Medicine

OPEN

Exploring the relationship between personality and chronic pain in adults with osteogenesis imperfecta
A cross-sectional study

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Abstract

Despite the growing body of research on chronic pain in adults with osteogenesis imperfecta (OI), there is still a lack of comprehensive understanding of the influence of psychological factors on pain experienced by individuals with this condition. This study aims to delve into the correlation between personality traits and various aspects of pain, such as frequency, intensity, appraisal, and coping mechanisms, in a significant sample of adults with OI. Additionally, the investigation seeks to identify whether certain personality profiles may be more susceptible to chronic pain within this specific population. A descriptive cross-sectional study was conducted on a sample of 418 adults diagnosed with OI. Participants completed an online survey that assessed sociodemographic and clinical variables, pain parameters, personality traits, pain appraisal, and coping strategies. Subsequently, descriptive, correlational, cluster and comparative analyses were performed. Up to 83% of the participants reported experiencing pain on a regular basis. Regarding personality dimensions, moderate scores were obtained, with no significant differences compared to the general population. Neuroticism emerged as the trait showing the most robust relationships with the evaluated variables. It positively correlated with pain intensity, frequency, and the perception of pain as threatening ($P < .001$). Conversely, higher levels of extraversion were associated with a reduction in pain and its threatening perception ($P < .001$). Finally, the cluster analysis revealed a personality profile that showed greater vulnerability in pain adaptation, characterized by high levels of neuroticism and low levels of extraversion, agreeableness, and conscientiousness. Chronic pain is prevalent in adults with OI. Personality dimensions maintain a significant relationship with this pain, acting as vulnerability or protective factors. Consequently, specific personality profiles are associated with poorer adaptation. Understanding these profiles would allow for a deeper comprehension of the pain experience in adults with OI.

Abbreviations: AHUCE = Asociación Nacional Huesos de Cristal, NEO-FFI = NEO five-factor inventory, OI = osteogenesis imperfecta, SD = standard deviation.

Keywords: chronic pain, osteogenesis imperfecta, personality, threat appraisal

1. Introduction

Osteogenesis imperfecta (OI) is a genetic disorder that affects the connective tissue, specifically type I collagen. It is characterized by significant bone fragility.[1] In addition to frequent bone fractures, OI is associated with other clinical manifestations such as dentinogenetic imperfecta, skin hyperlaxity, joint hypermobility, short stature, long bone deformities, hearing, and vision loss, cardiovascular and respiratory complications, as well as chronic pain.[2,3] To date, more than 15 distinct genetic mutations have been identified, each responsible for a specific type of OI.[4] However, simplified classifications are used in clinical practice, such as the 1 proposed by Van Dijk and Sillence,[5] which includes the following types: Type I, the most common and considered the mildest form of OI, with a lower incidence of fractures and bone deformities; Type II, an extremely severe form of OI, with a high mortality rate during pregnancy and in the first 4 weeks of life; Type III, a severe variant with multiple fractures, skeletal deformities, short stature, hearing problems, and more frequent cardiorespiratory complications; Type IV, of

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variable severity, with recurrent fractures, osteoporosis, bone deformities, and scoliosis; Type V, which exhibits moderate to severe bone fragility and a higher predisposition to developing hyperplastic callus after a fracture. OI is considered a rare disease, with a prevalence of 1 affected individual per 10,000 births.[6]

Numerous recent studies have consistently demonstrated the prevalence of chronic pain in adults with OI.[7–9] This particular type of pain has been shown to be resistant to both surgical and pharmacological interventions[10] and has been associated with functional limitations, sleep disturbances, fatigue, and a decreased health-related quality of life.[7,9]

Chronic pain, characterized as persistent or recurrent pain lasting for more than 3 months,[11] represents a significant source of stress in the healthcare domain.[12] Lazarus and Folkman Transactional Model of Stress[13] is frequently employed as a theoretical framework in the study of chronic pain from a psychological standpoint. This model allows for the exploration of various factors including environmental and individual characteristics, cognitive processes, behavioral responses, and subsequent consequences.[14] According to this model, pain and individual characteristics interact to create a stressful situation, initiating an appraisal and coping process that ultimately leads to consequences affecting the initial situation.[15] In a prior study involving adults with OI,[16] it was previously reported that a threatening appraisal of pain was associated with increased pain intensity and interference with daily activities. These variables were, in turn, linked to a decline in health-related quality of life. However, the initial study did not extensively examine individual-related factors beyond clinical and sociodemographic variables.

In relation to individual variables, personality dimensions have been frequently assessed in stress studies since the 1980s.[17–21] This trend aligns with the proposal of the 5-Factor Model of Personality by Costa and McCrae,[22] a model that encompasses 5 personality dimensions with a significant genetic influence, characterized by an endogenous nature and independent development from environmental events.[23] In summary, the 5 dimensions include Neuroticism, which represents a tendency to experience unpleasant emotions such as anxiety, sadness, or anger; Extraversion, describing sociable, assertive, and optimistic individuals; Openness to Experience, reflecting a willingness to accept new or unconventional ideas and to experience both negative and positive emotions; Agreeableness, indicating individuals who are empathetic and enjoy helping others; and Conscientiousness, describing individuals who are diligent, punctual, reliable, and exhibit outstanding academic and/or work performance.[24]

In chronic pain research, higher levels of neuroticism have consistently been associated with increased pain intensity, a more catastrophic and threatening appraisal of pain, the use of passive coping strategies, and poorer adaptation, characterized by limitations in physical activity and higher levels of depression and anxiety.[25–28] The other personality dimensions, although to a lesser extent, also maintain significant relationships with variables relevant to chronic pain. For instance, higher levels of extraversion have been linked to lower perceived pain intensity and interference in daily activities,[29] the use of active coping strategies,[30] and a lower tendency to avoid pain.[26] Similarly, openness to experience has been associated with an increased use of active coping strategies[28] and a lower tendency for catastrophizing and distress.[31,32] Individuals with a higher level of conscientiousness exhibit lower pain frequency,[33] reduced opioid consumption, improved adherence to medical treatment,[34,35] and a predominant use of active coping strategies.[28] Finally, agreeableness appears to have weaker associations with variables such as coping, appraisal, and consequences of chronic pain.[15,26,36]

Several studies have aimed to identify resilient or vulnerable profiles in individuals facing chronic pain by grouping them based on their scores on the mentioned personality dimensions.

