# **Development and Aging**

## Psychological tendencies of children with juvenile idiopathic arthritis

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A bulk of studies showed an association between stressful events and juvenile idiopathic arthritis (JIA) but failed to identify specific psychological tendencies that contribute to the patients' vulnerability to stress. The purpose of this paper is to identify psychological tendencies specific to JIA that would unravel characteristic sources of stress. The study is based on the cognitive orientation model of health, which enables us to identify these kinds of tendencies in terms of four belief types (beliefs about self, general beliefs, beliefs about norms, and goals) that refer to specific themes. This is a case-control-cohort study that included a sample of 36 patients (mean age = 12.44 years, SD = 2.97, 21 females) and 41 matched controls (mean age = 13.15 years, SD = 2.01, 22 females). The JIA cognitive-orientation questionnaire was administered, and relevant medical parameters were recorded. The belief types differentiated between the two groups, and the patients were characterized using six themes. Examples of the themes are being oversensitive, striving for success, and not fulfilling duties well. The themes differentiated between the participants' groups with an accuracy of 89.1%. The likelihood of the patients being characterized by the themes is 3.24-9.35 times more than the controls. The psychological tendencies of JIA were discussed as generators of stress (e.g., being over-sensitive) and cognitive conflicts (e.g., the contradiction between striving for success versus not fulfilling duties well). Also, the suggested reflections of these tendencies in the health workers' and patients' relationships, such as egalitarian interaction, and non-formal communication style, were described.

Key words: Juvenile idiopathic arthritis, psychological tendencies, psychological stress, cognitive conflicts, patients-doctors relationships, communication style in the clinical context.

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## INTRODUCTION

Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic disease in childhood, with an annual incidence rate of 1.6 to 23 per  $1 \times 10^5$  people (Thierry, Fautrel, Lemelle, & Guillemin, 2014). It is an autoimmune disease that results from an immune reaction caused or triggered by environmental factors, such as infectious agents in a genetically susceptible host (Horton & Shenoi, 2019; Rewers & Ludvigsson, 2016). The causes of JIA remain largely enigmatic; there is evidence suggesting that developing JIA depends on interactions between genetic and environmental variables (Shiff, Oen, Kroeker & Lix, 2019). The concordance rate of monozygotic twins having JIA is 25%–40% (Prahalad, 2017), suggesting that genetic factors are only one part of the puzzle. Other data indicate that infections, maternal smoking, and nutritional factors may also be contributing factors (Ellis, Munro & Ponsonby, 2010; Huang, 2012).

Another factor that plays a role in the etiology of JIA is psychological stress (Berkun & Padeh, 2010; Straub, Dhabhar, Bijlsma & Cutolo, 2005). The influence of stress before the onset of JIA, as a disease trigger, was evaluated in several studies and results support an association between psychological stress and JIA (Aasland, Flato & Vandvik, 1997; Herrmann, Scholmerich & Straub, 2000; Rubinstein, Bullock, Ardalan *et al.*, 2020). For example, a retrospective study conducted on 868 patients with JIA and 1,042 control subjects found that the patients' parents reported antecedent experiences of serious upsets, ill family members, parents' separations, and interpersonal difficulties to be 4.81, 2.29, 1.96, and 2.54, times more than the parents of the control group, respectively (Neufeld, Karunanayake, Maenz & Rosenberg, 2013). Another study found that JIA patients had a higher likelihood of having adoptive parents or being raised by a single parent than the control subjects (Henoch, Batson & Baum, 1978).

However, psychological stress is not specific to JIA, and it was reported as characteristic of many states of health and autoimmune diseases (Schiffenbauer & Miller, 2020). Furthermore, stress depends not only on external stressors, but also on internal tendencies, personality characteristics, and psychological dynamics which also could function as a source of stress (Ebstrup, Eplov, Pisinger & Jorgensen, 2011; Lazarus & Folkman, 1984; Lecic-Tosevski, Vukovic & Stepanovic, 2011; Subhashini, 2017; Vollrath, 2001). Therefore, the present study focuses on revealing specific psychological tendencies relevant to JIA and related to stress. The rationale behind identifying these kinds of tendencies is related to the current novel approach in healthcare systems toward utilizing personalized medicine in clinical frameworks (Cesario, Lohmeyer, D'Oria, Manto & Scambia, 2021; Fournier, Prebet, Dormal, Brunel, Cremer & Schiaratura, 2021). The main concept of this approach is to develop and provide tailored interventions in line with the personal needs and unique genetic and environmental profile of each individual patient and disorder (Mathur & Sutton, 2017). The framework of this approach avoids trial-and-error treatments and strives to plan selective optimal treatment in line with the unique bio-psycho-social characteristics of the patient. Since this approach is already applied in rheumatoid arthritis, such as during

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making decisions concerning providing biological treatment based on the patient's unique characteristics (Johnson, Sanchez & Schoenbrunner, 2019), it is necessary, therefore, to align with this approach at the psychological level by identifying specific psychological profiles and preparing the ground toward tailoring targeted interventions that correspond to the specific profile and characteristics of the patients (Kreitler, 2019).

The study is based on the cognitive orientation model of health (COH), which provides a comprehensive framework and empirical-based tools to identify specific psychological tendencies for a particular disease (Kreitler, 1999, 2019). According to this model, the occurrence of any physical disease depends on interactions between the following three factors: (1) the pathogen which is the carrier or instigator of the disease, such as viruses or microbes; (2) the background factors that modulate the vulnerability of the organism to succumb to the pathogen or resist its impact; and (3) the physiological mechanism or procedure that leads to the development and the occurrence of a given disease (Kreitler, 1999, 2019). This model focuses on the background factors and deals mainly with identifying specific psychological tendencies relevant to a given disease.

