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Case study

Emotional and behavioral influence of headache in Pediatric rheumatic diseases

Burak Dogangun [a], Mustafa Emir Tavsanli [a], Ezgi Sen [a], Murat Guntel [b,*], Ozgur Kasapcopur [c], Melis Sohtaoglu [d], Erdal Yanardoner Stat [a], Sabahattin Saip [d], Derya Uluduz [d]

[a] Cerrahpasa School of Medicine, Department of Pediatric Psychiatry, Istanbul, Turkey
[b] Elazığ Education and Research Hospital, Department of Neurology, Elazığ, Turkey
[c] Cerrahpasa School of Medicine, Department of Pediatric Rheumatology, Istanbul, Turkey
[d] Cerrahpasa School of Medicine, Department of Neurology, Istanbul, Turkey

1. Introduction

Primary headache is common among children and adolescents [1]. Migraine occurs 3% in the preschool; 4–11% in the primary school years and the frequency increases up to 8–23% in adolescents [2–10]. Headache can be the expression of psychological difficulties, such as anxiety and depression. Stressfullife events have been frequently implicated in the onset, exacerbation, and maintenance of headache. Conversely, headache itself can be a source of stress, leading to functional impairment, which can cause anxiety in patients [11–15].

Familial mediterrenean fever (FMF) is an inherited childhood periodic fever syndrome with recurrent painful attacks affecting the abdomen, chest or joints [16]. The children with chronic rheumatic diseases face a number of difficulties including dependency upon family, isolation from their peers and suffering of physical problems. Pediatric patients are uncertain with regard to disease prognosis and they may undergo a number of complicated medical procedures, all of which can influence their mood, affect and pain perception [17]. Neurological involvement is a rare condition in patients with FMF and also primary headaches might be encountered depending on depression and anxiety in terms of the severity [18]. Although many children and adolescents with chronic rheumatic disease function well, they may have behavioural problems, especially internalizing problems such as being withdrawn and depressed [17–19]. Behavioural problems possibly affect the outcome of treatment through multiple complementary pathways. Comorbid behavioural problems may cause poor adherence, poor management and poor functional health status. Also burden of the chronic disease may lead to behavioural problems [19].

Behavioral symptoms in patients with chronic rheumatic disease suffering primary headaches have not been emphasized since now. The purpose of our study was to examine; (a) the presence of behavioural and emotional problems among pediatric patients with chronic rheumatic disease suffering from primary headache, (b) the possible association between type of headache and psychological symptoms.

2. Methods

Patients with a diagnosis of FMF who had been following in a tertiary clinic of Pediatric Rheumatology Research Center for at least one year and who were in remission period were included in this study. The definite diagnoses of FMF were made by the same pediatric rheumatology expert according to the clinical characteristics and laboratory findings of the patients and depending on the definition criteria [20]. Patients were recruited during a clinical visit in pediatric rheumatology and were randomly referred to neurology clinic for the assessment of headache existence. Informed consent was obtained from the parents according to the procedures of the institutional review board. Data were collected from one parent, who was accompanying to the child. The evaluation of headache was done by a neurologist specialized with headache disorders using the ICHD diagnostic criteria (ICHD-2013). Patients were selected if they had either migraine or tension type headache (TTH) for at least 6 months; regarding the frequency and duration, intensity, localization, quality of pain, associated symptoms, and aggravating factors of each headache. The final diagnosis of headache was made by another neurologist according to the International Classification of Headache Disorders-II (ICHD-II) [21] during face-to-face interviews. Neurological examinations were performed by neurologists under the supervision of a specialized neurologist from the Headache Center. Patients with abnormal neurological examination and any neurological disorder were excluded. We only selected patients with TTH and migraine diagnosis, for at least 6 months and have attack frequencies of at least once a month. Participants meeting all but one of the diagnostic criteria for a given headache subtype were classified as 'probably' having that headache type. Those diagnosed as migraine or

Abbreviations: FMF, familial mediterrenean fever; TTH, tension type headache; ICHD-II, International Classification of Headache Disorders-II; CBCL, the child behaviour checklist.

* Corresponding author at: Elazıg Education and Research Hospital Neurology Clinic, Inonu Street, 23200 Elazıg, Turkey.
E-mail address: muratguntel@hotmail.com (M. Guntel).

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probable migraine were considered to have migraine and those classified as TTH or probable TTH were considered to have TTH.

FMF patients with no complaint of headache were selected as control group. All patients were evaluated by pediatric psychiatrists on the aspect of psychiatric comorbidity. The evaluation of behavioural disorders was done with The Child Behaviour Checklist (CBCL) which was completed according to the data given by the parents during the same session. The tests were commented by a specialist of child and adolescent psychiatry. CBCL is a 113-item questionnaire completed by the parents. The questionnaire consists of two parts: 'Competences' and 'Emotional and Behavioural Problems' scales. In the first section, social competence is covered by three scales: Activity scale, Social scale and School scale. CBCL rates behavioural and emotional problems both globally and with two dimensions; 'internalizing' symptoms composed of subscales Anxious/Depressed, Withdrawn, and Somatic Complaints, such as anxiety and depression, and 'externalizing' symptoms, such as aggression and hyperactivity [22]. Raw scores of each clinical factor were transformed to t-scores based on published norms, and scores of ≥64 were considered indicative of clinical impairment [23].