Indeed, it has been observed that individuals with high levels of neuroticism, low scores on extraversion, openness to experience, and conscientiousness, and moderate levels of agreeableness, experience higher levels of pain and analgesic consumption, sleep difficulties, lower levels of activity and functionality, and poorer quality of life across various domains, including physical activity, vitality, social relationships, and mental health.[15,37]

Although there has been increasing research on chronic pain in adults with OI, there remains limited knowledge regarding the role of psychological variables in pain within the context of this condition. The aim of this study is to examine the relationship between personality dimensions and the frequency, intensity, appraisal, and coping of pain in a large sample of adults with OI, using data collected from a previous investigation.[16] Furthermore, we will seek to identify the potential existence of a personality profile that is more vulnerable to chronic pain in this population.

2. Materials and methods

The present study emerged from a subsequent analysis of data collected in the previously published study titled "Chronic pain in adults with osteogenesis imperfecta and its relationship to appraisal, coping, and quality of life: A cross-sectional study."[16] Therefore, the sample, methodology, and instruments described below are those referenced in the mentioned publication.

The study was approved by the Committee of the Doctoral Program in Clinical and Health Psychology to the University of Valencia. All participants provided their informed consent regarding their participation in the study.

2.1. Sample

The sample consists of 418 adults with OI from 36 different countries who, having granted their consent, completed an online questionnaire battery hosted on the servers of the Asociación Nacional Huesos de Cristal (AHUCE) Foundation.

2.1.1. Inclusion criteria.

(i)   Be at least 18 years of age.
(i)   Having an OI diagnosis.
(iii) Having access to the internet.

2.1.2. Exclusion criteria.

(i)   Being under 18 years of age.
(ii) Not having an OI diagnosis.

2.2. Procedure

Initially, an online survey was developed and hosted on private servers of the AHUCE Foundation. The survey consisted of an informed consent form and several questionnaires addressing the variables of interest in the study. To ensure participant accessibility, the survey was available in both English and Spanish. When questionnaires were not available in both languages, the method of forward-backward translation was used, followed by subsequent statistical validation to ensure equivalence of the translated versions. The estimated time to complete the questionnaire battery was approximately 20 minutes, and participants had the flexibility to access and complete the survey from a computer, tablet, or mobile phone without the presence of an evaluator. Furthermore, anonymity of the collected data was guaranteed.

The survey dissemination among the target population was carried out by the AHUCE Foundation and other collaborating entities, such as the AHUCE and the Asociación Madrileña de Osteogénesis Imperfecta in Spain and Latin America, the

Brittle Bone Society in the United Kingdom, the osteogenesis imperfecta foundation in the U.S., and the European Federation of Osteogenesis Imperfecta, as well as other organizations in various European countries. The distribution was conducted through different channels, including social media, emails, brochures, and communications at conferences and events.

2.3. Assessment instruments

2.3.1. Sociodemographic variables.
The sociodemographic questionnaire employed in this research was developed by the research team and consists of 4 items assessing participants age, gender, type of OI, and country of residence. The questionnaire incorporates a varied response format, including open-ended questions and multiple-choice items.

2.3.2. Chronic pain and its characteristics.
Pain frequency was assessed using a single item consisting of the question, "Do you experience pain frequently?" with 5 response alternatives: no, once a month, several times a month, several times a week, or every day. Additionally, participants were asked to report the duration of their pain using an open-ended response format.

Pain intensity, location, and type were evaluated using the pain detect questionnaire[38] in its original English version and its adaptation and validation in Spanish.[39] This questionnaire comprises 4 sections. Section 1 includes 3 visual analogue scales with 11 possible points each, assessing current pain, worst pain experienced in the past 4 weeks, and average pain during the same period. Section 2 consists of an item featuring a graph accompanied by a legend, inquiring about the participant's pain pattern over time, with 4 response options: "constant pain with slight fluctuations," "constant pain with pain attacks," "pain attacks without pain between attacks," and "frequent pain attacks with pain between attacks." Section 3 includes a drawing depicting a front and back view of a human figure, accompanied by 3 items investigating pain location and radiation. Finally, Section 4 consists of 7 items with a 6-point Likert format, exploring different pain characteristics in the marked areas, such as burning and tingling sensations, sensitivity to touch, presence of sudden pain attacks, sensitivity to cold or heat, numbness, and sensitivity to light pressure. The response options for these items are: "no," "mild," "very mild," "moderate," "intense," and "very intense."

In the validation of the original questionnaire, the authors obtained a significant bivariate correlation between items ($P <$ .01) and satisfactory internal consistency (Cronbach alpha = 0.83).[38] The Spanish adaptation also presents good psychometric indices, with a Cronbach alpha of 0.86 and a test-retest intraclass correlation coefficient of 0.93.[39]

2.3.3. Personality dimensions.
The NEO 5-factor inventory (NEO-FFI), an abbreviated version of the revised neo personality inventory,[40] was used to assess personality dimensions. The NEO-FFI was designed to measure the personality traits proposed in the 5-factor theory.[41,42] It consists of a 60-item questionnaire with a 5-point Likert scale format, encompassing 5 scales: Neuroticism (N), Extraversion (E), Openness to Experience (O), Agreeableness (A), and Conscientiousness (C).[43] Each factor is assessed by 12 items, and responses are assigned numerical scores. These scores are summed to derive 5 raw scores, 1 for each personality trait. Subsequently, they are transformed into standard scores using a normative table, allowing for the determination of each participant's percentile in relation to the reference population for each trait.

Regarding its psychometric properties, different internal consistency indices were obtained for the original questionnaire and its Spanish validation,[44] depending on the analyzed factor. The Cronbach alphas obtained were 0.86 and 0.82 for Neuroticism, 0.77 and 0.81 for Extraversion, 0.73 and 0.76 for Openness to

Experience, 0.68 and 0.71 for Agreeableness, and 0.81 in both cases for Conscientiousness.

