Introduction

Anxiety is the psychological process by which the individual reacts to dangerous external stimuli, triggering responses (mediated by norepinephrine, GABA and serotonin) involving physiological reactions and cognitive strategies. Anxiety can be differentiated in state anxiety and trait anxiety.

Fragile X Syndrome (FXS) is a disorder most commonly caused by a triplet repeat expansion of >200 cytosine-guanine-guanine (CGG) repeats in the 50 untranslated region of the Fragile X mental retardation 1 (FMR1) gene.

Aim of the present study is investigating the state or trait anxiety in a sample of mothers of children affected by X-fragile.

Materials and methods: 84 mothers (mean age 48.36 ± 7.11) of children affected by FXS were recruited and compared with mother of 171 typical developing children (mean age 46.18 ± 9.53). The Scale State-Trait Anxiety Inventory for Adults (STAI) was used in order to assess the anxiety levels.

Results: All mothers of children affected by FXS showed high score of anxiety levels at the STAI evaluation (Table 1), with no differences were found between mothers of males and females children affected.

Conclusions: Mothers of children with FXS appear to be in a state of anxiety effects and suddenly even higher than those found later in mothers of children with other chronic diseases.

Keywords: anxiety disorder, X-fragile, scale state-trait anxiety inventory for adults.

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Fragile X Syndrome (FXS) is a disorder most commonly caused by a triplet repeat expansion of >200 cytosine-guanine-guanine (CGG) repeats in the 50 untranslated region of the Fragile X mental retardation 1 (FMR1) gene\textsuperscript{[11-12]}. Aim of the present study is investigating the state or trait anxiety in a sample of mothers of children affected by X-fragile.

**Materials and methods**

84 mothers (mean age 48.36 ± 7.11) of children affected by FXS were recruited and compared with mother of 171 typical developing children (mean age 46.18 ± 9.53). Exclusion criteria were the following: overweight (z-BMI > 85 pc) and obesity (z-BMI > 95 pc), cognitive disability (IQ <70), neurological disorders (ie headaches, epilepsy), chromosomal syndromes different from FXS (eg. Down, Prader-Willi, Crouzon, Pierre-Robin, trisomy 18), psychiatric illness (ie. mood disorders, anxiety disorders, psychosis) and specific neuropsychological disorders\textsuperscript{[13-38]}.

**Anxiety levels assessment**

The Scale State-Trait Anxiety Inventory for Adults (STAI) consists of 40 items, split into 2 multiple choice subscales of 20 items each. The first questionnaire measure states anxiety (S-anxiety), consisting of questions that address how respondents feel at the moment, including items that measure subjective feelings of calm, tension, apprehension, nervousness, worry, and other questions that assess autonomic nervous system activity. State anxiety scale items are rated on a 4 point Likert scale (1 = not at all, 2 = somewhat, 3 = moderately so, and 4 = very much so). The second subscale measures trait anxiety (T-anxiety), on how respondents generally feel, using items that measure general states of calmness, security, and confidence. Items were rated on a 4 point Likert scale (1 = almost never, 2 = sometimes, 3 = often, and 4 = almost always). The total STAI score for each subscale (S-anxiety and T-anxiety) is the addition of response scores for the 20 items, and ranges from 20-80, with higher scores indicating greater anxiety. The scores consent to distinguish different grades of anxiety levels: mild (20-40 points), moderate (41-60 points), severe (61-80 points). A score below 20 is indicative for the absence of anxiety.

**Results**

In our study, all mothers of children affected by Down syndrome showed high score of anxiety levels at the STAI evaluation. By comparison with the control population reveals the presence of significantly more prevalent anxiety levels of moderate and severe in mothers of children with X-Fragile Syndrome. (Table 1). Specifically, there was a distribution of different levels of anxiety: 27 mothers showed mild anxiety was (32.15% vs 66.66%, p <0.001); 31 mothers showed moderate anxiety was (36.9% vs. 5.21%, p = 0.011); 26 mothers showed severe anxiety was (30.95% vs 28.12%, p <0.001). 32 mothers showed mild anxiety tract (38.09% vs. 71.34%, p <0.001); 35 mothers showed moderate anxiety tract (41.66% vs. 18.71%, p <0.001); 17 mothers showed severe anxiety trait (20:23% vs. 9.94%, p = 0.038). (Table 1). No differences were found between mothers of males and females children affected.

<table>
<thead>
<tr>
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<th>FXS N=84</th>
<th>Controls N=171</th>
<th>p</th>
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</thead>
<tbody>
<tr>
<td>Mild state anxiety levels %</td>
<td>32.15</td>
<td>66.66</td>
<td>&lt;0.001</td>
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<tr>
<td>Moderate state anxiety levels %</td>
<td>36.9</td>
<td>21.05</td>
<td>0.011</td>
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<tr>
<td>Severe state anxiety levels %</td>
<td>30.95</td>
<td>12.28</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Mild trait anxiety levels %</td>
<td>38.09</td>
<td>71.34</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Moderate trait anxiety levels %</td>
<td>41.66</td>
<td>18.71</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Severe trait anxiety levels %</td>
<td>20.23</td>
<td>9.94</td>
<td>0.038</td>
</tr>
</tbody>
</table>

Table 1: shows comparison between pathological percentages scores between different anxiety grades among mothers of children affected by FragileX syndrome (FXS) and those of normal children (Controls). Chi-square test was applied. p values <0.05 were considered as statistically significant.

**Discussion**

Anxiety, like all emotional reactions, has an adaptive function, crucial for survival. When this chronically, anxiety has nevertheless a strong impact on the life of the subject. It first reduces the capacity for action, then it becomes pathological anxiety. Our data, like those reported in the literature, highlighting the difficulty of mothers of children with Fragile syndrome to accept and manage the disease, although to varying degrees and with different characteristics from disease pathology\textsuperscript{[39-47]}.
About FXS, the objective difficulties that these mothers encounter in the management of their children, it also adds to the marginalization and social prejudice that still reign. The social and emotional contacts were generally normal, although it was required greater empathy. The families were involved and usually controlled and experiences generally positive. We have seen, however, the result of the study we conducted and performed on a larger sample of the population has in fact contradicted this first observation. The mothers of children with FXS appear to be in a state of anxiety and suddenly even higher than those found later in mothers of children with other chronic diseases\(^{99-100}\).

References


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