

## ORIGINAL ARTICLE

## Musculoskeletal

# The relationship between chronic pain and psychosocial aspects in patients with haemophilic arthropathy. A cross-sectional study

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## Abstract

**Background:** Pain is a major characteristic in haemophilic arthropathy. Identifying the psychosocial variables affected by pain can help when addressing these patients.

**Aim:** To assess the relationship between perceived intensity of chronic pain and joint damage, kinesiophobia, catastrophism, anxiety and perceived quality of life in adult patients with haemophilic arthropathy.

**Methods:** Multicentre cross-sectional descriptive study. Seventy-seven adult patients with haemophilic arthropathy were recruited. The usual and maximum pain intensity (Visual Analog Scale), joint status (Haemophilia Joint Health Score), Kinesiophobia (Tampa Scale of Kinesiophobia), catastrophism (Pain catastrophizing scale), anxiety (State-Trait Anxiety inventory) and perceived quality of life (36-Item Short Form Health Survey) were evaluated. The correlation between usual and maximum pain intensity with quantitative variables was obtained with Spearman's correlation test. Kruskal-Wallis one-way ANOVA analysed differences in perceived pain according to the severity and type of treatment, and development of inhibitors.

**Results:** The usual intensity of perceived pain correlated positively with catastrophism, kinesiophobia, and state and trait anxiety. The same results were obtained when analysing the maximum perception of pain. We found an inverse correlation between the physical component of perceived quality of life and usual and maximum pain intensity perceived by patients.

**Conclusion:** Psychosocial factors affect the painful experience of patients with haemophilic arthropathy. Pain intensity affects the quality of life of these patients.

## KEYWORDS

anxiety, catastrophizing, haemophilia, joint pain, kinesiophobia, quality of life

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## 1 | BACKGROUND

Haemophilia is a congenital coagulopathy characterized by a deficiency of a clotting factor. The main clinical manifestations are haemorrhages. Most bleeding events occur in the musculoskeletal system, mainly in the joints (hemarthrosis).<sup>1</sup> Hemarthrosis entails extremely painful bleeding events, presenting with swelling, hotness, reflex muscle inhibition and functional deficit.<sup>2</sup>

Recurrence of joint bleeding leads to changes in the synovial membrane (chronic synovitis) and joint structure, which cause intra-articular alterations and functional limitations.<sup>3</sup> Hemarthrosis eventually leads to inactivity and, consequently, atrophy of the periarticular muscles and joint instability, which in turn significantly increases the risk of further bleeding.<sup>4</sup> This process progressively contributes to irreversible joint degeneration and the development of chronic haemophilic arthropathy, characterized by joint deformity, functional limitations, disability and chronic pain.<sup>5</sup> Prophylactic administration of clotting factor concentrates is the *gold standard* in the treatment of patients with haemophilia. This is the most effective treatment in preventing life-threatening bleeding events and disability resulting from recurrent hemarthrosis.<sup>6</sup>

Pain is an unpleasant sensory and emotional experience associated with or similar to that associated with actual or potential tissue damage.<sup>7</sup> Similarly, pain is always a personal experience, to an extent influenced by biological, psychological and social factors.<sup>7</sup> Some factors such as stress or anxiety can influence the experience of pain.<sup>8</sup> Pain experience and nociception are different phenomena. Pain experience not only involve sensory neurons.<sup>7</sup> The presence of endogenous pain modulating systems has been proposed and their approach may facilitate the development of more effective interventions in pain management.<sup>8</sup>

Pain is one of the main features of haemophilic arthropathy. An incidence of localized pain in more than one joint has been described<sup>9</sup> in more than 60% of patients with haemophilia. A multicentre study raised this figure to 89% of patients who reported pain that interfered with the performance of their daily activities.<sup>10</sup> Witkop et al.<sup>11</sup> reported that 20% of patients with haemophilia experience acute pain, 34% chronic pain, and 32% both types of pain.