2.3.4. Pain assessment.
To assess the variable "pain appraisal," the pain appraisal inventory[45] was used, based on Lazarus and Folkman transactional model.[13] This instrument consists of 16 items with 6 response options (strongly disagree, moderately disagree, somewhat disagree, somewhat agree, moderately agree, and strongly agree). The pain appraisal inventory comprises 2 factors: threat appraisal, which refers to a negative interpretation of pain and is typically associated with unpleasant emotions and greater activity limitation, and challenge appraisal, which is linked to a more positive interpretation of pain experience and is associated with better quality of life.

In terms of its psychometric properties, the original questionnaire demonstrated good internal consistency, with a Cronbach alpha coefficient of 0.86 for the threat factor and 0.81 for the challenge factor. Since no Spanish version of this questionnaire was available, a validation in Spanish was conducted using the back-translation method. In the validated version, Cronbach alpha coefficients of 0.86 for the threat factor and 0.86 for the challenge factor were obtained.

2.3.5. Coping strategies.
The Chronic Pain Coping Questionnaire - Short Form Cuestionario de Afrontamiento al Dolor Crónico (questionnaire on coping with chronic pain)[46] was employed to assess the coping strategies utilized by participants in response to pain. The Cuestionario de Afrontamiento al Dolor Crónico (questionnaire on coping with chronic pain) consists of 24 items with 5 response options (never, seldom, neither seldom nor often, often, and always) that evaluate 6 distinct factors. These factors are as follows: religion (cognitive or behavioral strategies based on religious or spiritual aspects), distraction (behaviors aimed at diverting attention from pain), mental self-control (cognitive efforts to reduce pain), self-affirmation (self-talk aimed at improving mood), catharsis (verbal expression of pain with others in search of relief), and information seeking (searching for additional knowledge about the problem and its possible solution).

The questionnaire demonstrates high internal consistency, with satisfactory Cronbach alpha values for each factor: 0.94 for religion, 0.84 for catharsis, 0.75 for distraction, 0.80 for mental self-control, 0.77 for self-affirmation, and 0.74 for information seeking. As the questionnaire was not available in English, a translation was conducted using the forward-backward method. In the English version, McDonald omega values were obtained, yielding 0.65 for distraction, 0.77 for information seeking, 0.93 for religion, 0.82 for catharsis, 0.80 for mental self-control, and 0.79 for self-affirmation.

2.4. Statistical analysis

The Statistical Package for the Social Sciences (SPSS) 21.0 software was used for statistical analyses. Descriptive and frequency analyses were conducted to examine the characteristics of the sample and their scores on the different questionnaires. Pearson correlation coefficient was employed to assess the relationships between variables. Two-step cluster and k-means analyses were performed on the personality dimensions, followed by mean comparison between groups using the Student t test. Data were presented as percentages, raw scores, means, and standard deviations. The statistical significance level was set at $P <$ .005.

3. Results

3.1. Descriptive statistics

The participants age ranged from 18 to 85 years, with a mean age of 41.20 years (standard deviation [SD] = 13.81). Of the
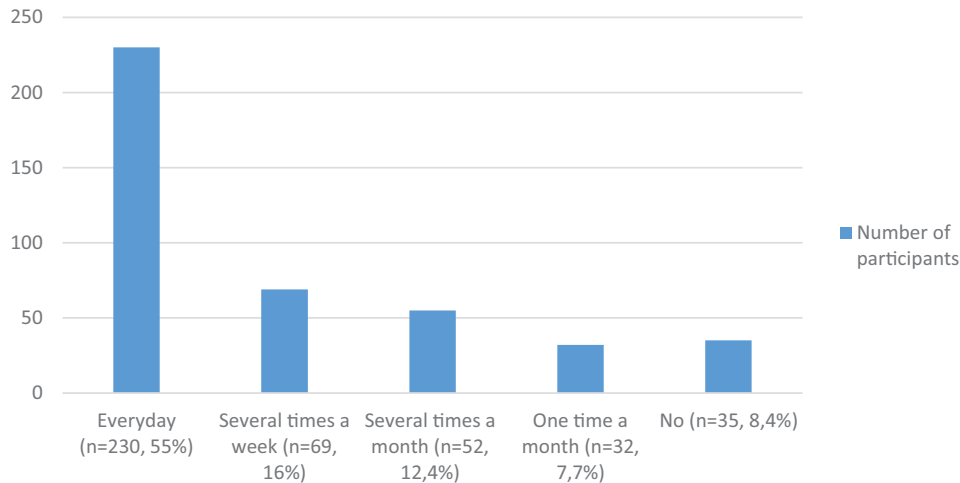
Do you experience pain frequently?

Figure 1. Pain frequency in the full sample (number of participants and % of the sample).

Table 1

Raw scores, standard scores, percentile, and T-scores of personality dimensions.

Personality dimensions	Raw score	Standard deviation	Centil	T-score	Profile rating
Neuroticism	22,7	9,08	53	50	Average
Extraversion	26,93	7,85	42,74	48	Average
O. to experience	28,5	6,16	55	51	Average
Agreeableness	31,36	6,65	36	46	Average
Conscientiousness	32,5	7,59	48	50	Average

T-Score = standardized scores of Neo-FFI.

Table 2

Correlations between personality and pain frequency and intensity.

		Pain intensity		
Personality dimensions	**Frequency of pain**	**In the current moment**	**Maximum past month**	**Average past month**
	r	r	r	r
Neuroticism	.224**	.240**	.239**	.266**
Extraversion	−.237**	−.192**	−.162**	−.173**
O. to experience	−.050	−.200	−.082	−.130**
Agreeableness	.028	.006	.015	.014
Conscientiousness	−.081	−.073	−.089	−.082

r = Pearson correlation coefficient.

participants, 75.6% were women (n = 316) and 24.4% were men (n = 102). The sample consisted of individuals from up to 36 different nationalities, with Spain (n = 118), the United States (n = 100), and the United Kingdom (n = 45) being the countries with the highest representation. The most common type of OI was type I, accounting for 45.5% of the cases, followed by type III (20.8%), type IV (16.5%), type II (4.1%), and type V (1.7%).

The frequency of pain was high, as previously described in a study with the same sample (Muñoz et al, 2022). Among the participants, 55% reported experiencing daily pain, while 16.5% experienced it several times a week and 12.4% experienced it several times a month. The remaining 16.1% stated that they did not experience pain regularly (see Fig. 1).