The tendencies are defined in terms of the following four belief types that refer to a set of disease-relevant themes: (1) beliefs about self, express information about oneself, such as one's habits, actions or feelings (for example, "I like being in control of my reactions"); (2) general beliefs, express information concerning reality and others (for example, "People usually like controlling their reactions"); (3) beliefs about rules and norms, express rules and social standards (for example, "One should always like being in control"); and 4) beliefs about goals, express one's wants, wishes, desires or dislikes (for example, "I want to be in control of my reaction"). The contents of the belief types identified by a standard procedure are called themes. The themes are not conscious; they are unique for a particular disease and constitute the underlying meanings that the patients provide or attribute to the diagnosis. Thus, the themes are indirectly related to the disease and unconnected to the explicit meanings of the disease itself (Kreitler, 2019).

Therefore, according to the COH model, the psychological tendencies regarding each state of health consist of the four belief types, but in each case, the content of the beliefs is different and represents a cluster of themes that are specifically relevant to a particular disease (Kreitler, 1999, 2019). This model enabled the identification of specific psychological tendencies related to stress for anorexia (Kreitler, Bachar, Canetti, Berry & Bonne, 2003), ischemic heart disease (Drechsler, Bruner & Kreitler, 1987), gynecological infections (Kreitler, Kreitler & Schwartz, 1991), type 2 diabetes (Kreitler, Weissler & Nurymberg, 2004), colon and colorectal cancer (Figer, Kreitler, Kreitler & Inbar, 2002; Kreitler, Kreitler & Barak, 2013), asthma (Roth & Kreitler, 2020), Crohn's and colitis in children (Badarnee, Weiss, Shouval & Kreitler, 2022).

#### The study objectives

Previous studies reported an association between JIA and increased rates of experiencing psychological stress and severe life events before the onset of the disease (Neufeld *et al.*, 2013).

However, psychological stress is not specific to JIA and it was reported as a characteristic of other autoimmune diseases, such as inflammatory bowel diseases (Badarnee et al., 2022; Rampton, 2009). In addition, stress is not related only to life events but also depends on personality tendencies that make an individual vulnerable or resistant to stress (Ebstrup et al., 2011; Lazarus & Folkman, 1984; Lecic-Tosevski et al., 2011; Subhashini, 2017; Vollrath, 2001). Thus, revealing some of those tendencies may deepen our view in regard to identifying several internal sources of stress, such as cognitive conflicts, and as a result, providing essential clinical tools for lowering the stress levels based on the psychological dynamics of the patients. Identifying psychological tendencies relevant to JIA may pave the way for developing a new generation of targeted psychological interventions that correspond specifically to the characteristics of children with JIA. From the perspective of health psychology, this study has the potential to contribute to the personalized medicine approach, which proposes to establish clinical decisions based upon the individual profile of a patient, tailoring the treatment to his or her characteristics, needs, and preferences (Fournier et al., 2021; Perna, Grassi, Caldirola & Nemeroff, 2018).

Therefore, the present study was set to identify specific psychological tendencies related to stress and cognitive conflict in children with JIA. It is done in the framework of the COH model that defines the tendencies in terms of four belief types (beliefs about self, general beliefs, beliefs about norms, and goals) which refer to specific themes (Kreitler, 1999, 2019). The belief types represent the motivational disposition towards being characterized by the themes, and the themes represent the psychological dynamics of the patients – their internal sources of stress and cognitive conflict.

This study does not claim cause–effect relationships between psychological tendencies and JIA but tries to identify autonomous tendencies, which means that they are supposed to be associated with being diagnosed with JIA but not with the medical parameters of the disease, such as the duration and the severity of the disease. If these tendencies prove to be associated with the disease but not with its different conditions, this will indicate that they are relevant to the disease itself and are not a result of coping with it (Kreitler, 2019). The autonomy of the tendencies will be statistically verified.

## Hypotheses

This study deals with examining two hypotheses. The first is related to the differences between children with JIA and the control ones in each belief type – beliefs about self, general beliefs, beliefs about norms, and goals. The diagnosed children are expected to score higher than the control subjects in the belief types. The second hypothesis deals with 26 themes that were identified in the pretest process. A set of those themes is expected to characterize children with JIA and to differentiate them from others in the control group. The complete list of themes is presented in Table 1.

The two hypotheses expect the psychological variables, which consist of the beliefs and the themes, to be related mainly to the disease itself rather than to its characteristics, such as the duration and the age of the patient at the onset of the disease. This Table 1. The themes' labels in the CO-JIA questionnaire

Table 2. The medical characteristics of patients with JIA (N = 36)