In the absence of standardized data and guidelines, the pharmacological management of pain in patients with haemophilia in clinical practice is based on empirical experience.<sup>12</sup> The administration of painkillers is a therapeutic option in 36% of patients with hemophilia.<sup>9</sup>

Although pain has an adaptive function, it can produce adverse effects on both function and physical and social well-being.<sup>7</sup> Therefore, patients with pain are more likely to develop depression, obesity and a poorer quality of life. Several studies<sup>10,13</sup> have pointed to the relationship between chronic pain and poor psychological functioning and low quality of life related to physical and mental health in patients with haemophilia.

The fact of suffering from a chronic disease, alone, does not increase the risk of depression or anxiety. However, pain associated with chronic illness increases the risk of developing such disorders. Suboptimal coping strategies often affect relationships and reduce motivation to

engage in activities that increase self-esteem.<sup>14</sup> Pain, depression and anxiety have complex, two-way interrelationships. Although depression is underdiagnosed in patients with haemophilia, it is associated with anxiety, pain and decreased adherence to treatment.<sup>15</sup> Identifying the patient's behavioural, sociological and psychosocial aspects is important for establishing the best strategy for pain management in patients.<sup>16</sup>

The aim of this study is to evaluate the relationship between the perceived intensity of chronic, usual and maximum pain, and joint damage, kinesophobia, catastrophism, anxiety and the perception of quality of life in adult patients with haemophilic arthropathy.

## 2 | METHODS

### 2.1 | Design and setting

Multicentre cross-sectional descriptive study conducted in adult patients with haemophilia from all over Spain. Patients were recruited prospectively between March 2018 and October 2019.

The study was conducted in accordance with the Declaration of Helsinki. Written informed consent was obtained from all patients included in the study. Ethics approval was granted by the Clinical Research Ethics Committee of the Virgen de la Arrixaca University Hospital of Murcia (id. 2020-2-9-HCUVA). Prior to patient recruitment, the study protocol was registered with the international registry of clinical trials [www.clinicaltrials.gov](http://www.clinicaltrials.gov) (id. NCT03499522).

### 2.2 | Participants

Inclusion criteria for taking part in the study: patients with a medical diagnosis of haemophilia A or B; diagnosed with haemophilic arthropathy (at least three points on the *Haemophilia Joint Health Score*)<sup>17</sup>; aged over 18 years; and not having undergone orthopaedic surgery or radioactive synovectomy in the 6 months prior to the study. Exclusion criteria of the study: patients who failed to complete all the evaluation instruments; patients with difficulty in understanding and writing the psychosocial evaluation questionnaires themselves; patients who had had hemarthrosis in the 12 weeks prior to the study; patients with walking difficulties; and patients who failed to sign the Informed Consent Document.

### 2.3 | Procedure

Patients interested in participating in the study and who met the selection criteria, contacted the researchers through their respective Association. Patients were recruited from eight regions of Spain (Andalusia, Aragon, Castilla y León, Galicia, Madrid, Basque Country, Murcia region and Valencia Community). The participants were informed through a telematic survey. Participation was voluntary. All patients who showed their interest in participating in the study were informed by the

evaluator and after verifying that they met the selection criteria, they signed the informed consent. According to the census of the Spanish Federation of Haemophilia, the number of patients with haemophilia who met the selection criteria to participate in the study was 450 ( $n = 450$ ). Eventually, the data of 77 respondents were used for the final analysis.

Through personal interviews, data on the psychosocial variables of the patients were collected. A physiotherapist, experienced in the treatment and evaluation of patients with haemophilia, performed the physical evaluation of patients to measure joint health and pain intensity.

## 2.4 | Assessment

The variables evaluated in the study were pain intensity (usual and maximum), joint health, kinesiophobia, catastrophism, anxiety and perception of quality of life.

The perceived pain intensity of patients with haemophilia was evaluated using the visual analogue scale. This scale has shown good reliability and validity for the assessment of pain intensity.<sup>18</sup> This measuring instrument evaluates the intensity of pain on a 10 cm line (0 being “no pain” and 10 being “the worst pain”). Patients were asked to make a mark on the line to represent the average intensity of the usual joint pain they endure in the last 7 days. Similarly, they were asked to mark the maximum degree of pain they report in the last week.