The results obtained from the NEO-FFI questionnaire indicated that the scores on the different personality traits were similar to those of the general population. Specifically, the mean raw score for neuroticism was 22.7 (SD = 9.08; percentile: 53; T-score = 50), for extraversion was 26.93 (SD = 7.85; percentile: 42.74; T-score = 48), for openness to experience was 28.5 (SD = 6.16; percentile: 55.45; T-score = 51), for agreeableness was 31.36 (SD = 6.65; percentile: 35.78; T-score = 46), and for conscientiousness was 32.50 (SD = 7.59; percentile: 48; T-score = 50) (see Table 1).

3.2. Correlations

Regarding the observed correlations between personality dimensions and pain, it was found that higher levels of neuroticism were significantly associated with increased pain frequency (r =

.224, $P < .000$) and current pain intensity (r = .240, $P < .000$), as well as with average pain intensity (r = .266, $P < .000$) and maximum pain intensity in the past month (r = .239, $P < .000$). In contrast, extraversion showed a significant negative correlation with pain frequency (r=−.237, $P < .000$) and current pain intensity (r=−.192, $P < .000$), maximum pain intensity in the past month (r=−.162, $P = .001$), and average pain intensity in the past 4 weeks (r=−.173, $P < .000$). The remaining correlations between personality dimensions and pain frequency and intensity can be observed in Table 2.

In relation to pain appraisal, there was a significant increase in perceiving pain as a threatening event with higher levels of neuroticism (r = .431, $P < .000$). On the other hand, perceiving pain as a threat showed a significant negative correlation with extraversion (r= −.187, $P < .000$), openness (r= −.137, $P = .005$), and conscientiousness (r= −.144, $P = .003$). The correlations between these personality dimensions and the appraisal of pain as a challenge were less pronounced and can be found in Table 3.

Finally, regarding the relationship between personality and pain coping, several significant correlations were found. Neuroticism showed positive correlations with mental self-control (r = .168, $P = .001$) and catharsis (r = .129, $P = .008$). Extraversion demonstrated significant correlations with distraction (r = .153, $P = .002$), information seeking (r = .122, $P = .012$), and self-affirmation (r = .205, $P < .000$). Openness maintained significant correlations with distraction strategies (r = .207, $P < .000$) and mental self-control (r = .161, $P = .001$). Agreeableness was negatively

related to mental self-control (r= −.123, P = .012) and positively associated with catharsis (r = .110, P = .025). On the other hand, conscientiousness established a significant correlation only with the self-affirmation strategy (r = .216, P < .000) (Table 4).

For a comprehensive understanding of the relationship between pain appraisal, coping strategies, interference in daily activities, and quality of life, we recommend referring to the study conducted by Muñoz et al[16] using the same sample.

3.3. Cluster analysis

Initially, a hierarchical cluster analysis was conducted on the personality dimensions, revealing that the data could be best classified into 2 distinct groups. To delve deeper into this finding, a k-means cluster analysis was performed, where 2 groups were selected based on the scores obtained from the NEO-FFI questionnaire. The results of the cluster analysis can be found in Table 5.

Cluster 1 exhibited significantly higher levels of neuroticism (80th percentile) compared to Cluster 2, which had low scores on this dimension (30th percentile). Additionally, Cluster 1 showed lower scores on extraversion (20th percentile), agreeableness (20th percentile), and conscientiousness (30th percentile) compared to Cluster 2, which had moderate scores on these dimensions (extraversion: 55th percentile, agreeableness: 45th percentile, conscientiousness: 60th percentile). Both clusters obtained moderate scores on openness to experience (55th percentile).

Upon comparing the 2 groups using a mean contrast, notable distinctions emerged. Cluster 1 displayed a significantly higher frequency of pain (P = .003), as well as greater intensity of pain in the current moment (P = .001), a higher average pain intensity over the last month (P < .000), and a higher maximum pain intensity in the past 4 weeks (P = .002) compared to Cluster 2. Furthermore, participants in Cluster 1 perceived pain as more threatening (P < .000) and engaged in exercise less frequently (P < .000) than those in Cluster 2. However, no significant differences were observed between the 2 groups in terms of coping strategy use, appraisal of pain as a challenge, and other clinical and sociodemographic variables (refer to Table 6).

4. Discussion

The aim of this study was to investigate the relationships between personality dimensions based on Costa and McCrae 5-Factor Model[22] and various aspects of chronic pain, including frequency, intensity, appraisal, and coping strategies, in a diverse international sample of adults with OI. This study builds upon previous research conducted with the same simple,[16] and readers can refer to that study for additional information on the topic.

The frequency of pain was high in 83.4% of the sample. Specifically, 55% of participants reported experiencing daily pain, while 16% reported pain several times a week, and 12.4% reported pain several times a month. These findings are consistent with similar studies conducted with the same population[7,47] and indicate a high prevalence of chronic pain compared to the general population. For example, in Spain, the United States, and the United Kingdom, which were the countries with the largest representation in the sample (n = 263), chronic pain is present in approximately 17.6%, 20.4%, and 34% of the general population, respectively.[48–50]

The mean scores on the 5 personality dimensions, assessed using the Neo-FFI questionnaire,[51] indicate a moderate presence of traits in the participants, with no significant differences compared to the general population. These data suggest that, despite the consensus regarding the joint influence of genetic and environmental factors on personality development,[52–54] the complications associated with OI do not seem to be significant enough to establish differences from the rest of the population.

Table 3

Correlations between personality and pain appraisal.

Personality dimensions	Threat pain appraisal r	Chellenge pain appraisal r
Neuroticism	.431**	.063
Extraversion	−.187**	.073
O. to Experience	−.068	.096*
Agreeableness	−.137**	−.021
Conscientiousness	−.144**	.100*

r = Pearson correlation coefficient.

Table 4

Correlations between personality and coping strategies.

Personality dimensions	Distraction r	Information seeking r	Mental self-control r	Self-affirmation r	Religion r	Catharsis r
Neuroticism	−.047	.044	.168**	−.004	.052	.129**
Extraversion	.153**	.122**	.007	.205**	.021	.073
O. to experience	.207**	.035	.161**	.084	−.073	−.019
Agreeableness	.024	.086	−.123*	.091	.058	.110*
Conscientiousness	.093	.092	−.038	.216**	.030	−.074

r = Pearson correlation coefficient.