The data was analyzed by SPSS 20.0 software (Chicago, IL, USA). Descriptive statistical analyzes were used for socio-demographic data. Kolmogorov–Smirnov test showed that the data has not a normal distribution and for this reason Mann Whitney *U* and Kruskal Wallis tests were used for non parametric analysis. Spearman correlation was used to compare two variables and Chi-square test was used for the distribution of categorical variables. The alpha level for significance is accepted as $p < 0.05$.

3. Results

The study group consisted of 69 patients with FMF, 33 females (48%) and 36 males (52%). The mean age of the group was 12.57 ± 2.98 years. The subjects were divided into two groups according to have a complaint of headache. The group of the subjects with headache (Group H+) consisted of 27 subjects and without headache (Group H−) consisted of 42 subjects. There were 15 girls (56%) and 12 boys in Group H+ and the mean age was 12.60 ± 2.44 years. The mean education time of the mothers was 5.19 ± 2.07 years and the fathers were 6.56 ± 2.40 years. Group H− had 18 girls and 24 boys (57.1%) with a mean age of 12.57 ± 2.98 years. The education time was 5.64 ± 2.58 years for mothers and 7.0 ± 3.09 for fathers. There was no significant difference between gender, mean age and education times of the mothers and the fathers among both groups (Table 1).

Table 2 demonstrated the results of Child Behaviour Checklist (CBCL) total and subdimension scores of the groups. Group H+ had significantly higher total scores than the subjects of Group H− ($p = 0.020$). The analysis of internalization problems showed higher scores in anxiety/depression subtitles ($p = 0.002$), although unsignificant in withdrawn subtitle ($p = 0.062$), in group H+. Aggressive behaviour score was also higher in the analysis of externalization problems of group H+ ($p = 0.01$). Interpretation of T scores for Total Problems, Externalizing Problems and Internalizing Problems can be evaluated in three groups: T scores less than 60 are considered as normal; 60–63 as borderline scores and over 63 (≥64) as clinical range [23].

The comparison of H+ and H− groups in terms of clinical relevance ($T \geq 64$), revealed that subjects with headache had significantly higher frequency on total scores (44%) ($p = 0.043$) and externalizing problem scores (33%) ($p = 0.03$) than subjects without headache. Frequencies of subjects of group H+ on anxious/depressed and rule breaking subscales were 67% ($p = 0.012$) and 30% ($p = 0.013$), which was also a significantly higher percentage than children of H− group (Table 3).

Spearman Correlation of the CBCL total and subdimension scores regarding to how long the subject has headache showed no significant relationship between subjects' total score and headache histories ($r = 0.33$, $p = 0.091$). The correlation of the CBCL total and subdimension scores regarding to the duration and frequency of the headache for the last month showed no significance between subjects ($r = 0.12$, $p = 0.557$ and $r = 0.11$; $p = 0.468$ respectively). However there was a significant correlation between regression scores for either the duration and the frequency of headache ($r = 0.39$, $p = 0.042$ and $r = 0.31$; $p = 0.040$ respectively). For the rest of the subdimensions, there was not any significant correlation. Mann Whitney *U* analysis of the comparison for total and subdimension scores of CBCL and having migraine (M) or tension type headache (TTH) revealed no significant difference ($p = 0.979$).

4. Discussion

Pediatric headache impacts quality of life and seems to be associated with behavioral, emotional, and social problems and competencies [19,24]. To best of our knowledge, the present study is the first study to examine the behavior and emotional correlates of primary headaches in chronic rheumatic disease population.

Starting point of our research was to reach an association between headache and psychopathology on children and adolescents with a chronic rheumatological disease. Comprehensive meta analytic studies have confirmed our idea that arthritis, headache and chronic pain seem to have association with behavioral disorders on children and adolescents [19,24,25]. However in reviewing the literature, no previous data was found about comparison of the affects of different types of arthritis on children's and adolescents' psychological well being. In the current study, except thought problems domain, we did not found significant differences on CBCL scores of subjects with FMF. Nevertheless, we found that subjects with headache had significantly higher scores on total, internalizing, and externalizing problem scores; which can be concluded as the psychology and behavior of children who have a chronic rheumatological disease are negatively affected if they also experience headaches. In terms of clinical relevance (*T* score greater than 64), subjects with headache also had higher ratios than subject without headache on total and externalizing problem domains.

Table 1
Demographic features of the groups.

	FMF cases with headache			FMF cases without headache	*P*
	Migraine (*n*)	TTH (*n*)	Total (*n*)	(*n*)	
Gender (F/M)	11/7	4/5	15/12	18/24	0.897
Mean age	11 ± 2.82	10.33 ± 2.65	12.6 ± 2.44	12.57 ± 2.98	0.122
Mean education time of mother	5.67 ± 1.64	4.22 ± 2.59	5.19 ± 2.07	5.64 ± 2.58	0.442
Mean education time of father	6 ± 1.78	7.67 ± 3.16	6.56 ± 2.40	7.00 ± 3.09	0.529
Mean of headache duration (h)	1.94 ± 1.51	0.93 ± .97	1.60 ± 1.42		

FMF: familial mediterrenean fever, TTH: tensiontype headace, F: female, M: male.