The joint condition of knees, ankles and elbows was evaluated using version 2.1. of the *Haemophilia Joint Health Score*.<sup>17</sup> This scale, specific for the evaluation of joint damage in patients with haemophilia, evaluates eight items (*swelling, duration of swelling, atrophy, crepitus, range of motion, loss of flexion and extension, muscle strength and pain*). This measuring instrument scores 0–20 points for the state of each joint. In addition, gait is scored on a range of 0–4 points. This scale has shown excellent inter and intraobserver reliability ( $ICC = .90$  and  $ICC = .91$ , respectively).<sup>19</sup>

Kinesiophobia, or fear of movement or physical activity, was measured with the Spanish version<sup>20</sup> of the *Tampa Scale of Kinesiophobia* (TSK-11).<sup>21</sup> This self-administered measuring instrument consists of 11 items with a 4-point Likert scale. The score range is 11–44 (a higher score indicates a greater fear of re-injury from movement). This test has shown good test-retest reliability (intraclass correlation coefficient [ $ICC$ ] = .72).<sup>22</sup>

Catastrophism was evaluated with the validated Spanish version of the *Pain catastrophizing scale* measurement instrument.<sup>23</sup> This scale, composed of 13 items, uses a 5-point Likert scale (from 0 “mild symptoms” to 4 “worst symptoms”). The total score is derived from the sum of the individual scores of the 13 items. Scores range from 0 to 52 points (the higher the score the greater is the number of catastrophic characteristics of pain in the patient). Ong et al.<sup>24</sup> reported a good internal consistency of this scale (Cronbach's  $\alpha = .94$ ) in patients with knee osteoarthritis.

Anxiety was evaluated with the validated Spanish version of the *State-Trait Anxiety inventory* (STAI) questionnaire.<sup>25</sup> This measuring

instrument has two self-assessment scales to measure two independent anxiety concepts: state and trait. Both scales have 20 items each, which are scored on a Likert scale having four possible replies (from 0 to 3). This questionnaire presents a good internal consistency in the Spanish version in anxiety/state (Cronbach's  $\alpha = .90 - .93$ ) and in anxiety/trait (Cronbach's  $\alpha = .84 - .87$ ).<sup>25</sup>

The perceived quality of life was measured with the Spanish version<sup>26</sup> of the *36-Item Short Form Health Survey* (SF-36) scale, version 2. This self-administered generic scale, providing a health profile can be applied to patients and the general population to assess health-related quality of life. It consists of 36 items divided into eight domains: physical functioning, limitations due to physical problems, physical pain, social role or function, mental health, limitation due to emotional problems, vitality and general perception of health. This scale has shown good internal consistency (Cronbach's  $\alpha > .85$ ; reliability coefficient  $> .75$ ).<sup>27</sup> A higher score indicates a better perception of quality of life.

Similarly, the main clinical and therapeutic variables of the patients recruited in the study were collected.

## 2.5 | Statistical analysis

Data analysis was carried out with the statistical package SPSS Statistics version 25.0 (IBM, Inc., Armonk, NY, USA). The main descriptive statistics of central trend (mean) and dispersion (standard deviation) of the quantitative variables were calculated. Frequencies and proportions of qualitative variables were obtained. The statistical analysis should be performed using non-parametric tests. The correlation between usual and maximum pain intensity with quantitative dependent variables was performed with Spearman correlation test. Kruskal-Wallis one-way ANOVA was used to analyse differences in pain perception depending of the severity of haemophilia, development of inhibitors, type of treatment, and history of knee arthroplasty and radio-synovectomy in the last 5 years. All results are considered significant for a level  $< .05$ .

## 3 | RESULTS

Ultimately, 77 patients with haemophilia were included in the study. The mean age of the sample was 44.04 (SD: 10.17) years. Most patients had a diagnosis of haemophilia A (83.1%), received prophylactic treatment (71.4%) and a severe phenotype of the disease (87.0%). None of the patients performed regular physical activity or participated in directed activity courses (for example gym). Table 1 shows the clinical and therapeutic characteristics of the patients included in the study.