Table 5

Results of the k-means cluster analysis on personality dimensions.

Dimensions	Cluster 1	Cluster 2	F	Sig.
Neuroticism	29	16	412,725	.000
Extraversion	23	31	140,231	.000
O. to experience	28	29	7192	.008
Agreeableness	29	34	58,809	.000
Conscientiousness	28	36	155,736	.000

Sig = Significance.

Table 6

Mean comparison between clusters on different variables.

Pain	Groups	Mean	SD	t	P value	Effect size
Frequency	Cluster 1	3,22	1163	3,04	.003	d = .30
	Cluster 2	2,83	1431			
Pain intensity in the current moment	Cluster 1	5,06	2,23	3,47	.001	d = .34
	Cluster 2	4,28	2336			
Maximum pain intensity past month	Cluster 1	5,55	2,17	3,16	.000	d = .35
	Cluster 2	4,76	2,28			
Average pain intensity past month	Cluster 1	7,33	2,23	3.6	.002	d = .31
	Cluster 2	6,57	2,66			
Pain appraisal						
Threat	Cluster 1	4,31	1,04	6,8	.000	d = .67
	Cluster 2	3,57	1,16			
Challenge	Cluster 1	2,83	1,14	−.71	.480	
	Cluster 2	2,91	1,23			
Coping strategies						
Distraction	Cluster 1	11,35	3,22	−.64	.524	
	Cluster 2	11,55	3,12			
Information seeking	Cluster 1	10,67	3,9	−.507	.612	
	Cluster 2	10,87	4,07			
Mental self-control	Cluster 1	10,31	4,01	1,47	.143	
	Cluster 2	9,75	3,8			
Self-affirmation	Cluster 1	13,96	3,62	−1,77	.78	
	Cluster 2	14,6	3,08			
Religion	Cluster 1	8,05	4,94	.271	.786	
	Cluster 2	7,92	5,12			
Catharsis	Cluster 1	10,04	4,08	.709	.479	
	Cluster 2	9,75	4,1			
Healthy habits						
Physical exercise	Cluster 1	0,78	0,864	−3,74	.000	d = .37
	Cluster 2	1,1	0,873			
Physical therapy	Cluster 1	0,49	0,956	.451	.652	
	Cluster 2	0,45	0,957			

d = Cohen's d value, P = probability value, SD = Standard deviation, t = t value in Student t test.

Indeed, other studies point in a similar direction, finding no significant association between chronic illnesses and personality changes,[55] or observing changes of modest magnitude (1–2 T-score units).[56]

Certain personality traits, on the contrary, have been found to contribute to the vulnerability of developing diseases,[57] their progression,[58] and engaging in health-risk behaviors.[59] Likewise, personality dimensions have frequently been associated with the presence of chronic pain. Specifically, neuroticism has demonstrated a stronger link to pain compared to other traits. Elevated levels of neuroticism have been consistently linked to higher pain intensity, increased pain frequency, a perception of pain as more threatening, and poorer pain adaptation.[25–28] Consistent with prior evidence, our study's results support these findings. As neuroticism scores increased, so did pain frequency (r = .224, P < .001) and intensity across 3 measured indices: "current moment" (r = .240, P < .001), "maximum in the last month" (r = .239, P < .001), and "average over the past 4 weeks" (r = .266, P < .001). Additionally, higher levels of neuroticism were associated with a tendency to perceive pain as more threatening (r = .431, P < .001).

The remaining personality traits also showed significant relationships with the evaluated variables, although these relationships were weaker and had opposite directions compared to those observed in neuroticism. Consistent with findings from previous studies,[29] higher levels of extraversion were associated with lower pain frequency (r = −.237, P < .001) and decreased pain intensity at the current moment (r = −.192, P < .000), maximum intensity in the last month (r = −.162, P < .000), and average intensity over the past 4 weeks (r = −.173, P < .000). Moreover, higher scores in extraversion, agreeableness, and

conscientiousness were related to a decrease in the perception of pain as threatening (extraversion: r = -.187, P < .000; agreeableness: r = −.137, P < .000; conscientiousness: r = −.144, P < .000). The dimension of openness to experience showed some significant but weaker correlations.

The presented results suggest that neuroticism can act as a vulnerability factor in the appraisal of pain, while the other dimensions, except for openness to experience, may have a protective effect. These results align with previous research. For instance, Gunthert et al[60] found that individuals with higher neuroticism scores tend to perceive stressful events as more threatening and feel less equipped to cope with them. In the context of chronic pain, Ramírez Maestre et al[61] found that a high level of neuroticism was a strong predictor of a catastrophizing interpretation of stressors. Additionally, Ebstrup et al[62] discovered a significant negative correlation between extraversion, agreeableness, and conscientiousness scores, and perceived stress, whereas the relationship between perceived stress and openness to experience was not statistically significant.

These findings are relevant to the adaptation to chronic pain, as it has been described how the propensity to value pain as a threat is related to an increase in the perception of its intensity and its interference in daily activities, and a decrease in physical and mental health.[16,63,64] The high consistency in the correlations that neuroticism maintains with stress may be due, as Suls[65] argues, to the fact that it is an affective trait (characterized by the frequent and intense experience of emotions such as fear, sadness, or anger), as opposed to the rest of the traits, which are more behavioral. In the same vein, it has been described that people with high neuroticism cope with everyday stressors with a higher basal level of negative affect, show hyperreactivity to

these events, tend to perceive them as more threatening, take longer to recover from their unpleasant emotional states, and have more difficulty solving recurring problems. These interrelated characteristics configure what is known as the "neurotic cascade."[65–67]

Regarding the relationship between personality and coping strategies, distinct patterns were found. The use of distraction as a coping strategy was more frequent in extraverted individuals (r = .153, *P* < .001) and those open to experience (r = .207, *P* < .001). On the other hand, the strategy of information seeking, which involves efforts to acquire more knowledge about pain, its causes, and ways to address it, showed a significant correlation only with extraversion (r = .122, *P* < .001). As for the strategy of mental self-control, which refers to cognitive efforts to control pain, it was more frequent in individuals with higher levels of neuroticism (r = .168, *P* < .001) and openness to experience (r = .161, *P* < .001). Additionally, this strategy negatively correlated with agreeableness (r = −.123, *P* < .005). The strategy of self-affirmation was more frequent in individuals who were more extraverted (r = .205, *P* < .001) and conscientious (r = .216, *P* < .001). On the other hand, catharsis, which involves verbalizing pain in order to seek understanding, was related to neuroticism (r = .129, *P* < .001) and agreeableness (r = .110, *P* < .005). Finally, the religious coping strategy did not show significant correlations with personality dimensions.

