia |
|--------------------------|--------------------------|---------------|-------------------------------------|
|                          | n   | %   | Sp  | n   | %   | Sp  | n   | %   | Sp  |
| Sex                      |     |     |     |     |     |     |     |     |     |
| Male                     | 31  | 100.0 | –  | 108 | 100.0 | –  | 11  | 33.33 | 8.21 |
| Female                   | 0   | 0.00  | –  | 0   | 0.00  | –  | 22  | 66.67 | 8.21 |
| Region distribution      |     |     |     |     |     |     |     |     |     |
| North Western            | 16  | 51.61 | 8.98 | 1   | 0.92  | 0.92 | 14  | 42.42 | 8.60 |
| North Eastern            | 7   | 22.58 | 7.51 | 53  | 49.07 | 4.81 | 7   | 21.21 | 7.12 |
| South Eastern            | 3   | 9.68  | 5.31 | 13  | 12.04 | 3.13 | 3   | 9.09  | 5.00 |
| South Central            | 1   | 3.23  | 3.17 | 18  | 16.67 | 3.59 | 2   | 6.06  | 4.15 |
| South Western            | 4   | 12.90 | 6.02 | 10  | 9.26  | 2.79 | 7   | 21.21 | 7.12 |
| North Central            | 0   | 0.00  | –  | 13  | 12.04 | 3.13 | 0   | 0.00  | –   |
| Education                |     |     |     |     |     |     |     |     |     |
| Preschool                | 0   | 0.00  | –  | 3   | 2.78  | 0.84 | 0   | 0.00  | –   |
| Primary                  | 13  | 41.94 | 8.86 | 30  | 27.78 | 2.21 | 1   | 3.03  | 2.98 |
| Lower secondary          | 9   | 29.03 | 8.15 | 37  | 34.26 | 2.33 | 1   | 3.03  | 2.98 |
| Upper secondary          | 9   | 29.03 | 8.15 | 38  | 35.19 | 2.34 | 12  | 36.36 | 8.37 |
| Undergraduate degree      | 0   | 0.00  | –  | 0   | 0.00  | –  | 19  | 57.58 | 8.60 |

n – total number; % – percentage; Sp – standard error of percentage.

There was no statistically significant difference between level of education and residence by region among the children with hemophilia ($P>0.05$). The proportions of children with and without hemophilia by regions in Bulgaria were similar, with more from urban areas. The largest proportion of children with hemophilia were in the regions of Sofia ($n=12$, 38.71±8.75%), Plovdiv ($n=5$, 16.13±6.61%), and Pleven ($n=2$, 6.45±4.41%). The distribution of children and parents by regions was similar. Moreover, parents motivated their children to participate in the same social events that they participated in.

**Dental Habits**

Children from both groups confirmed that they used a toothbrush and toothpaste in performing oral hygiene procedures (Table 2). The respondents from all groups were aware of adjunctive oral hygiene methods, but not all were familiar with the correct techniques for dental plaque removal. However, there was no significant difference between the groups in methods used for oral hygiene ($P>0.05$). A high preference for gum chewing was observed in children, whereas none of the adult respondents reported chewing gum. Adults preferred
adjunctive methods, such as using toothpicks, which were not used in the group of children with hemophilia. When asked to explain how to use toothpicks, children were unsure. They were usually hesitant to cause gingival bleeding after cleaning the interdental space. Interdental floss was used as an alternative oral hygiene method in about 20% of the respondents from families with hemophilia. Children explained that the interdental brush was easier to operate than interdental floss.

The duration of oral hygiene procedures was about 1 min (n=5, 16.13±6.61%) or 2 min (n=13, 41.94±8.86%), which is considered insufficient for proper oral hygiene (Table 3). There was a smaller proportion of children with hemophilia who brushed their teeth for 3 min or more. Longer toothbrushing duration was associated with other hygiene habits. For example, there was an association between longer toothbrushing and the use of dental fluoride in different forms ($\chi^2=6.64, P<0.05$). Children in both groups used fluoride supplements, but children with hemophilia were more likely to consider it as a method for caries prevention.

