Living with a child with familial Mediterranean fever: Does it affect the quality of life of the parents?

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Familial Mediterranean fever (FMF), quality of life, children, parents.

ABSTRACT

Objective
The aim of the present study was to assess the quality of life (QOL) and the psychological status of parents of children with familial Mediterranean fever (FMF).

Methods
The QOL, anxiety and depression of the parents of 35 children with FMF were evaluated and compared to the parents of 23 healthy children.

Results
Mothers of FMF children had lower QOL scores than mothers of healthy children: 5.5±1.1 versus 6.0±0.6 (p = 0.048). They also expressed higher levels of anxiety and depression. Within each group, mothers were more anxious and depressed than fathers. Parents with several FMF children were not significantly different from parents with only one FMF child.

Conclusion
The QOL and psychological well being of parents with FMF children were found to be slightly impaired, especially that of the mothers.

Introduction
The quality of life (QOL) of patients with rheumatic diseases is increasingly recognized as an important factor in assessing health and in planning treatment and management strategies. Indeed, evidence exists that persons with systemic rheumatic disease such as rheumatoid arthritis, systemic lupus erythematosus or fibromyalgia (FM) have a decreased QOL (1-4). Furthermore, we have shown that even relatives of FM patients have an impaired QOL, which may be attributed to the psychological distress in families with a chronic rheumatic disease (5).

Familial Mediterranean fever (FMF) is an autosomal recessive disorder occurring in persons of Mediterranean and Armenian descent. The disease is characterized by episodic fever accompanied by arthritis, peritonitis, pleurisy, skin rash and other signs and symptoms. Treatment with colchicine has dramatically changed the prognosis of FMF patients, decreasing the appearance of febrile attacks and preventing amyloidosis (6, 7).

Between the acute attacks, patients can lead a normal life. The disease-related gene has recently been discovered and coded for a protein that guides the neutrophil’s participation in inflammation (8).

We have found (9) that adult FMF patients reported considerably more dissatisfaction with various aspects of their life than healthy controls. One may speculate that the QOL of parents with FMF children will be impaired as well, because the disease of the child may pose a psychological and functional burden on his/her parents. The aims of the present study were: (a) to evaluate the QOL and psychological status of parents with FMF children compared to that of parents of healthy children; (b) to compare the QOL of mothers and fathers within each group, i.e. the groups with and without FMF children; and (c) to determine the effect of multiple cases of FMF in a single family on the QOL of the parents.

Materials and methods

Subjects
The parents (both mother and father) of 35 children with FMF and the parents of 23 healthy children participated in the study. No significant differences were found between the ages of the mothers (40±6 versus 38±5 years) and fathers (43±7 versus 42±6 years) in the FMF and control groups, respectively. They also had similar educational levels: 12±2 versus 13±3 years for mothers and 13±2 versus 14±3 for fathers.

The FMF children were randomly chosen from a list of 100 FMF patients being followed at the pediatric rheumatic disease clinic in Soroka Medical Center, Beer-Sheva, Israel. The patients are regularly seen every 3 to 6 months. Their parents were previously informed about the nature of the disease, the outcome and the treatment. The control group consisted of parents of 23 healthy children of similar ages and were recruited from the hospital personnel. The study was approved by the Helsinki Committee of the Soroka Medical Center; all participants gave their written consent after having received detailed information about the study.

The demographic and clinical background of the FMF children and the
healthy controls are shown in Table I. The gender and age distributions were similar in both groups. The mean duration of FMF was 4.7 ± 3.3 years. None of the FMF children had amyloidosis or protracted arthritis. One patient had erysipelas-like erythema, and 6 patients had acute arthritis. We also calculated a severity score of FMF, according to recently established criteria (10). The mean severity score was 5.8 ± 1.5. According to this system, the FMF disease of 19 children (54%) was mild (score ≤ 5), and in 16 children (46%) it was moderate (score: 6-10).

The FMF children missed significantly more school days than the healthy controls (Table I): 6.7 ± 8.0 vs 1.4 ± 2.0 days in the past 3 months (P < 0.01). The quality of life and the psychological status were assessed in all parents in both groups using the same questionnaires.

Table I. Demographic and clinical background of children with and without familial Mediterranean fever (FMF).

<table>
<thead>
<tr>
<th>Variable</th>
<th>FMF group (n = 35)</th>
<th>Control group (n = 23)</th>
</tr>
</thead>
<tbody>
<tr>
<td>M/F ratio (%)</td>
<td>18/17 (51/49)</td>
<td>13/10 (57/43)</td>
</tr>
<tr>
<td>Age, years</td>
<td>10.2 (3.9)</td>
<td>8.6 (2.7)</td>
</tr>
<tr>
<td>FMF duration, years</td>
<td>4.7 (3.3)</td>
<td>—</td>
</tr>
<tr>
<td>FMF severity score</td>
<td>5.8 (1.5)*</td>
<td>—</td>
</tr>
<tr>
<td>Siblings</td>
<td>2.5 (1.7)</td>
<td>2.3 (1.9)</td>
</tr>
<tr>
<td>School days missed</td>
<td>6.7* (8.0)</td>
<td>1.4* (2.0)</td>
</tr>
</tbody>
</table>

Family physician visits (last 6 months)

<table>
<thead>
<tr>
<th></th>
<th>FMF group (n = 35)</th>
<th>Control group (n = 23)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a) Due to FMF</td>
<td>1.5 (2.3)</td>
<td>—</td>
</tr>
<tr>
<td>b) Other causes</td>
<td>1.4 (1.9)</td>
<td>1.0 (1.0)</td>
</tr>
</tbody>
</table>