Num.	Themes' labels	Medical variables JIA type <sup>a</sup>		
$T_1^{a}$	Likes being free without limitations			
$T_2^{a}$	Likes being successful and persistent in everything	Oligoarticular n (%)	19 (52.77)	
T <sub>3</sub>	Accepting help and advice from others	Polyarticular n (%)	7 (19.44)	
$T_4$	Likes routines and fixed schedules	Systemic <i>n</i> (%)	2 (5.55)	
$T_5$	Not caring if everyone loves and spends time together	Enthesitis-related arthritis n (%)	1 (2.77)	
T <sub>6</sub>	Not caring about doing tasks well	Undifferentiated n (%)	7 (19.44)	
$T_7$	Not caring whether impressions match real feelings	JIA duration (years) M $\pm$ SD	$5.26\pm4.99$	
T <sub>8</sub>	Not recognizing, identifying, or sensing others' feelings	Patients' age at the onset of JIA (years) M $\pm$ SD	$7.15 \pm 4.76$	
T <sub>9</sub>	Willing to get instructions on how to perform things	Inflamed joint count at the study time M $\pm$ SD	$2.33\pm2.38$	
T <sub>10</sub>	Likes being different and unique	Treatment type <sup>b</sup>		
T <sub>11</sub>	Planning everything, up to the smallest details	Anti-TNF alpha n (%)	12 (33.33)	
T <sub>12</sub>	Being close to people and feeling good, even if one has to give up own	Steroids n (%)	21 (58.33)	
	needs	NSAID $n$ (%)	5 (13.88)	
T <sub>13</sub>	Moving freely without limitations	Cytotoxic n (%)	6 (16.66)	
T <sub>14</sub>	Controlling the body's reactions, movements, and desires	Comorbidity		
T <sub>15</sub>	Being over-sensitive and avoid being insulted or hurt	Uveitis n (%)	5(13.88)	
T <sub>16</sub>	Not sharing feelings, experiences, or difficulties	Asthma n (%)	1 (2.77)	
T <sub>17</sub>	Likes being surprised without knowing the clear outcomes			
T <sub>18</sub>	Likes being active all the time - doing and achieving things	Notes: anti-TNF alpha = anti-tumor necrosis factor-alp	pha; JIA = juvenile	
T <sub>19</sub>	Not caring about one's body or health	idiopathic arthritis; NSAID, non-steroidal anti-inflamma	atory drug.	
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T20 Likes having a lot of adventures and fun

- T<sub>21</sub> Keeping promises to family and friends; sees oneself as a responsible person
- T<sub>22</sub> Likes being orderly and organized
- T<sub>23</sub> Not caring about facts; knowing things more or less is okay

T<sub>24</sub> Befriending everyone, and likes being close

T<sub>25</sub> Avoidance of being angry, hiding it when it happens

 $T_{26}{}^a$   $\;$  Having control over one's reactions, in regard to saying, feeling, and doing

*Notes*: CO-JIA Questionnaire = cognitive orientation questionnaire of JIA; JIA = iuvenile idiopathic arthritis; Num. = number; T = theme.

<sup>a</sup>Those themes' labels are supported by two belief types instead of three belief types as the COH model suggests.

expectation was based on previous findings of other diseases indicating that these kinds of psychological variables are not dependent on the state of the disease (Badarnee *et al.*, 2022; Drechsler *et al.*, 1987; Figer *et al.*, 2002; Kreitler, 2019). Therefore, a preliminary analysis will be conducted in order to examine whether the psychological tendencies are autonomous and not dependent on the relevant medical parameters of JIA.

## MATERIALS AND METHODS

#### Participants

The sample size was determined by running statistical power analysis using G\*Power 3.1 software (Faul, Erdfelder, Buchner & Lang, 2009). The overall suggested sample size for *t*-test with  $\alpha = 0.05$ , power = 0.95, and a large effect size of d = 0.8 (Cohen, 1988) was 70 participants. The estimated sample size for MANOVA analysis, *F*-test with  $\alpha = 0.05$ , power = 0.95, a large effect size of  $f^2 = 0.35$  (Cohen, 1988), two groups, and four independent variables (the belief types), was 60 participants. And the suggested total sample size for logistic regression analysis with  $\alpha = 0.05$ , power = 0.95, and odds ratio = 2.9 [determined based on the average odds ratio of stressful factors in children with JIA (Neufeld *et al.*, 2013)] was 69. Therefore, the power analyses suggested an overall sample size of between 60 and 70 participants. According to these findings, 77 children were recruited in this study.

Thirty-six children with JIA, 14 males and 21 females, whose mean age was 12.44 years old (SD = 2.97) were recruited for this study. Table 2

*Notes*: anti-TNF alpha = anti-tumor necrosis factor-alpha; JIA = juvenile idiopathic arthritis; NSAID, non-steroidal anti-inflammatory drug. <sup>a</sup>Most patients in this study are diagnosed with oligoarticular JIA subtype which agrees with previous epidemiological studies that showed this subtype of JIA to be the most prevalent one in the Middle East (Al-Mayouf *et al.*, 2021), and the worldwide (Consolaro *et al.*, 2019).

<sup>b</sup>The treatment type refers to the medical treatment the patients received during the study.

shows the medical characteristics of the patients. Forty-one children with no chronic diseases were recruited to the control group, 19 males and 22 females, whose mean age was 13.15 years old (SD = 2.01). The groups match in age (t [60.21] = -1.20, p = 0.24) and gender ( $\chi^2$  [1, 76] = 0.31, p = 0.65). With respect to the children school function, all participants study at regular education environments, and the grade point average of the patients (M = 87.95, SD = 5.82) and the controls (M = 88.67,SD = 5.09) are matched (t [75] = -0.57, p = 0.56). 88.88% of children with JIA and 87.80% of the controls are right-handed ( $\chi^2$  [1, 76] = 0.02, p = 1.00). Regarding the participants' family size, there was no difference in the number of siblings between the two groups (t [47.15] = 1.76, p = 0.09), while the patients have on the average 3.19 siblings (SD = 2.09), the compared group has 2.24 siblings (SD = 1.33). In addition, the two groups are matched in regard to the birth order of the participant ( $\chi^2$  [1, 76] = 0.68, p = 0.71). 30.6% of children with JIA are firstborn, 38.9% are middle born, and 30.6% of them are the youngest children in their families. Concerning the controls, 39.0%, 36.6%, and 24.4% of them are first, middle, and youngest born children, respectively. And finally, 41.6% of the patients and 29% of the controls lead a religious-traditional lifestyle, whereas 58.4% of the patients and 71% of the controls adopt a secular or non-religious lifestyle. There were no differences between the two groups concerning the degree of leading a religious lifestyle ( $\chi^2$  [1, 76] = 1.29, p = 0.33).

