



Assessment of dental anxiety and fear levels of pediatric patients diagnosed with familial mediterranean fever

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Abstract

Aim: Familial Mediterranean Fever (FMF) is an autoinflammatory illness featured by recurrent fever, arthritis, and inflammation of the serous membranes, with an autosomal recessive with recurrent attacks and is a disease that begins in childhood and lasts a lifetime. In this study, it was aimed to evaluate the correlation between disease activity and dental fear-anxiety, and dental behaviour level in children with FMF who had pain and hospital experience in early childhood.

Materials and Methods: The study included 207 children ranging in age from 5 to 15 years; 93 patients with FMF and 114 healthy controls. Dental anxiety and dental fear were scored using Frankl Scale and Children's Fear Survey Schedule-Dental Subscale (CFSS-DS). Age, sex, disease severity activity, and duration of illness were examined.

Results: There was not statistical difference in dental anxiety and fear between the FMF and healthy group ($p > .05$). However, there was a statistically significant difference in the effect of disease severity activity on the CFSS-DS. The anxiety and fear in FMF patients was similar to healthy children, which was low.

Conclusion: Dental anxiety and fear level in FMF patients was similar to healthy children, which was low. It is necessary to avoid the idea that all chronic diseases will cause dental anxiety and fear, and they should be evaluated according to the nature of the chronic disease.



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Introduction

Familial Mediterranean Fever (FMF) is an autoinflammatory disease characterized by recurrent fever, inflammation of the serous membranes, peritonitis, and arthritis with an autosomal recessive transition with recurrent attacks [1], FMF is frequently observed in societies of Eastern Mediterranean origin (Jewish, Arab, Armenian, Greek, and Turkish) [2-4]. This disease begins in childhood and lasts a lifetime, requiring treatment throughout life. Although there are regional differences, the prevalence of the disease in Turkey varies from 1/400 to 1/1,000 and the carriage rate is known to be 1/5. In Turkey, unlike other countries, it is more frequently observed in individuals with origins in Central Anatolia, Black Sea, and Eastern Anatolia compared to those living on the Mediterranean coast [2], Tokat province is also among the provinces where FMF is most frequently observed.

Pain and high fever are the most important symptoms

of FMF and are among the criteria required for diagnosis. Nonspecific findings, such as malaise, fatigue, myalgia, arthralgia, headache, back, and low back pain are also commonly accompanied by high fever [5]. Abdominal pain caused by inflammation of the peritoneal membrane is the most common clinical complaint in FMF. While joint findings are frequently observed in hips and knees, they can also be observed in other joints, such as temporomandibular joint, shoulder, ankle and sternoclavicular joints [6]. Oral findings observed in FMF patients are recurrent oral aphthae and periodontitis. Oral aphthae are rare mucocutaneous findings and may accompany attacks [7].

While chronic diseases cause physical and mental difficulties in a child, they can affect a family, both mentally and economically and can cause serious adjustment problems and psychiatric disorders [8]. Gortmaker et al. stated that 10% of adolescents with chronic diseases experience behavioural problems, such as social withdrawal, aggression, and depression, higher anxiety levels, and are 1.55 times more frequent psychiatric disorders than children without the chronic disease [9]. Depression and anxiety

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levels are increased in children with chronic disease and their parents [10]. Children and adolescents with chronic diseases also have lower self-esteem, weaker body images than healthy ones, and are more problematic in their psychological health, behaviour, and social cohesion [11].

The Children's Fear Survey Schedule-Dental Subscale (CFSS-DS) covers an assessment consisting of 15 questions about various stages of dental treatment, such as dental treatment and clinical examination, the necessity of keeping the mouth open, the injection dentist's use of rotary instruments. The questions are answered by choosing one of the answer options with a score of 1-5 between fearless and extremely afraid. Total scores range from 15 to 75 [12]. A score of 15-31 points indicated a low level of anxiety, 32-38 points indicated a moderate level of anxiety, and 39 points and above indicated a high level of anxiety.