In a previous study conducted by Soriano et al[28] with an exclusively Spanish sample, similar correlations were found to those observed in the present study, such as the relationship between neuroticism and mental self-control, responsibility and self-affirmation, and the negative association between agreeableness and mental self-control. However, some relationships described in that study were not found in the present work, such as the association between openness to experience and distraction and self-affirmation, among others. Another study conducted by the same authors[15] evaluated the correlations between personality traits and coping strategies in a sample of 296 Spanish patients with chronic pain. In this study, similarities as well as differences were found compared to the aforementioned results. For instance, a significant correlation was found between extraversion and responsibility with the use of self-affirmation as a coping strategy. However, in this current investigation, no significant relationship was found between neuroticism and the use of mental self-control strategy.

Based on the findings and mentioned research, it can be inferred that, according to the conclusions of Soriano et al,[28] although personality appears to influence stress mediating processes, it does not establish a strong and consistent relationship to be considered a robust predictor of such processes, at least in terms of coping strategies. However, it is important to note that in the previous study conducted with the same sample,[16] coping strategies did not show significant relationships with pain interference in daily activities and quality of life, which has also been noted in other studies.[68]

Finally, in order to investigate the presence of a vulnerability profile to chronic pain, a cluster analysis was conducted using personality dimensions. As a result, 2 groups were identified: 1 characterized by high levels of neuroticism and moderate levels of extraversion, agreeableness, and conscientiousness, and another group with low levels of neuroticism and higher levels of extraversion, agreeableness, and conscientiousness. The dimension of openness to experience remained at moderate scores in both groups. Previous studies[15,28,37] have also found similar group distributions based on personality traits in patients with chronic pain. In these publications, the authors identified the group with higher scores in neuroticism as vulnerable, as they showed greater pain intensity, more symptoms (such as increased use of analgesics, poorer sleep quality, and fewer hours of sleep), lower use of coping strategies such as

self-affirmation and distraction, higher utilization of religious coping, and poorer quality of life. In the present study, the group identified as vulnerable (cluster 1) significantly experienced higher pain frequency and intensity, as well as a tendency to perceive pain as more threatening. However, no differences were found in the use of coping strategies between the groups. These characteristics may indicate greater difficulty in adapting to chronic pain for the vulnerable group compared to the resilient group.

5. Conclusion

In the present study, the 5 personality dimensions assessed in adults with OI obtained scores within moderate ranges, suggesting that personality in this population does not significantly differ from that observed in the general population. This could indicate that life events associated with the illness do not appear to have a substantial impact on personality development.

On the other hand, personality dimensions seem to play a relevant role in the experience of chronic pain, acting as protective or vulnerability factors. Neuroticism emerged as the trait with the strongest associations, being linked to higher pain frequency and intensity, as well as a perception of chronic pain as more threatening. On the other hand, although to a lesser extent, extraversion was associated with decreased pain, and together with agreeableness and conscientiousness, were related to a lower perception of threat.

Finally, the data supported the existence of a personality profile vulnerable to chronic pain, characterized by high levels of neuroticism and moderate to low levels of extraversion, agreeableness, openness to experience, and conscientiousness. Compared to the more resilient profile, this group exhibited significantly higher levels of pain frequency and intensity, as well as a more threatening appraisal of pain. These findings suggest that personality may play an important role in the adaptation to chronic pain and could be relevant in the design of personalized therapeutic interventions.

6. Limitations

This study has some limitations that should be considered. Firstly, the study design is descriptive and cross-sectional, which means that it cannot determine the direction of the observed associations or establish causal relationships. However, we have attempted to provide explanations for these associations based on relevant theoretical frameworks and findings from similar studies.

Additionally, the data collection method relied on open online surveys. While this approach allowed for a larger sample size, especially when studying conditions with low prevalence or dispersed populations, it comes with certain limitations. For example, there is less control over participants, which makes it challenging to verify the accuracy of the provided information, such as sociodemographic data. This may have led to an overrepresentation of female participants in the study. Furthermore, it is possible that only individuals with internet and social media proficiency and access completed the questionnaire, potentially introducing a selection bias.

The study's topic and the use of an open survey format for voluntary participation could introduce self-selection bias.[69,70] In other words, adults with OI who experience chronic pain more frequently may be more motivated to participate than those who do not regularly experience it. However, the data regarding pain frequency align with the findings of similar studies.[7,71]

Regarding the variables examined, it would have been interesting to consider participants' emotional responses to pain as indicators of adaptation. These aspects could provide additional

insights into the experience of chronic pain and could have enhanced the study's findings.

Acknowledgments

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References

[1] Marini JC, Cabral WA. Osteogenesis imperfecta. Genet Bone Biol Skelet Dis Second Ed. 2018:397–420.

[2] Chougui K, Addab S, Palomo T, et al. Clinical manifestations of osteogenesis imperfecta in adulthood: an integrative review of quantitative studies and case reports. Am J Med Genet Part A. 2020;182:842–65.

[3] Glorieux FH, Rowe D. Osteogenesis Imperfecta. In: Pediatric Bone. Elsevier Inc.;2012:511–39. doi:10.1016/B978-0-12-382040-2.10019-X

[4] Forlino A, Marini JC. Osteogenesis imperfecta. Lancet. 2016;387:1657–71.

[5] Van Dijk FS, Sillence DO. Osteogenesis imperfecta: clinical diagnosis, nomenclature and severity assessment. Am J Med Genet Part A. 2014;164:1470–81.

[6] Martin E, Shapiro JR. Osteogenesis imperfecta: Epidemiology and pathophysiology. Curr Osteoporos Rep. 2007;5:91–7.

[7] Arponen H, Waltimo-Sirén J, Valta H, et al. Fatigue and disturbances of sleep-in patients with osteogenesis imperfecta - a cross-sectional questionnaire study. BMC Musculoskelet Disord. 2018;19:3.