#### Tools

The Psychological Questionnaire. The Cognitive Orientation Questionnaire of JIA (CO-JIA) was specially developed for this study. It is based on both components of the COH model: the four belief types and the disease-relevant themes (Table 1). Each theme in this questionnaire is presented as a short story about two children. The participants were requested to make four decisions that represent their beliefs regarding a given theme: the first is in line with whether the theme describes oneself; while the second deals with what usually happens in reality, or whether the theme describes others; the third is in line with what should be; and the fourth is in line with one's goals or desires. The following example is cited from the CO-JIA questionnaire. It refers to the theme "Likes being successful and persistent in everything" and it is presented here to illustrate the participants' tasks:

"It is very important for Tommy to succeed in everything he does and persevere in all tasks to achieve good results. On the other hand, it is not so important for Adam to succeed in everything or in all tasks he does."

- *A*: Who are you like? Whom do you resemble? Tommy, who strives for success? \_\_\_\_\_ or Adam, who does not mind succeeding in all?
- B: In your opinion, how are most children? Do they like to succeed in all tasks, like Tommy? \_\_\_\_, or are they like Adam, who does not mind succeeding in all? \_\_\_\_.
- *C:* How should children be: should they be like Tommy, who likes to succeed in all? \_\_\_\_, or should they be like Adam, who does not strive for success in all?
- D: How do you want to be: Do you wish to be successful in all and preserve in all tasks, like Tommy? \_\_\_\_, or would you rather be like Adam who does not mind about being successful in all \_\_\_?

*Scoring.* Each item in the questionnaire is assigned a dichotomy score of one or two. A score of two was given when the participant chose the theme of interest and score one was given when the opposite theme was chosen. The CO-JIA questionnaire provides two kinds of scores' sets:

1. The means for each belief type across all themes. These scores represent the motivational disposition for JIA tendencies.

2. The sum for each theme across the four belief types. This set represents the relevant themes that characterize the patients and provides some insights into their psychological dynamics.

The reliability of the CO questionnaire in different medical conditions is satisfactory. For example, the value of Cronbach's alpha was 0.97 for the CO-Anorexia questionnaire (Kreitler *et al.*, 2003), 0.72 *for the* CO-type 2 diabetes questionnaire (Kreitler *et al.*, 2004), and 0.93 for the CO-Asthma questionnaire (Roth & Kreitler, 2020). In this study, the value of Cronbach's alpha based on the four belief types was 0.64, which indicates an acceptable moderate level of internal consistency (Daud, Khidzir, Ismail & Abdullah, 2018).

The rationale behind the JIA-CO questionnaire format. The format of the JIA-CO questionnaire for pediatric patients was designed in line with several recommendations to vield accurate responses from children in selfreported questionnaires (Mellor & Moore, 2014). First, since there are previous findings that reported an association between using short stories and enhanced reading comprehension among children (Okumuf-Ceylan, 2016; Puji-Handayani, 2013), we decided to present the items in the JIA-CO questionnaire as short stories in order to make the abstract concepts of the items (e.g., being over-sensitive or being successful) easier for the children to understand. Notably, this format of short stories was satisfactorily applied in a study focused on identifying a set of psychological characteristics of children with inflammatory bowel disease (Badarnee et al., 2022). Second, regarding the use of the dichotomous response scale, there is evidence that children show a full understanding of questionnaires when the response options are presented on a dichotomous scale, more than on other scales, such as a Likert scale (Mellor & Moore, 2014; Tomlinson, Hyslop, Stein et al., 2019). And third, it is also suggested to utilize a word-based response format, such as "agree" and "disagree" instead of numbering formats, to measure abstract concepts, such as beliefs, emotions, and attitudes (Mellor & Moore, 2014). Thus, all these recommendations were applied in the process of developing the JIA-CO questionnaire with the purpose of constructing a self-reporting tool that meets the children's cognitive capacities. Therefore, the items in this questionnaire were presented as short stories and dichotomous word-based response scale.

The procedure used for identifying the themes. The CO-JIA questionnaire was constructed in line with the standard procedure in a pretest with 12 patients selected from the same clinic. The procedure is

based on interviewing the participants in line with specific guidelines that consist of four interviewing phases (Kreitler, 2019). The first deals with the interpersonal meaning of the disease and is followed by three phases with each of them based on the answers previously obtained. These answers are divided into keywords and the patients requested to explain the personal meanings of each. For instance, during an interview with a 13-year-old girl with JIA, she expressed the interpersonal meaning of her diagnosis "that it is a painful and restrictive disease." This meaning communication was used by extracting two keywords for the later interview's phases "painful" and "restrictive." In the next phase, the patient was asked to express the personal meanings of each previous keyword. For example, "what does painful mean to you? What it is for you, regardless of what it means to others?" The patient's response was "It is hard and bothers me; I cannot jump or do many things." Then, the participant was asked to elaborate and explain the personal meanings of the last answer, whereby the response was "It is a challenge." In the last phase, the patient was asked about the personal meaning of "challenge." In this case, the interviewee's response was "something you are not good at, but you should persevere and succeed." The last response was considered as an underlying personal meaning of JIA, and it marks the end of this track of questioning. The same procedure of questioning was repeated regarding each keyword separately. The responses provided by the subjects in the final stage of interviewing are classified for similarity in content, while those classified in the same category are accepted as themes

This procedure of the interviews yielded 26 themes. Table 1 shows the themes' labels.

## The medical data

The following variables were collected from the medical reports: (1) JIA type; (2) the medical treatment type; (3) the number of the inflamed joints; (4) the disease duration; and (5) the ages of the patients at the diagnosis.