The usual perception of pain in patients with haemophilia correlated positively ( $P < .001$ ) with catastrophism ( $r = .68$ ), kinesiophobia ( $r = .67$ ), state anxiety ( $r = .58$ ) and trait anxiety ( $r = .39$ ). When analysing the maximum pain perception, we found a correlation ( $P < .001$ ) for all variables: catastrophism ( $r = .49$ ), kinesiophobia ( $r = .52$ ), state anxiety ( $r = .49$ ) and trait anxiety ( $r = .39$ ). When relating the intensity of pain and the perceived quality of life, we found

**TABLE 1** Clinical characteristics of the patients included in this study and descriptive statistics of dependent variables

Variables	n	%
Type of haemophilia		
Haemophilia A	64	83.1
Haemophilia B	13	16.9
Severity of haemophilia		
Severe	67	87.0
Moderate	10	13.0
Inhibitor		
No	65	84.4
Yes	12	15.6
Treatment		
On demand	22	28.6
Prophylaxis	55	71.4
Knee arthroplasty		
No	66	85.7
Yes	11	14.3
Radio-synovectomy		
No	36	53.2
Yes	41	46.8
	Mean	SD
Age (years)	44.04	10.17
Knee joint status (range: 0–40)	18.01	7.21
Right knee (0–20)	9.25	3.94
Left knee (0–20)	8.79	3.75
Ankle joint status (range: 0–40)	20.53	5.48
Right ankle (0–20)	10.49	2.85
Left ankle (0–20)	10.17	3.17
Elbow joint status (range: 0–40)	15.81	5.58
Right elbow (0–20)	8.29	2.96
Left elbow (0–20)	7.52	3.14
All joints status (range: 0–124)	56.17	16.47
Intensity of current joint pain (range: 0–10)	4.52	1.65
Maximal intensity of joint pain (range: 0–10)	7.99	1.37
Catastrophism	10.49	8.81
Kinesiophobia (range: 11–44)	30.53	7.17
State anxiety (range: 0–60)	17.48	5.49
Trait anxiety (range: 0–60)	18.44	6.86
Physical component summary	30.35	8.77
Mental component summary	56.05	5.80

Abbreviations: n, number of patients; %, percentage; SD, standard deviation; NSAID, Non-Steroidal Anti-inflammatory drugs.

an inverse correlation ( $P < .001$ ) between the total physical health score and usual pain ( $r = -.64$ ) and maximum pain ( $r = -.66$ ) perceived by patients. Table 2 shows the correlation analysis of pain intensity with all other dependent variables.

**TABLE 2** Correlations between joint pain and joint damage, and catastrophism, kinesiophobia, anxiety and quality of life

Variables	Intensity of current joint pain	Maximal intensity of joint pain
Knee joint status (0–20)	.04	.07
Ankle joint status (0–20)	-.12	-.06
Elbow joint status (0–20)	.16	.04
All joints status (0–124)	.05	.03
Age	-.24*	-.12
Catastrophism	.68**	.54**
Kinesiophobia	.67**	.49**
State anxiety	.56**	.47**
Trait anxiety	.40**	.36**
Physical component summary	-.64**	-.64**
Mental component summary	-.16	-.01

\*Significant difference ( $P < .05$ ).

\*\*Significant difference ( $P < .01$ ).

When comparing the perception of pain based on the severity of haemophilia, patients with severe haemophilia showed a greater perception of pain ( $P = .003$ ) than patients with moderate haemophilia, with respect to usual pain (4.73 vs 3.10) and maximum pain (8.16 vs 6.80). The development of antibodies to treatment with clotting factor concentrates showed differences ( $P = .01$ ) regarding the usual intensity of pain in patients with haemophilia (5.67 vs 4.31). In terms of the type of treatment, we found no differences ( $P > .05$ ) in the intensity of pain among patients with prophylactic or on-demand treatment. Finally, we found differences regarding the history of knee arthroplasty ( $P < .05$ ) and radio-synovectomy ( $P < .001$ ) in the usual intensity of pain and maximum pain. Table 3 shows the results of the analysis of independent samples between perceived pain and the clinical variables of the patients included in the study.