Frequent oral hygiene habits lead to a significant reduction of anaerobic microorganisms, which cause periodontal diseases in the mouth cavity and create the need to visit the dentist. The proportions of dental fluoride in different forms were $21.85\%$ ($n=5, 16.13\%\pm 6.61\%$) or for a dental treatment ($n=5, 16.13\%\pm 6.61\%$). A lack of knowledge about the reasons for dental visits was a common response in children with hemophilia ($n=11, 35.48\%\pm 8.59\%$). We interpreted this as a denial of a dental visit, or as there being no reason for visiting. The real reasons could be analyzed better during an interview with every child, which is a recommendation for future studies. It is important for the long-term outcomes of oral health improvement in children with comorbidities for the dentist to achieve a high degree of cooperation with the children. Receiving professional dental advice was associated with the duration of toothbrushing in children with hemophilia ($\chi^2=6.85, P<0.05$). However, most of the patients with hemophilia responded that they had not received any professional help for oral hygiene improvement ($n=18, 58.06\%\pm 8.86\%$). More than half of the children in the control group were educated about correct oral hygiene techniques by a dental professional ($n=68, 62.96\%\pm 4.65\%$). In the present study, children who received oral hygiene advice by a dentist brushed their teeth significantly longer ($n=9, 69.23\%\pm 12.80\%$) than those who had not received advice, which was also found in previous studies [8,9].

In addition to professional dental motivation in clinics, which was provided periodically, motivation for oral hygiene improvement was associated with family education. Most of the children in both groups had received advice from their parents regarding oral hygiene. Nevertheless, more than half of the children with hemophilia had experienced bleeding episodes from the oral cavity (Figure 1).

### Table 2. Methods of oral hygiene.

| Characteristics                  | Children with hemophilia | Control group | Parents of children with hemophilia |
|----------------------------------|--------------------------|---------------|------------------------------------|
|                                  | n | %     | Sp | n | %     | Sp | n | %    |
| Use of oral hygiene methods      |   |       |    |   |       |    |   |      |
| Toothbrush                       | 31 | 100.00 | –  | 108 | 100.00 | –  | 33 | 100.00 | –  |
| Toothpaste                       | 31 | 100.00 | –  | 108 | 100.00 | –  | 31 | 93.94 | 4.15 |
| Mouthwash                        | 4  | 12.90 | 6.02 | 20  | 18.52 | 3.74 | 18  | 45.45 | 8.67 |
| Interdental brush                | 7  | 20.00 | 7.30 | 27  | 25.00 | 4.17 | 12  | 36.36 | 8.37 |
| Toothpick                        | 1  | 3.23  | 3.17 | 34  | 31.48 | 4.47 | 28  | 15.15 | 6.24 |
| Chewing gum without sugar        | 6  | 19.35 | 7.10 | 17  | 15.74 | 3.50 | 0   | 0.00  | –   |

$n$ – total number; $%$ – percentage; $Sp$ – standard error of percentage.

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Discussion

Several epidemiological studies have been conducted in Bulgaria to register people with coagulopathies, of which there are a total of 610 patients with hemophilia A (7 per 100,000 people) and 85 patients with hemophilia B (0.7 per 100,000 people). Evaluation of the oral status of such patients was based on groups with a small number of patients [14].

The sample size of similar studies included the observation of children only, or both adults and children with hemophilia. In the present study, the focus was to assess only the pediatric population with respect to the habits of the family environment. This study aimed to evaluate and compare the oral hygiene habits among children with hemophilia, their parents, and age-matched controls. Most studies explaining the health behavior of children with hemophilia included boys only, with few including female participants.

Table 3. Oral-health related awareness among children with hemophilia and parents.