#### Procedure

The CO-JIA questionnaire was administered to the patients on the clinic's premises during their regular follow-ups, and the control subjects were interviewed in the framework of the school. Before responses, an experimenter introduced the study purpose to the children, providing a pertinent explanation to the parents, and did ask them to sign an informed consent form. The form specified that the responses to the questionnaires are confidential, and the participants have the choice of not participating or to withdraw from the study at any point without any effects on the treatments or the services received in the hospital or school. The current study was conducted in line with Helsinki guidelines and approved by the appropriate ethics committees in the hospital (approval numbers: 1832) where the patients were recruited, and by the ministry of education (approval numbers: 9586), which is in charge of the school where the control subjects were recruited.

#### Statistical analysis

Descriptive statistics (i.e., means and SDs) were calculated for each study variable. Pearson correlation coefficients were calculated between relevant independent variables to identify the interrelationships between them. *t*-tests, multiple analysis of variance, and a logistic regression were applied to examine the study hypotheses.

## RESULTS

Preliminary analyses were conducted to find out the interrelations among the four belief types (the independent variables of the first hypothesis), and to examine whether the psychological tendencies are autonomous, namely, associated with JIA but not with the medical parameters of the disease. The results showed that four out of six correlations between the four belief types were significant (Table S1 in the supplementary materials). The range of their shared variances was between 11.56% and 19.36%. Table S2 summarized the correlations between the psychological tendencies and the medical parameters of JIA patients. Significant correlations were found between  $T_5$ ,  $T_{10}$ , and the number of the inflamed joints; between  $T_{13}$ ,  $T_{20}$ , and JIA duration. These the disease; and between  $T_{13}$ ,  $T_{20}$ , and JIA duration. These themes ( $T_5$ ,  $T_{10}$ ,  $T_{11}$ ,  $T_{13}$ , and  $T_{20}$ ) were excluded from the analyses of the second hypothesis that focused on autonomous themes.

The first hypothesis examined the role of the four belief types constituting the motivational disposition toward psychological tendencies for JIA. This hypothesis was tested by a model of a multiple analysis of variance (MANOVA) that yielded significant differences between children with JIA and others in the control group (F [4, 72] = 24.89, p < 0.001, Wilks'  $\Lambda = 0.42$ , partial  $\eta^2 = 0.58$ ). The procedure of Holm's sequential Bonferroni (Holm, 1979) was applied for multiple comparisons correction. Figure 1 shows that the mean score of JIA patients is higher than the control subjects in all belief types and that the range of their effect sizes (between 0.20 and 0.34) is large (Richardson, 2011).

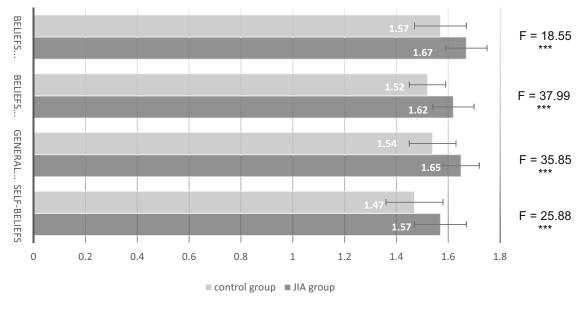
The second hypothesis that dealt with characterizing specific themes of children with JIA was tested in two stages. First, *t*-tests were conducted, as preliminary analyses, to determine which themes have the potential to be selected (based on *p*-value <0.005) for the second stage with the purpose of running a prediction model to characterize the patients in terms of the themes. Table 3 summarizes the tests' results and shows that 10 themes differentiate between the two groups with a *p*-value

<0.005. The range of their effect sizes (between 0.05 and 0.21) indicates medium to large effects (Richardson, 2011).

Second, a backward stepwise logistic regression analysis was performed to identify the categorization of the participants into one of the two groups, based on the themes which differentiated between the patients and the control children with *p*-value <0.05 (Table 3) as predictors. Table 4 shows that six themes were included in the final model, whereas the other four themes were excluded during the analysis steps. The overall model was statistically significant ( $\chi^2$  [7] = 51.90, *p* = 0.000), and showed that being in the patients' group is associated with an increased likelihood of being characterized by the themes at 3.24 to 9.35 times. In addition, the model allows the identification between the two groups with an accuracy of 89.1%, namely, 39.1% better than would be possible based on chance (50%). The difference between the two percentages (50% vs. 89.1%) is significant ( $\chi^2$  [1] = 22.92, *p* = 0.000).

## DISCUSSION

This paper dealt with identifying specific psychological tendencies of JIA defined by four belief types that refer to a set of specific themes. The first belief type is about self which expresses information about oneself, for example, "I dislike sharing my feeling with others." The second belief type is general beliefs which express information about reality and others, for example, usually, people do not share their feelings with others. The third belief type is about rules and norms which express an individual belief about social rules and standards, for example, one should not share feelings with others. The fourth belief type is about goals that express one's wants and desires, for example, "I do not



\*\*\* p < .001.

*Fig. 1.* The results of MANOVA analysis based on comparing the means and the *SD*s of Patients with JIA (N = 36) and the Control Group (N = 41) in regard to the four belief types. \*\*\*p < 0.001. The X-axis represents the mean scores of each belief type. The means of each group are presented within the bars. The error bars represent the standard deviations. The *p*-values were adjusted in line with the criteria of Holm's sequential Bonferroni procedure (Holm, 1979). The four comparisons were statistically significant in accordance with this procedure. The degree of freedom in each *F*-test = 1. The effect size was calculated in terms of partial eta squared (Richardson, 2011). It is 0.20, 0.34, 0.31, and 0.26 for beliefs about self, general beliefs, beliefs about norms, and goals, respectively. JIA = juvenile idiopathic arthritis; MANOVA = multiple analysis of variance.