## 4 | DISCUSSION

The objective of this study was to evaluate the perceived pain intensity, both usual and maximum, in adult patients with haemophilic arthropathy and the physical and psychosocial variables most related to it. Catastrophism, kinesiophobia, perception of quality of life, state anxiety and trait anxiety correlated with perception of both usual and maximum pain intensity. Patients with increased pain intensity presented higher rates of catastrophism, kinesiophobia, state anxiety and trait anxiety. Similarly, they presented a poorer perceived quality of life on the physical health scale. In addition, severity of haemophilia was also related with both degrees of perceived pain intensity, and the existence of inhibitors with usual pain. Those with a more severe haemophilia presented a greater intensity of pain. On the other hand, usual pain was increased in those who developed antibodies to the treatment.

**TABLE 3** Relationship between intensity of joint pain and severity of haemophilia, inhibitor development, and type of treatment

Variables	Severity of haemophilia		Inhibitor		Type of treatment		Knee arthroplasty		Radio-synovectomy	
	MD	95%CI	MD	95%CI	MD	95%CI	MD	95%CI	MD	95%CI
Intensity of current joint pain	-1.63*	-2.68; -.57	-1.35*	-2.35; -.36	.53	-.29; 1.36	1.24*	.20; 2.28	1.68**	1.03; 2.33
Maximal intensity of joint pain	-1.36*	-2.24; -.48	-.80*	-1.64; .03	.04	-.64; .73	1.15*	.29; 2.00	1.95**	1.51; 2.39

\*Significant difference ( $P < .05$ ).

\*\*Significant difference ( $P < .001$ ).

Abbreviations: MD, mean difference; 95% CI, 95% confidence interval.

Usual pain is a common condition in patients with haemophilic arthropathy.<sup>28</sup> Patients included in this study showed moderate values of usual pain,<sup>29</sup> with moderate-severe peaks of maximum pain.<sup>29</sup> These results are consistent with those reported by Pinto et al.<sup>13</sup> where maximum pain intensities were between moderate and severe<sup>30</sup> (mean: 5.67; SD = 2.09).

Few studies have evaluated catastrophism in patients with haemophilia. Santavirta et al.<sup>31</sup> pointed to catastrophism as one of the coping strategies developed in these patients. Normative values of catastrophism for male patients with chronic pain are established (mean: 30.9; SD = 13.6).<sup>32</sup> Patients with haemophilia included in this study had lower values in the lower limbs and upper limbs. Such low values of catastrophism may be due to the pain experience in these patients. Haemophilia is a congenital disease in which patients endure joint bleeding and severe pain from a very early age. This experience with pain may explain why the cognitive-emotional response is not so high or negative. This hypothesis would be consistent with the explanations given by the *International Association for the Study of Pain*,<sup>7</sup> whereby the concept of pain is learned through our life experiences.

The patients in our study presented a high mean value of kinesiophobia, in contrast to the values reported by Calatayud et al.<sup>33</sup> in adult patients with haemophilic elbow arthropathy. Values over 27 points on the measuring instrument used are considered high scores.<sup>34</sup> However, if these results are compared against the normative values described<sup>35</sup> for populations with chronic musculoskeletal pain, it would only be higher than the values described by patients with osteoarthritis. The correlation of kinesiophobia values with pain intensity observed in our results are in line with an association of a high degree of kinesiophobia with high levels of pain intensity.<sup>36</sup> The high degree of kinesiophobia can be related to the fear of developing new hemarthrosis that further deteriorate the joint condition, being an important nociceptive source. This experience of previous trauma and its haemorrhagic consequences may justify the high degree of kinesiophobia in adult patients with haemophilia.

The values for both state and trait anxiety observed in the study patients correspond to a 45 percentile,<sup>25</sup> slightly lower than the average for the Spanish population. These values in patients with haemophilia are considerably lower than those referred in patients with other pathologies that are undergoing chronic pain due to joint degeneration or disc pathology.<sup>37</sup> Anxiety is an emotional state pro-

jected towards the future, in which the source of the threat is anticipated without having yet been detected, provoking a stimulation of the prevention response.<sup>38</sup> Patients with haemophilia suffer from hemarthrosis and pain from an early age, so uncertainty and anticipation of the stimulus are mitigated by the patient's own self-knowledge. On the other hand, anxiety is associated with defensive behaviours, such as hypervigilance, catastrophic thoughts, and avoidance behaviours.<sup>38</sup> Therefore, these low values of anxiety would be in accordance with the values noted for catastrophism.