| Children with hemophilia | Control group | Parents of children with hemophilia | Test |
|--------------------------|---------------|-------------------------------------|------|
| n | % | Sp | n | % | Sp | n | % | Sp | χ² | P |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Dental visits during the last year** | | | | | | | | | | |
| 1-2 times | 15 | 48.39 | 8.98 | 46 | 42.59 | 7.06 | 19 | 57.58 | 8.60 | 13.30 |
| More than 2 times | 8 | 25.81 | 7.86 | 53 | 49.07 | 7.14 | 8 | 24.24 | 7.46 | |
| I don’t remember/I haven’t visited | 8 | 25.81 | 7.86 | 9 | 8.33 | 3.95 | 6 | 18.18 | 6.71 | |
| **Reason for the last dental visit** | | | | | | | | | | 22.10 |
| Toothache/pain in the gums | 5 | 16.13 | 6.61 | 30 | 27.78 | 4.31 | 8 | 24.24 | 7.46 | |
| Treatment prophylactic | 5 | 16.13 | 6.61 | 41 | 37.96 | 4.67 | 11 | 33.33 | 8.21 | |
| Examination | 10 | 32.26 | 8.40 | 30 | 27.78 | 4.31 | 11 | 33.33 | 8.21 | |
| I don’t know/I don’t remember | 11 | 35.48 | 8.59 | 7 | 6.48 | 2.37 | 3 | 9.09 | 5.00 | |
| **Frequency of toothbrushing** | | | | | | | | | | 0.61 |
| Once per day | 13 | 41.94 | 8.86 | 22 | 20.37 | 3.88 | 6 | 18.18 | 6.71 | |
| 2 or more times per day | 23 | 74.19 | 7.89 | 86 | 79.63 | 3.88 | 27 | 81.82 | 6.71 | |
| **Duration of toothbrushing** | | | | | | | | | | 3.37 |
| Less than 3 min | 18 | 58.06 | 8.86 | 81 | 75.00 | 4.17 | 0 | 0.00 | – | |
| 3 min or more | 13 | 41.94 | 8.86 | 27 | 25.00 | 4.17 | 0 | 0.00 | – | |
| **Received oral hygiene advice from parents** | | | | | | | | | | 0.02 |
| No | 13 | 41.94 | 8.86 | 46 | 42.59 | 4.76 | 0 | 0.00 | – | |
| Yes | 18 | 58.06 | 8.86 | 62 | 57.41 | 4.76 | 0 | 0.00 | – | |
| **Received oral hygiene advice from a dentist** | | | | | | | | | | 1.44 |
| No | 18 | 58.06 | 8.86 | 40 | 37.04 | 4.65 | 0 | 0.00 | – | |
| Yes | 13 | 41.94 | 8.86 | 68 | 62.96 | 4.65 | 0 | 0.00 | – | |
| **Use of fluoride** | | | | | | | | | | 3.20 |
| No | 11 | 35.48 | 8.59 | 58 | 53.70 | 4.80 | 0 | 0.00 | – | |
| Yes | 20 | 64.52 | 8.59 | 50 | 46.30 | 4.80 | 0 | 0.00 | – | |

n – total number; % – percentage; χ² – chi-square value; P – significance value; Sp – standard error of percentage.
In providing quality dental services for people with coagulopathies, despite the macro-economic stability, Bulgaria still strives to attain the EU standards in medical development and oral health policy. Until now, there are no official dental protocols for the treatment of people with bleeding disorders in Bulgaria. There is no published research conducted on the dental status and dental treatment needs of children with hemophilia in Bulgaria. Only after a detailed professional evaluation of the dental needs of children with disabilities can recommendations be implemented in the programs for oral health improvement. However, reducing inequality in the access to primary and specialized medical services and dental care for children with chronic diseases is within the key recommendations of UNICEF, which is among the main medically and socially justified policies, defining the priority in oral health improvement, including for children with hemophilia [15].

The latest active oral health program in Bulgaria including the pediatric population is the National Program for Prophylaxis of Oral Diseases in Children Ages 0 to 18 Years, from 2021-2026, which continues the national program from 2014-2020. In its focus is proper oral hygiene among children with the use of toothbrushes, toothpastes with fluoride, and dental floss. It is the only national and regional activity established regarding the oral hygiene improvement of children. All children and their parents (guardians) should be trained and educated in all basic prophylactic methods. As a result, they should acquire adequate knowledge and skills as part of their value system [16].

After a detailed study and comparison of oral hygiene status in children with and without hemophilia, researchers found conflicting results [8]. Age is associated with the ability of individuals to perform oral hygiene procedures [12-14]. This is because children with hemophilia have a high awareness of the options for prophylaxis as prevention of life-threatening dental conditions. Some authors found poor oral hygiene status in the hemophilia groups (P<0.05) [17-19]. More than three-quarters of all children needed dental procedures and more than half of them needed dental prophylaxis. It could be because most of the patients with hemophilia (n=36, 72%) performed oral hygiene habits once a day, while few of them (n=9, 18%) brushed their teeth twice a day. Only around 10% skip tooth brushing because of a fear of gingival bleeding. Moreover, the better the periodontal status is, the lower the severity of oral cavity bleeding is in people with hemophilia. The overall oral hygiene status in children with hemophilia was poorer than that of age-matched controls, but did not show a significant difference. In a study by Parvae et al, almost a third of the participants with hemophilia reported that they do not brush their teeth (n=28, 31.5%, P<0.05), and a majority brush teeth once per day (n=23, 25.8%, P<0.05) or once or twice per week (n=23, 25.8%, P<0.05). The study was done in comparison to the healthy children, in which indices were lower, but not significantly [18].