One of the techniques commonly used in scoring the behaviors is the Frankl Behavior Scale. The Frankl Behavior Scale is based on the observational evaluation and scoring of behaviors by the dentist. The behavior of the child is evaluated by dividing them into four groups. Absolutely negative; refuses treatment, is very afraid, shows extremely negative behaviors and cries. Negative; is maladaptive, reluctant to treatment, and displays some negative behaviors such as resentment. Positive; It is compatible with the dentist, accepts the treatment, but acts cautiously. Absolutely positive; He has a good relationship with the dentist and is cheerful, accepts the treatment and acts interested. Behaviours were scored between 1 and 4 [13].

It was found that an acquired fear of the hospital and boredom was high in children with chronic systemic disorders. The continuous presence of the child in the hospital environment due to ongoing disorders may lead to the development of white coat phobia. Our study aimed to evaluate the relationship between disease severity activity score and dental anxiety-fear levels, and dental behaviour level in children with FMF who had pain and hospital experience in early childhood.

Materials and Methods

This study was planned as a randomized controlled clinical study with 207 children. There were two main groups in this study, FMF group and healthy control group, and all patients were of Turkish origin, from the inner Black Sea Region of Turkey. An individual identification form was used to survey healthy and FMF children to evaluate demographic characteristics, disease severity activity scoring and duration, diagnosis age of the disease, Frankl Behavioural Rating Scale (FBRS), and Children's Fear Survey Schedule-Dental Subscale (CFSS-DS).

Approval of the clinical research ethics committee (Ethics No: 19 KAEK 167) was obtained for the study. This study was managed in Departments of Pediatrics and Pediatric dentistry. The informed consent form was acquired from the parents of the children included in the study. Sample size was calculated at a 80% confidence interval and a significance level of 0.05 and effect size was 0.57, according to the Hollis et al [14]. At the analysis calculated using the G*Power software program (version 3.1.9.6) indicated that a group sample size of 39.

Patients with a diagnosis of FMF who applied between 2019 and 2020 were included in our study. In order to provide randomization in grouping the patients, drawings were made from closed, opaque envelopes. Ninety-three children between the ages of 5-15 years who were admitted to Faculty of Dentistry and had no FMF attack during the acute attack period in the past six months were included in study for dental examination. FMF was diagnosed according to Tel Hashomer criteria by the Department of Pediatrics at Medicine Faculty. Disease severity was determined according to previously published Pras criteria [15]. One hundred-fourteen children without systemic disorders who were admitted to Faculty of Pediatric Dentistry for dental examination and were considered healthy were included as a control group. Only dental examination was performed in FMF and control study groups.

1. Children were not between 5 -15 years old,
2. Children had additional chronic disease in addition to Familial Mediterranean Fever,
3. Children who could not answer or answered the questions too late were not included in the study.

The patient's dental fear-anxiety levels were recorded with Frankl Behavioural Rating Scale and Children's Fear Survey Schedule-Dental Subscale (CFSS-DS Index) by same operator (M.B).

Priori hypothesis

There were hypotheses as follow: The dental anxiety level of children with FMF is similar to the level of dental anxiety of healthy children. The dental fear level of children with FMF is similar to the level of dental fear of healthy children.

Statistical analysis

The data were analysed using SPSS (IBM SPSS Statistics 19, SPSS inc., an IBM Co., Somers, NY) and $p < 0.05$ was considered as statistically significant. Data are expressed as n (%) or mean and standard deviation (SD). Independent sample t-tests were used in comparisons of the FMF and healthy groups. A linear regression analysis, Spearman's rho correlation, was administered to determine the relationship between disease duration and disease severity activity scoring with dental anxiety and dental fear.

Results

FMF patients (n=93) and healthy children (n=114) aged 5-15 years were included. 94 (45.4%) of the patients were female and 113 (54.6%) were male and average age was 10.31 ± 2.82 years (Table 1). There was no statistically significant difference between the two groups in terms of age or sex ($p=0.62$). Disease severity score was computed according to modified Pras scoring system for children. (15) In our study, the mean disease severity score was 1.89. Out of 93 cases, 22 had mild, 59 had moderate, and 12 had severe disease activity (Table 1).