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Table 3. Comparing the means and the SDs of patients with JIA and the control group in regard to the themes

JIA patients $(N = 36)$			Control group $(N = 41)$				
Num.	М	SD	М	SD	t (df)	<i>p</i> -value	Effect size <sup>a</sup>
T <sub>1</sub> <sup>b</sup>	6.67	0.72	6.15	0.83	2.88 (74)	0.005	0.10
T <sub>2</sub>	6.06	0.68	5.68	0.72	2.31 (74)	0.024	0.07
T <sub>3</sub>	6.25	1.20	5.67	1.22	2.06 (74)	0.043	0.05
$T_4$	6.83	1.18	6.09	1.49	2.41 (74.23)	0.019	0.07
T <sub>6</sub>	5.42	1.27	4.92	0.88	1.93 (61.74)	0.058	0.05
T <sub>7</sub>	6.06	0.81	5.75	1.00	1.43 (72)	0.156	0.03
T <sub>8</sub>	5.74	0.74	5.43	0.68	1.86 (72)	0.067	0.05
T <sub>9</sub>	6.88	0.88	6.57	1.10	1.33 (71.70)	0.188	0.02
T <sub>12</sub>	7.17	1.01	6.90	0.92	1.21 (73)	0.230	0.02
T <sub>14</sub>	7.70	0.59	7.56	0.68	0.88 (70)	0.382	0.01
$T_{15}^{b}$	6.18	1.16	5.12	0.97	4.19 (70)	0.000	0.20
T <sub>16</sub>	5.91	1.07	5.43	0.94	2.00 (70)	0.050	0.05
T <sub>17</sub>	5.88	0.96	5.61	1.06	1.12 (70)	0.265	0.02
T <sub>18</sub>	7.21	0.99	7.02	1.03	0.79 (72)	0.433	0.01
T <sub>19</sub>	5.42	0.66	5.27	0.56	0.99 (67)	0.326	0.01
T <sub>21</sub>	7.27	0.72	6.87	0.78	2.24 (72)	0.028	0.07
T <sub>22</sub>	7.16	0.88	6.91	0.86	1.13 (67)	0.264	0.02
T <sub>23</sub>	5.78	0.87	5.44	0.76	1.71 (68)	0.091	0.04
T <sub>24</sub>	6.91	0.58	6.48	0.79	2.61 (68.68)	0.011	0.09
T <sub>25</sub>	6.67	0.89	6.31	0.84	1.71 (69)	0.092	0.04
$T_{26}^{\ b}$	5.82	1.04	4.92	0.72	4.15 (55.58)	0.000	0.21

*Notes*: Num = theme's number; T = theme. The themes labels are presented in Table 1. T<sub>5</sub>, T<sub>10</sub>, T<sub>11</sub>, T<sub>13</sub>, and T<sub>20</sub> were excluded from this analysis due to their association with the medical parameters of JIA (see Table S2).

<sup>a</sup>The effect size was calculated in terms of partial eta squared. The values of 0.00, 0.05, 0.13 indicate small, medium, and large effects, respectively (Richardson, 2011).

 ${}^{b}T_{1}$ ,  $T_{15}$ , and  $T_{26}$  were significant to a satisfactory level of the criteria of Holm's sequential Bonferroni procedure (Holm, 1979).

want to share my feelings with others" (Kreitler, 1999, 2019). The contents of the belief types are called themes which represent the cognitive contents (e.g., sharing feelings with others) of the beliefs relevant to the disease (Kreitler, 1999, 2019). The results of this study showed that the joint vector which consists of the four belief types significantly differentiated between children with JIA and others in the control group. These results match the motivational component of the COH model, which points out the necessity of at least three belief types for establishing the psychological motivational disposition for specific tendencies relevant to a disease (Kreitler, 1999, 2019). The role of the four belief types in regard to differentiating between children with JIA and controls accords with previous studies that reported a similar role in patients with asthma (Roth & Kreitler, 2020), inflammatory bowel diseases (Badarnee et al., 2022), type 2 diabetes (Kreitler et al., 2004), and in related to health behavior, such as attending sport activities (Badarnee, Aslih, Goldman & Kreitler, 2020).

Those belief types refer to a set of specific themes relevant to a particular disease and represent the psychological dynamics that characterize patients diagnosed with that disease (Kreitler, 1999, 2019). Accordingly, this paper found that children with JIA are characterized by the following themes: being free without

Table 4. A backward stepwise logistic regression analysis of classification of the participants into the correct group (JIA versus control) based on the themes<sup>†</sup> that yielded significant results in the t-tests

Themes <sup>‡</sup>	$B^{\S}$	SE	Exponent $B^{\parallel}$	95% CI for exponent <i>B</i> LL-UL	р
T <sub>1</sub>	1.17	0.57	3.24	1.06-9.90	0.039
T <sub>2</sub>	2.24	0.83	9.35	1.83-47.85	0.007
T <sub>3</sub>	1.29	0.53	3.62	1.27-10.30	0.016
T <sub>4</sub>	0.77	0.40	2.16	0.99-4.71	0.053
T <sub>6</sub>	2.06	0.74	7.86	1.85-33.42	0.005
T <sub>15</sub>	2.06	0.66	7.82	2.13-28.75	0.002
T <sub>24</sub>	1.52	0.72	4.57	1.12–18.59	0.034

*Notes*: The regression model is significant ( $\chi^2$  [7] = 51.90, p = 0.000), n = 64,  $R^2 = 0.74$ . % of correct predication = 89.1%. The differences between this % of correct predication (89.1%) and the % of chance prediction (50%) is significant ( $\chi^2$  [1] = 22.92, p = 0.000). CI = confidence interval; JIA = juvenile idiopathic arthritis; LL = lower limit; UL = upper limit; T = theme.

<sup> $\dagger$ </sup>See the results of *t*-tests in Table 3. The themes' labels are presented in Table 2.

<sup>‡</sup>The themes  $T_{16}$ ,  $T_{21}$ , and  $T_{26}$  were excluded during the steps of the regression analysis.