[8] Celin MR, Kruger K, Caudill A, et al. Multicenter study of pain in osteogenesis imperfecta. Arch Phys Med Rehabil. 2021;102:e2.

[9] Orlando G, Pinedo-Villanueva R, Reeves ND, et al. Physical function in UK adults with osteogenesis imperfecta: a cross-sectional analysis of the RUDY study. Osteoporos Int. 2020;32:157–64.

[10] Nghiem T, Chougui K, Michalovic A, et al. Pain experiences of adults with osteogenesis imperfecta: an integrative review. Can J Pain. 2018;2:9–20.

[11] Treede RD, Rief W, Barke A, et al. A classification of chronic pain for ICD-11. Pain. 2015;156:1003–7.

[12] Dysvik E, Natvig GK, Eikeland OJ, et al. Coping with chronic pain. Int J Nurs Stud. 2005;42:297–305.

[13] Lazarus RS, Folkman S. Stress, Appraisal, and Coping. New York: Springer Publishing Company;1984.

[14] Soriano J, Monsalve V. El afrontamiento del dolor crónico. Boletín Psicol. 2005;84:91–107.

[15] Soriano J, Monsalve V, Gómez-Carretero P, et al. Perfil de personalidad vulnerable en pacientes con dolor crónico: relación con el afrontamiento, la calidad de vida y la adaptación a la enfermedad. Int J Psychol Res. 2012;5:42–51.

[16] Muñoz R, Soriano JF, Monsalve V. Chronic pain in adults with osteogenesis imperfecta and its relationship to appraisal, coping, and quality of life: a cross-sectional study. Medicine (Baltim). 2022;101:e30256.

[17] Amirkhan JH, Risinger RT, Swickert RJ. Extraversion: a "hidden" personality factor in coping? J Pers. 1995;63:189–212.

[18] David JP, Suls J. Coping efforts in daily life: role of big five traits and problem appraisals. J Pers. 1999;67:265–94.

[19] Hooker K, Frazier LD, Monahan DJ. Personality and coping among caregivers of spouses with dementia. Gerontologist. 1994;34:386–92.

[20] Rim Y. Ways of coping, personality, age, sex, and family structural variables. Pers Individ Dif. 1986;7:113–6.

[21] Watson D, Hubbard B. Adaptational style and dispositional structure: coping in the context of the five-factor model. J Pers. 1996;64:737–74.

[22] Costa PT, McCrae RR. The NEO Personality Inventory Manual. Odessa, FL: Psychological Assessment Resources; 1985.

[23] Sánchez RO. Los Cinco Grandes Factores: ¿cómo entender la personalidad y cómo evaluarla? In: Conocimiento Para La Transformación. Serie Investigación y Desarrollo. Universidad Atlántida Argentina;2007:131–60. https://www.researchgate.net/publication/338111357_Los_Cinco_Grandes_Factores_como_entender_la_personalidad_y_como_evaluarla. [accessed May 6, 2021].

[24] Costa PT, McCrae RR. Inventario de Personalidad Neo Revisado (NEO PI-R). Manual Profesional. 2nd ed. Madrid: TEA Ediciones; 2002.

[25] Bucourt E, Martaillé V, Mulleman D, et al. Comparison of the big five personality traits in fibromyalgia and other rheumatic diseases. Jt Bone Spine. 2017;84:203–7.

[26] Ibrahim ME, Weber K, Courvoisier DS, et al. Big five personality traits and disabling chronic low back pain: association with fear-avoidance, anxious and depressive moods. J Pain Res. 2020;13:745–54.

[27] Kadimpati S, Zale EL, Hooten WM, et al. Erratum: associations between neuroticism and depression in relation to catastrophizing and pain-related anxiety in chronic pain patients (PLoS One (2015) 10:4 (e0126351) (doi:10.1371/journal.pone.0126351)). PLoS One. 2015;10:e0129871.

[28] Soriano J, Monsalve V, Ibañez E, et al. Personalidad y afrontamiento en dolor crónico neuropático: un divorcio previsible. 2010.

[29] Phillips JM, Gatchel RJ. Extraversion–introversion and chronic pain. In: Personality Characteristics of Patients with Pain. American Psychological Association; 2004:181–202. doi:10.1037/10376-008

[30] Gómez Carretero P, Soriano J, Monsalve V, et al. Personalidad y afrontamiento como predictores de la resiliencia en pacientes con dolor crónico. In: Conferencia.; 2011. https://psiquiatria.com/bibliopsiquis/volumen.php?wurl=personalidad-y-afrontamiento-como-predictores-de-la-resiliencia-en-pacientes-con-dolor-cronico. [accessed June 17, 2021].

[31] Magyar M, Gonda X, Pap D, et al. Decreased openness to experience is associated with migraine-type headaches in subjects with lifetime depression. Front Neurol. 2017;8:270.

[32] Williams PG, Rau HK, Cribbet MR, et al. Openness to experience and stress regulation. J Res Pers. 2009;43:777–84.

[33] Newth S, Delongis A. Individual differences, mood, and coping with chronic pain in rheumatoid arthritis: a daily process analysis. Psychol Heal. 2004;19:283–305.

[34] Krok JL, Baker TA. The influence of personality on reported pain and self-efficacy for pain management in older cancer patients. J Health Psychol. 2014;19:1261–70.

[35] Sutin AR, Stephan Y, Luchetti M, et al. The prospective association between personality traits and persistent pain and opioid medication use. J Psychosom Res. 2019;123:109721.

[36] Asghari MA, Nicholas MK. Personality and adjustment to chronic pain. Pain Rev. 1999;6:85–97.

[37] Soriano J, Monsalve V. Profiles of personality and resilience in chronic pain: utility of the CD-RISC-10 to discriminate between resilient and vulnerable types. Rev la Soc Española del Dolor. 2019;26:72–80.

[38] Freynhagen R, Baron R, Gockel U, et al. painDETECT: a new screening questionnaire to identify neuropathic components in patients with back pain. Curr Med Res Opin. 2006;22:1911–20.

[39] De Andrés J, Pérez-Cajaraville J, Lopez-Alarcón MD, et al. Cultural adaptation and validation of the pain detect scale into Spanish. Clin J Pain. ;28:243–53.