Table 2 shows the distribution of patients' frequency according to the FBRS and CFSS-DS. The FBRS values of the patients in control group and the patients in FMF group were statistically similar ($p = 0.09$). The mean

Table 1. Demographic datas, diagnosis age of the disease and disease severity scores.

	FMF (n=93)	Control (n=114)	p value
Age Mean			
Minimum	5	6	
Maximum	15	13	
Mean ± Standard Deviation	10.55 ± 3.22	9.91±1.97	p=0.62
Sex			
Girl	46	48	p=0.62
Boy	47	66	
FMF diagnostic age	7.75 ± 3.32		
Disease duration	4.47 ± 3.05		
Disease severity activity score	n (%)		
Mild	22 (23.7%)		
Moderate	59 (63.4%)		
Severe	12 (12.9%)		

Table 2. Evaluation of the Frankl Behavioural Rating Scale and Children’s Fear Survey Schedule-Dental Subscale.

	FMF (n=93)	Control (n=114)	χ ²	p value
Frankl				
Definitely negative	1 (1.1%)	0		
Negative	7 (7.5%)	12 (10.5%)		
Positive	22 (23.7%)	46 (40.4%)		
Definitely positive	63 (67.7%)	56 (49.1%)		
Mean Frankl Score	3.58±0.68	3.39±0.67	6.28	0.09
CFSS-DS				
Low	64 (68.8%)	64 (56.1%)		
Moderate	17 (18.3%)	22 (19.3%)		
High	12 (12.9%)	28 (24.6%)		
Mean CFSS-DS	27.90±8.72	31.16±11.37	3.68	0.15

χ²Chi-square test.

Table 3. Regression results for disease severity activity score with Frankl Behavioural Rating Scale and Children’s Fear Survey Schedule-Dental Subscale.

Dependent variable	Independent variable	R ²	β	t	Signature
Frankl	Disease duration	0.04	0.19	1.85	0.07
	Disease severity activity		-0.07	-0.69	0.49
CFSS-DS	Disease duration	0.05	-0.11	-1.06	0.29
	Disease severity activity		-0.21	-2.05	0.04*

*Significant difference according to the linear regression analysis, Spearman’s rho correlation.

FBRS score was 3.58 (SD 0.68) for the FMF group and 3.39 (SD 0.67) for control group (Table 2). There were not significant differences in the dental anxiety between FMF and control groups. Mean CFSS-DS score was 27.90 (SD 8.72) for FMF group and 31.16 (SD 11.37) for control group. Dental fears of FMF and control groups were similar (Table 2).

The correlation coefficient revealed a weak correlation between dental anxiety with disease duration and disease severity activity in the FMF group (R²= 0.04) (Table 3). There was not relationship between the FBRS scores and disease duration, disease severity activity (p=0.07 and p=0.49). The relationship factor showed a weak, non-significant correlation between dental fear with disease duration (R²= 0.04, p=0.29) (Table 3). There was a negative low correlation between the CFSS-DS scores and disease severity activity in the FMF group (R²= 0.05, β=-0.21, p=0.04) (Table 3).

Discussion

It has been reported that behaviours of pediatric patients with the systemic disease during dental treatment are affected by the period they stayed in the hospital, depending on their previous medical stories [14]. This pioneering study examined the relationship between dental behaviour and dental fear patterns in children with FMF, and the activity of the disease was evaluated [16, 17].

A child’s age is among the factors affecting occurrence of dental anxiety and fear [18]. Average age of FMF patient group was 10. 55 years, and duration of disease was 4.47 years, in our study. It has been reported in the literature that medical fear and anxiety are more commonly seen in young children [19]. Our results also indicate that the chronic disorder of FMF does not negatively affect a patient’s initial dental compliance. Our results may show that their ability to cope with problems is more developed due to the occurrence of the disease at a young age compared to their healthy peers.