Emergency room visits (last 6 months)

<table>
<thead>
<tr>
<th></th>
<th>FMF group (n = 35)</th>
<th>Control group (n = 23)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a) Due to FMF</td>
<td>0.3 (0.6)</td>
<td>—</td>
</tr>
<tr>
<td>b) Other causes</td>
<td>0.4 (1.1)</td>
<td>0.2 (0.4)</td>
</tr>
</tbody>
</table>

Table II. Quality of life and psychological variables of parents in the study and control groups (means and standard deviations)

<table>
<thead>
<tr>
<th>Variable (range)</th>
<th>FMF group (n = 35)</th>
<th>Control group (n = 23)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mother</td>
<td>Father</td>
</tr>
<tr>
<td></td>
<td>Mother</td>
<td>Father</td>
</tr>
<tr>
<td>Quality of life (1 - 7*)</td>
<td>5.5 (1.1)</td>
<td>5.5 (0.9)</td>
</tr>
<tr>
<td>Anxiety (SCL-90) (1 - 5**)</td>
<td>2.0 (0.8)</td>
<td>1.7 (0.6)</td>
</tr>
<tr>
<td>Anxiety (AIMS) (0 - 10**)</td>
<td>4.8 (1.8)</td>
<td>3.4 (1.1)</td>
</tr>
<tr>
<td>Depression (AIMS) (0 - 10**)</td>
<td>3.0 (2.3)</td>
<td>2.2 (1.2)</td>
</tr>
</tbody>
</table>

*Best possible score; **worst possible score.

Psychological status

Psychological status was assessed by the anxiety and depression subscales of the AIMS: Arthritis Impact Measurement Scales (14), each consisting of 6 questions to be answered on a 6-point scale. The final indices are the average scores normalized to range from 0 to 10, with “0” reflecting the best condition. Anxiety was also measured using a subscale of the SCL-90 psychiatric instrument (15). This subscale includes 10 questions to be answered on a 5-point scale. The final scores consist of the average scores, ranging from 1 to 5, with 5 reflecting the worst condition.

Statistical analysis

T-tests for independent samples were used to compare the study and control groups, while mothers and fathers within the same group were compared by paired t-tests.

Results

Table II compares the quality of life and the psychological variables for mothers and fathers within the study and the control groups, as well as for the mothers and fathers across the two groups. Mothers of FMF children were significantly less satisfied with their lives than the mothers of healthy children: 5.5 ± 1.1 versus 6.0 ± 0.6 (p = 0.048). No differences were observed between the fathers in the two groups. Within each group, both parents similarly evaluated their QOL.

The anxiety level as measured by SCL-90 was similar for mothers and fathers in both groups. However, the AIMS anxiety score was different between the two groups, as well as between mothers and fathers within each group: the mothers expressed more anxiety than the fathers (p = 0.009 and p = 0.05, for the mothers and fathers, respectively, in the FMF group and the control group).

The mothers in the FMF group expressed significantly higher levels (p = 0.012) of depression than their husbands, and slightly higher levels than the mothers in the control group (p = 0.062).

The parents of children whose FMF dis-
ease was considered mild did not differ from the parents of children whose disease was moderate: their QOL scores and levels of depression and anxiety were similar (p > 0.05). Also, no significant correlations were observed between these variables in the parents and the severity score of the children’s FMF disease.

We furthermore addressed the question as to whether having several children affected with FMF in the same family posed a greater burden on the parents. Therefore, we compared the reported quality of life and levels of anxiety and depression in 24 families with one FMF child to that of 11 families with 2 to 4 FMF children (Table III). Interestingly, no differences were detected between these families. However, once again within each group the mothers expressed more anxiety (p = 0.008 and p = 0.020 in the two groups, respectively) and more depression (p = 0.011 and p = 0.018, respectively) than the fathers.

**Discussion**

The quality of life of patients with rheumatic diseases is adversely affected to an extent comparable with that of other chronic diseases, such as chronic obstructive pulmonary diseases and insulin dependent diabetes (3). However, the impact of such diseases, specifically FMF, on the QOL of their relatives has not yet been studied. In the present study, mothers of FMF children reported a lower QOL, and expressed slightly higher levels of anxiety and depression than mothers of healthy children. Though some of the differences were statistically significant, the actual differences were not remarkable. Surprisingly, no differences were observed between the fathers in the two groups. Within each group, the mothers were more anxious and more depressed than the fathers. In a previous study we similarly addressed the issue of QOL in relatives (parents, siblings and children) of patients with FM. We found the QOL to be impaired, especially in female relatives and those with undiagnosed FM (5). In both the FM study and the present one, female relatives expressed more anxiety and depression than male relatives.

It should be pointed out that our negative results, namely the finding of no differences regarding the parents’ psychological well-being, should be interpreted cautiously. The power of the performed t-tests was not high, due to the small sample sizes (n = 35 and n = 23). Therefore, the differences in the anxiety and depression levels between mothers and fathers in the FMF group, and between mothers in the two groups, did not reach statistical significance.

The low QOL of mothers with FMF children may be attributed to the atmosphere of stress and anxiety characteristic of families with members affected by a chronic disease. Treatment with colchicine has changed dramatically the prognosis of FMF patients, decreasing the appearance of febrile attacks and preventing amyloidosis. Between acute attacks, the patients can lead a normal life. Thus, one may speculate that since all FMF patients are now regularly treated with colchicine, their QOL and as a consequence that of their relatives have been improved. This may also explain our finding that parents with several FMF children were not different from parents with only one FMF child.

One of the limitations of the present study is the lack of a positive control group, for example parents of patients with juvenile chronic arthritis (JCA).

### References