[40] Costa PT, McCrae RR. Normal personality assessment in clinical practice: the NEO personality inventory. Psychol Assess. 1992;4:5–13.

[41] Digman JM. Personality structure: emergence of the five-factor model. Annu Rev Psychol. 1990;41:417–40.

[42] McCrae RR, Costa PT. Validation of the five-factor model of personality across instruments and observers. J Pers Soc Psychol. 1987;52:81–90.

[43] Cordero A, Pamos A, Seisdedos N. RESUMEN DE LA VALORACIÓN DEL TEST Descripción General Características Descripción Nombre Del Test NEO PI-R, Revised Neo Personality Inventory Autor de La Adaptación Española.

[44] Manga D, Ramos F, Morán C. The Spanish norms of the NEO five-factor inventory: new data and analyses for its improvement | IJPSY. 2004:639–48. https://www.ijpsy.com/volumen4/num3/104.html. [accessed November 28, 2019].

[45] Unruh AM, Ritchie JA. Development of the pain appraisal inventory: psychometric properties. Pain Res Manag. 1998;3:105–10.

[46] Soriano J, Monsalve V. Validación del cuestionario de afrontamiento al dolor crónico reducido (CAD-R). 2004:407–14. http://scielo.isciii.es/scielo.php?script=sci_arttext&pid=S1134-80462004000700002. [accessed December 1, 2019].

[47] Balkefors V, Mattsson E, Pernow Y, et al. Functioning and quality of life in adults with mild-to-moderate osteogenesis imperfecta. Physiother Res Int. 2013;18:203–11.

[48] Català E, Reig E, Artés M, et al. Prevalence of pain in the Spanish population: telephone survey in 5000 homes. Eur J Pain. 2002;6:133–40.

[49] Dahlhamer J, Lucas J, Zelaya, C, et al. Prevalence of chronic pain and high-impact chronic pain among adults – United States, 2016. MMWR Morb Mortal Wkly Rep. 2018;67:1001–6.

[50] Public Health England. Chronic Pain in Adults 2017 - An Analysis of Self-Reported Chronic Pain from 2017. Health Survey for England Data. 2020. https://www.gov.uk/government/publications/chronic-pain-in-adults-2017. [accessed March 22, 2023].

[51] Costa PT, Robert McCrae JR. Inventario de personalidad neo revisado inventario neo reducido de cinco factores (neo-ffi) manual profesional. 1978.

[52] Briley DA, Tucker-Drob EM. Genetic and environmental continuity in personality development: a meta-analysis. Psychol Bull. 2014;140:1303–31.

[53] Krueger RF, South S, Johnson W, et al. The heritability of personality is not always 50%: gene-environment interactions and correlations between personality and parenting. J Pers. 2008;76:1485–522.

[54] Vukasović T, Bratko D. Heritability of personality: a meta-analysis of behavior genetic studies. Psychol Bull. 2015;141:769–85.

[55] Sutin AR, Zonderman AB, Ferrucci L, et al. Personality traits and chronic disease: implications for adult personality development. J Gerontol Ser B Psychol Sci Soc Sci. 2013;68:912–20.

[56] Jokela M, Hakulinen C, Singh-Manoux A, et al. Personality change associated with chronic diseases: pooled analysis of four prospective cohort studies. Psychol Med. 2014;44:2629–40.

[57] Deary IJ, Weiss A, Batty GD. Intelligence and personality as predictors of illness and death: how researchers in differential psychology and chronic disease epidemiology are collaborating to understand and address health inequalities. Psychol Sci Public Interest. 2010;11:53–79.

[58] Ironson GH, O'Cleirigh C, Weiss A, et al. Personality and HIV disease progression: role of NEO-PI-R openness, extraversion, and profiles of engagement. Psychosom Med. 2008;70:245–53.

[59] Turiano NA, Whiteman SD, Hampson SE, et al. Personality and substance use in midlife: conscientiousness as a moderator and the effects of trait change. J Res Pers. 2012;46:295–305.

[60] Gunthert KC, Cohen LH, Armeli S. The role of neuroticism in daily stress and coping. J Pers Soc Psychol. 1999;77:1087–100.

[61] Ramírez Maestre C, Esteve Zarazaga R, López Martínez AE. Neuroticismo, Afrontamiento Y Dolor Crónico | Anales de Psicología/Annals of Psychology. An Psicol. 2001;17:129–37.

[62] Ebstrup JF, Eplov LF, Pisinger C, et al. Association between the five factor personality traits and perceived stress: is the effect mediated by general self-efficacy? Anxiety, Stress Coping. 2011;24:407–19.

[63] Gatchel RJ, Peng YB, Peters ML, et al. The biopsychosocial approach to chronic pain: scientific advances and future directions. Psychol Bull. 2007;133:581–624.

[64] Soriano J. Valoración, afrontamiento y emoción en pacientes con dolor crónico - Dialnet. Boletín Psicol. 1999;62:43–64.

[65] Suls J. Affect, stress, and personality. In: Forgas JP, ed. Handbook of Affect and Social Cognition. Mahwah, NJ: Lawrence Erlbaum Associates Publishers; 2001:392–409. https://psycnet.apa.org/record/2000-16445-018. [accessed November 30, 2020].

[66] Semmer NK. Personality, stress, and coping. In: Handbook of Personality and Health. Chichester, UK: John Wiley & Sons Ltd; 2008:73–113. doi:10.1002/9780470713860.ch4

[67] Suls J, Martin R. The daily life of the garden-variety neurotic: Reactivity, stressor exposure, mood spillover, and maladaptive coping. J Pers. 2005;73:1485–509.

[68] Soucase B, Monsalve V, Soriano J, et al. Estrategias de afrontamiento ante el dolor y calidad de vida en pacientes diagnosticados de fibromialgia. Rev la Soc Española del Dolor. 2004;11:4551.

[69] Elston DM. Participation bias, self-selection bias, and response bias. J Am Acad Dermatol. 2021;0.

[70] García Villar C. Introducción a la lectura crítica de artículos: diseño de estudios y sesgos. Radiologia. 2015;57:3–13.

[71] Barlow S, Dove L, Jaggi A, et al. The prevalence of musculoskeletal pain and therapy needs in adults with Osteogenesis Imperfecta (OI) a cross-sectional analysis. BMC Musculoskelet Disord. 2022;23.