Gender is also a factor that affects the behaviours of pediatric patients. In a study where aetiology of dental fear in ten different societies was investigated, it was revealed that girls had more dental anxiety than boys, but there was no gender difference in two of the populations [20]. Additional studies have also shown that dental anxiety was higher in girls, although differences were not statistically significant [21, 22]. In their study, Fägerstad et al. compared state of dental anxiety among children from different communities living in Sweden, and they reported that girls had higher levels of dental anxiety regardless of social background. They also did not see statistically significant correlation between dental anxiety and age [23]. In our study, similar to previous literature, although not statistically significant, dental anxiety was found higher in girls.

Determination of dental anxiety, dental behaviour and dental fear patterns before starting dental treatment and the investigation of their causes positively affects the success of the patient’s treatments [21, 22]. The anxiety level in FMF patients was similar to healthy children, which was low. As far as we know, there are no previous studies published on dental anxiety, behaviour, and dental fear

patterns in children with FMF. Therefore, we have associated our results with the studies on medical anxiety and fear. A study managed by Makay et al. showed that anxiety values of patients with FMF and the healthy group were similar in 96 children between the ages of 5-15 years [24]. However, Deger et al. and Sonmez et al. reported that patients with FMF were more probably to have anxiety compared to control group [25, 26].

The reasons underlying the development of dental fear and anxiety in pediatric patients are evaluated in 3 sections, including individual factors, environmental factors, and dental factors [27]. Systemic disorders and the medical history of the patient are evaluated within the individual factors. In our study, the disease severity of patients with FMF is mainly medium and low. In patients have FMF, severity of attacks is easily reduced with oral administration of Colchicine. Our results showed dental fear in patients with FMF was low and similar to those of healthy children. Although not statistically different, the average level of dental fear was higher in healthy children compared to patients with FMF. Patients with FMF may have more experience and knowledge about medical treatments as per their healthy equals, leading to reduce in dental fears.

In current study, we also investigated the severity score between the disease severity score and dental anxiety. As far as we know, there are very few studies in literature that show relationship between disease severity score and clinical findings Ekinçi et al. In their study, they emphasized that the disease severity score in FMF was affected by diseases accompanying FMF, especially JIA [28]. In contrast, Keskindemirci et al. They reported that there was not relationship between disease severity score and hearing loss in FMF [29]. Our study showed that disease severity scoring in FMF had a statistically significant effect on dental anxiety and fear. Our study was the first study to show the relationship between disease severity score and dental anxiety in FMF.

The low level of fear of the dentist among healthy children may be because young patients do not undergo any interventional procedures with only a dental examination performed. In a study conducted on children with cleft lip and palate, it was observed that they behaved more comfortably during their dental treatments and that their fear was reduced in time [30]. In our study, dental fears may be low in patients with FMF depending on acceptance, acclimation and normalization. Another explanation of the results obtained is that low levels of anxiety and fear of patients with FMF and control group can be associated with the character of illness. FMF is a chronic inflammatory illness, however, attacks are short-lived (12-72 h) and limits itself to full periods of prosperity. Therefore, high levels of anxiety and fear may not be observed as expected from patients with a chronic illness [24].

A limitation of this study is that it was directed in a single center where only patients living in Tokat were evaluated. It may not be appropriate to generalize results; however, it is possible to overcome prejudice in a single center since Tokat is one of the provinces where FMF is most frequently observed. While all patients were incorporated in the study, they were in the attack-free period, which may alter their psychology, during evaluation. Fur-

thermore, patients with active illness in the attack-free cycle may be more effective in interpreting the psychological effect of FMF in patients.

Conclusion

FMF is a endemic illness, which is not only constricted to eastern Mediterranean region. Tokat is one of the provinces where FMF is most frequently observed. As far as we know, there are no previous studies published on dental anxiety, behaviour and fear patterns in children with FMF. Dental anxiety-fear level in FMF patients was similar to healthy children, which was low. This may be due to the short duration of FMF attacks and limiting itself to full periods of prosperity. It is necessary to avoid the idea that all chronic diseases will cause dental anxiety and fear, and they should be evaluated according to the nature of the chronic disease.

Ethics approval

Approval of the clinical research ethics committee of Tokat Gaziosmanpaşa University (Ethics No: 19 KAEEK 167) was obtained for the study.

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