The same tendency was observed in the frequency of brushing habits between children with hemophilia (n=100, 23%) and a control group (n=100, 46%) in India, with more brushing in the healthy group (P<0.05). These results differ from the study in Bulgaria, as in the Balkans there are clear similarities between the groups in practicing this habit [14,20]. Other researchers have shown that oral hygiene practices were similar between a hemophilia group and control group [11,17,18]; most children performed oral hygiene habits twice per day, and there was no significant difference in oral hygiene practices between the groups. It is due to the interdisciplinary approach in Western European countries, where dentists have been an essential part of the treating team of people with hemophilia for decades [17]. It is worth making additional efforts to motivate children with hereditary conditions to maintain good oral hygiene before their dental status is compromised. Motivational methods give a considerable advantage when started from an early age [13].

A main principle of dental prevention for people with hemophilia is that everyone should be given the opportunity for self-assessment of the risk factors of early childhood caries. Standard dental prophylaxis is suitable for children with a low risk level of caries, while specific prophylaxis is used in children with an increased caries risk or in those assigned to a category with a high risk owing to other factors (eg, medical conditions) [3]. Teaching patients from an early age about the methods of maintaining personal oral hygiene, such as the correct toothbrushing technique, should be implemented in educational programs for children with hemophilia [4]. Health information and education for risk groups is important because irregular oral hygiene leads to significant bleeding. If toothbrushing causes bleeding, a temporary solution is mouth rinsing after each meal and improvement of the personal hygiene technique. Sufficient oral hygiene reduces the

![Figure 1. Experience of bleeding episode connected with oral tissues.](image-url)
necessity of invasive dental treatment and the number of dental appointments [5,7]. Regular self-assessment and professional assessment of the oral status of families with hemophilia make it possible to evaluate not only the quality of life, but also whether the treatment provided to each patient is appropriate for them.

Educating patients about correct preventive methods has significant health benefits, which are proven over time. Fluoride has been used in dentistry for nearly a century and is considered an essential protective factor in decreasing tooth decay. However, to achieve the maximum benefit of fluoride prophylaxis, it should be used in combination with different types of methods for personal oral hygiene. No significant difference was found in the fluoride used during oral hygiene or fissure sealant application in the literature [19,20].

Our study found a statistically significant difference between the reasons for dental visits between both groups of children, which was also found by other authors. However, a study in Malaysia found that there was a difference between groups (P<0.05) [17]. Dental visits in an ambulatory setting are generally for prophylaxis or scheduled in advance [17,18]. On a regional level in Bulgaria, children with hemophilia are referred mainly for dental care to the Dental Faculties of Medical universities, working in collaboration with the hemophilia team. It could be an insurmountable issue if a referral is for a child living in an area far from the Dental Faculties, regional hemophilia treating center, or specialized dental practice. The distribution of families mainly in the big cities is a consequence of today’s move toward urbanization. Many families from rural areas move to urban areas in search of employment opportunities, modernization, and better quality of life. For this population, the access to dental care is easier [15,16], and patients with hereditary coagulopathies who are looking for professional dental care have different options to choose from. Those living in the rural areas are either transferred by the general dentist to a specialist or oral or maxillofacial surgeon, or they prefer to visit pediatric dental practices for their children. The whole period between the first dental visit and the end of the treatment could be delayed or complicated by factors such as distance, transport, expenses, and procrastination [17].

Children’s behavior largely matches that of their parents; therefore, the family environment of children with hemophilia is a key to understand and build oral hygiene habits in the young generation. A significant difference in overall oral health has been seen between some groups of children with and without hemophilia. As a result of the positive impact of the family habits implemented in children, the overall dental health of children with hemophilia was evaluated as better than that of children without hemophilia [19]. Although this may partly be explained by genetic factors, environmental factors, such as established preventive dental routines at home, also contribute to this finding. Dental health culture should be extended from an individual to group level so that all family members receive the proper education. In practice, it is difficult for families with hemophilia to implement the recommendations of dental professionals at home. The focus should be in the control of children performing their daily dental routine so that they become autonomous.

Further, incorporating dental care as a part of the specialized hemophilia centers may enhance confidence and acceptance of the dental care and help to decrease the risk of bleeding during dental appointments [21,22].

Even though different national and international protocols and programs have considered dental treatment of children with hemophilia, there are still no strict recommendations or absolute statements in dental guidelines for Bulgaria. It may be because most studies showed diverse attitudes of medical and dental professionals to the treatment approach regarding this topic [23].

**Conclusions**

The findings of this study of children with hemophilia in Bulgaria support those of previous studies from other countries and support the need to improve oral hygiene practices in this group of patients. Children with hemophilia have a high awareness of oral hygiene methods, but more attention should be focused on additional individually based education at the dental office. More frequent self-assessment of children with hemophilia and further evaluation of their oral hygiene routine could identify the factors compromising the dental status in this vulnerable population.

**Declaration of Figures’ Authenticity**

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.
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