<sup>§</sup>Since the *B*'s values in this kind of regression are in log-odds units, it is suggested to interpret them based on the values of the Exponent *B*'s.

Exponent *B*'s values express how many times each theme increases the likelihood of being in the JIA group. The broad range in the Exponent Bs values of the themes may indicate that the themes have various levels of functioning as psychological tendencies of the patients. For example, since the likelihood of the patients being characterized by  $T_2$  is 9.35 and by  $T_3$  is 3.62 times more than the controls, this may indicate that  $T_2$  is more established than  $T_3$  in the psychological profile of children with JIA. Thus, it seems that the tendencies contribute at different intensities or levels to the psychological profile of children with JIA.

limitations (T<sub>1</sub>); being successful and persistent in everything (T<sub>2</sub>); accepting help and advice from others (T<sub>3</sub>); not caring about doing tasks well (T<sub>6</sub>); being over-sensitive and avoid being insulted or hurt (T<sub>15</sub>); and finally, befriending everyone and like being close (T<sub>24</sub>).

The identified themes affect the psychological state of patients and function, on an ongoing basis, as a source of stress and tension (Kreitler, 2019). More particularly, continuously striving for success and strictly persistent in every task (T2) is an aspect of perfectionism (Hewitt & Flett, 2002) which is associated with stress (Flett, Nepon, Hewitt & Fitzgerald, 2016), depression (Mandel, Dunkley & Moroz, 2015), and health difficulties (Molnar, Sadava, Flett & Colautti, 2012). In addition, the tendency of being an over-sensitive person (T15), associated with mood disorders (Liss, Timmel, Baxley & Killingsworth, 2005) and internalizing problems (Boterberg & Warreyn, 2016), contributes to the stressful psychological dynamics of the patients. And finally, the tendency of not fulfilling duties well (T<sub>6</sub>) seems to reflect on the social life quality of the patients since not doing responsibilities at home, school, and other settings is associated with social conflicts and interpersonal problems (Bulotsky-Shearer, Fernandez, Dominguez & Rouse, 2011).

Another contributor to the patients' stressful dynamics is reflected by the contradicted tendencies that express individuals' cognitive conflicts (Festinger, 1957; Harmon-Jones & Mills, 2019). The tendency of children with JIA to like being free without limitations (T<sub>1</sub>) opposes their motivation to success and persevere in tasks (T<sub>2</sub>). Moreover, it seems that striving for success (T<sub>2</sub>) contrasts with not fulfilling duties or tasks well (T<sub>6</sub>). Keeping close relations with others (T<sub>24</sub>) could be challenging and even impossible when one is over social-sensitive (T<sub>15</sub>). Thus, this kind of dynamics constitutes, continuously, a focus for stress and conflicts, and it is in agreement with previous studies that suggested an association between cognitive conflicts and health problems (Badarnee *et al.*, 2022; Compan, Feixas, Varlotta-Domínguez *et al.*, 2011; Dada, Feixas, Compañ & Montesano, 2012; Feixas, Montebruno, Dada, Del Castillo & Compan, 2010).

Besides, the results of this study could be essential in the clinical context where personality factors play a role in establishing the health-workers-patient relationships, which were found to influence illness trajectories, treatment adherence (May, Allison, Chapple et al., 2004), and care quality (Bensing, 1991; Street, Gordon & Haidet, 2007). Examples of interaction styles in the medical field are the egalitarian style preferred by assertive patients and the paternalistic interaction style which was reported as optimal for powerful-others patients (those who attribute control and power to people in positions of authority, such as doctors and other health professions) (Braman & Gomez, 2004). Therefore, the identified tendencies in this study could enhance the psycho-medical care quality by suiting health care workers' interactions and communication styles to the patients' needs. For example, the tendency of children with JIA to easily accept help and advice (T<sub>3</sub>), and befriending others (T<sub>24</sub>) may reflect their preference for a non-formal communication style in a clinical setting. In addition, the over sensitivity of the patients  $(T_{15})$  may request psychologists and other health professions to be particularly sensitive to the patients' non-spoken needs in the clinical encounters.

Thus, the identified tendencies provide an insight into the patients' stressful psychological dynamics and deepen our understanding concerning the preferred JIA patients' interactions and communications style. These tendencies seem to be stable under different conditions of the disease since they are not associated with the patients' age at the onset, the duration, and the severity of JIA. That in no way proves that there are cause-effect relations, but it indicates that the identified psychological tendencies are stable regardless of the disease itself rather than with its state.

## Implications

The results of this paper add another layer to understanding the sources of stress reported by previous studies as a characteristic of patients with JIA (Henoch *et al.*, 1978; Neufeld *et al.*, 2013). The identified tendencies could be essential in the stress management domain since it seems likely that patients with JIA would feel threatened or tensioned, for example, when they do not perfectly succuss in all tasks and life domains ( $T_2$ ) or due to a tendency of being overly sensitive ( $T_{15}$ ) and their preference not to perform tasks well ( $T_6$ ). Therefore, to reduce stress, it is advisable to make patients aware of the potential stressful effect of these tendencies and assist them to acquire optimal coping strategies to deal, for

example, with aspects of perfectionism as reflected in T2. Another aspect of stress management refers to decreasing the intensity of tendencies related to conflicts or those that make individuals venerable to stress (Badarnee et al., 2022; Kreitler, 2019). Since the COH model provides practical guidelines for changing psychological tendencies, it is possible to refine or reduce the extent to which the patients support a theme by mobilizing or inhibiting their beliefs concerning that theme (Kreitler, 2019). For example, when the theme related to a conflict is not caring about doing tasks well  $(T_6)$ , then the intervention will deal with recalling situations or tasks that the patient performed well (reflects a belief about self), upon inquiring about tasks that the child wish to be able to perform well (reflects a belief about goals), about tasks that usually people perform well (reflects a general belief), and finally, about kinds of tasks that should be performed well (reflects a belief about norms). The effect of this theme on the patient will be reduced when four or at least three of their belief types referring to that theme are changed (Kreitler, 1999). However, changing a single theme is insufficient to bring about a significant effect on the patient's stress level. The success of the training depends on establishing a stable and wellgrounded motivational disposition representing the four belief types regarding a sufficient number of relevant themes (Kreitler, 2019).