High kinesiophobia values are associated with a poorer quality of life.<sup>36</sup> Our results are in line with previous studies in hemophilia<sup>10</sup> where a greater intensity of pain is associated with a poorer perceived quality of life. This relationship may be due to how pain affects the development of activities and, thus the more physical components are perceived.

These results concerning intensity, catastrophism, kinesiophobia and quality of life are consistent with what was observed in other populations with chronic musculoskeletal pain.<sup>36,39</sup> High levels of catastrophism and kinesiophobia are associated with higher pain intensities and a more deteriorated perception of quality of life, suggesting the importance of psychosocial factors in the perception of pain and quality of life in patients with haemophilia and chronic pain.

Patients exhibited a correlation between the severity of haemophilia and the intensity of their usual and maximum pain. This relationship is logical in patients suffering severe degenerative joint damage and recurrent hemarthrosis over many years. However, the existence of antibodies against clotting factor concentrates (inhibitors) only showed an association with the usual pain. This may be due to a history of severe hemarthrosis without effective haemostatic treatment, which has evolved into prophylactic treatments with new drugs that allow for controlled and effective haemostasis.

Finally, we found a relationship with respect to the intensity of their usual and maximum pain in those patients who had undergone total knee arthroplasty or previous radio-synovectomy. These results are consistent with previous studies<sup>40,41</sup> that observed a reduction in pain perception in haemophilia patients who underwent these surgeries. These improvements in pain perception may be due to the reduction of nociceptive inputs because of both interventions. The joint replacement component of arthroplasty eliminates friction between bone surfaces that favours pain relief. In radio-synovectomy, the reduction



of synovial tissue and, therefore, the decrease in the frequency of hemarthrosis and inflammation justifies this improvement in patients with haemophilia.

#### 4.1 | Limitations of the study

This study has several limitations. The main limitation is sample size and sample recruitment. The measurement of the level of physical activity practice could have offered more information on the perception of joint pain in patients with haemophilic arthropathy. A study design including such level of physical activity practice (e.g., with IPAQ questionnaire) is an option to assess the relation of this variable with joint pain perception in these patients.

The use of different measuring instruments to measure the same variables (quality of life, intensity of pain, etc.) makes it difficult to compare results with findings of other studies. Unified assessment tools would promote the reporting of results and the generalization of conclusions. The values of daily pain and maximum pain in the same body region were collected. The absence of a joint-by-joint analysis and its relationship with the psychosocial variables included in the study is another limitation of the study. Finally, the assessment of analgesic alternatives was an important limitation of the study. The administration of diverse and varied analgesic drugs, the different pharmacological treatment guidelines, self-medication and the variability in consumption make it difficult to specifically analyse this variable on pain control in patients with haemophilia.

#### 4.2 | Relevance to clinical practice

The literature that addresses psychosocial factors such as catastrophism or kinesiophobia in patients with haemophilia is scarce. The absence of studies limits an understanding of these factors and possible approaches, and the evaluation of these variables needs to be addressed in future studies on pain in these patients.

The results of this study can guide us in the understanding of psychosocial factors that are related to pain in patients with haemophilia. As in other populations with chronic pain, these variables need to be addressed to improve the quality of life and functionality of patients. Establishing their existence enables us to approach their management. Kinesiophobia, catastrophism and anxiety are determining factors in the painful experience of patients with chronic pain and approaching them can be essential in the treatment of patients with haemophilia.

### 5 | CONCLUSIONS

Patients with haemophilia exhibit a high prevalence of chronic pain. The intensity of such pain is moderate, yet reaching severe maximum peaks.

The intensity of pain in patients with haemophilic arthropathy is correlated with anxiety, catastrophism, and the degree of kinesiophobia.

The perception of quality of life in patients with haemophilia, especially the physical component, negatively correlates with the intensity of pain.

Psychosocial factors influence the painful experience of patients with haemophilic arthropathy, affecting their perception of quality of life.

#### CONFLICTS OF INTEREST

The authors have no competing interests.

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#### DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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