Finally, psychologists and other health care workers could benefit from the identified tendencies by suiting their interactions and communication style to the patient needs to enhance their collaboration, adherence, and clinical quality care. It would be advisable to keep aware of the patients' tendency not to care about doing tasks well ( $T_6$ ) and initiate support programs to encourage their performance in related to medical treatment tasks. In addition, sharing information and involving the patients in medical decisions processes could be essential for cultivating their sense of being free and independent (refers to  $T_1$ ), and using an informal conversations style ( $T_{24}$ ) seems to be optimal in the clinical ground for children with JIA who willing to accept advice and help ( $T_3$ ). Finally, it is also suggested to be aware of patients' over sensitivity ( $T_{15}$ ) in different clinical settings and disease states, such as during treatment courses and inflamed periods.

## Related to the disease state tendencies

Besides the autonomous tendencies (not associated with the disease state), the supplementary results of this study showed that there are several tendencies related to the JIA state. The count of the inflamed joints is negatively associated with the tendency of not caring about being loved by others (T<sub>5</sub>) and positively with liking to be different and unique  $(T_{10})$ . Thus, it seems that when the state of the disease is severe - in terms of the number of inflamed joints - the patients try to spend more time with others, probably as an attempt to benefit from the social support, as reported by previous studies as a disease resilience source associated with positive outcomes of chronic diseases management (Fuertes, Rubinstein, Yarandi & Cohen, 2021) including JIA (Ahola Kohut, Forgeron, Amaria et al., 2016; Hynes, Saetes, McGuire & Caes, 2019; Ireys, Sills, Kolodner & Walsh, 1996). Concerning the tendency of feeling different and unique  $(T_{10})$ , it seems that the increased number of inflamed

joints restricts the moving abilities and life activities of the patients (Malattia & Martini, 2020; Martini, Lovell, Albani *et al.*, 2022; Packham & Hall, 2003), which strengthens this tendency among them. This may indicate an aspect of acceptance of the disease consequences, such as movement limitations and pain which apparently deepen the feeling of difference and uniqueness among the patients.

In addition, the supplementary results showed that the age of the patients at the onset of the disease is negatively associated with the tendency of planning things up to the smallest details  $(T_{11})$ . Thus, it seems that patients who were diagnosed at a young age had more frequent contact with health facilities and health professionals, which made them more likely to be given repetitive instructions concerning managing the disease and adapting their lifestyle (Dineen-Griffin, Garcia-Cardenas, Williams Benrimoj, 2019) in line with the state of the disease. And finally, the duration of the disease is negatively associated with the tendency to like moving freely without limitations  $(T_{13})$  and positively associated with having a lot of adventures and fun  $(T_{20})$ . Therefore, it seems that a longer disease duration enables adaptation to JIA symptoms including accepting moving limitations and physical pain. A systematic review study focused on resilience resources for coping with JIA among 6-18 years old children with JIA, found that pain acceptance plays an important role in relation to positive psychological and physical outcomes among the patients (Hynes et al., 2019). The acceptance of the disease symptoms and the physical limitation may be compensated by the tendency to like having a lot of adventures and fun which was found in this study as positively associated with the disease duration.

Thus, the supplementary results of this study indicate that there are several psychological tendencies associated with the state of the disease. These tendencies, which could be essential for deepening our understanding concerning the psychological coping throughout JIA prognosis, point to the need for further investigations that focus on the psychological tendencies related to the state of the disease.

#### Limitations and future studies

The findings of this paper must be seen in the light of some limitations. First, this study was based on a convenience sample recruited in a certain medical center. Not all the diagnosed children in other hospitals had the opportunity to participate in the study. Thus, future multi-center studies based on random samples are suggested. In addition, in line with epidemiological studies which found that the most JIA patients are diagnosed with Oligoarticular subtype (Al-Mayouf, Al Mutairi, Bouayed et al., 2021; Consolaro, Giancane, Alongi et al., 2019), most patients (52.77%) in this study were also diagnosed with this subtype. However, the number of patients diagnosed with other JIA subtypes was insufficient to compare them in regard to their psychological tendencies. Therefore, it is suggested that future studies focus on comparing the psychological characteristics of each subtype of JIA. Second, this is a case-control cohort study that identified autonomous psychological tendencies without examining the stability of these tendencies over time. Therefore, a longitudinal study that follows the patients' psychological

tendencies over time is suggested. And third, the interpretation of this study's results was based on the theoretical framework of the COH model and previous studies' findings concerning specific personality tendencies and stress. The stressful aspects of the identified psychological tendencies require empirical validation, and future studies are suggested to focus on it and the involvement of these tendencies in doctor-patient relationships.

In conclusion, the results of this paper add another layer to our understanding concerning the psychological tendencies of children with JIA. These tendencies deepen our knowledge regarding the patients' stressful psychological dynamics and their preferred relationships and communication style with physicians and health professions.

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## DATA AVAILABILITY STATEMENT

The dataset of this study is available in Figshare, doi: 10.6084/ m9.figshare.11888766

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#### SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article:

**Table S1.** The interrelations between the four belief types in the CO-JIA Questionnaire

**Table S2.** The correlations<sup> $\dagger$ </sup> between the psychological and some of the medical variables.

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