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Original article

Body perception distortions correlate with neuropathic features in Italian fibromyalgic patients: Findings from a self-administered online survey

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ABSTRACT

Background: Recent studies found that fibromyalgia may underly neuropathic conditions affecting the peripheral nervous system. Moreover, clinical observations and preliminary reports suggest the existence of body perceptions distortions (BPD) like "phantom" feelings of swollen hands and feet, similar to those complained by patients with other neuropathic conditions or subjected to experimental procedures affecting the peripheral nervous system.
Objectives: To investigate the prevalence of self-reported BPD in Italian people with fibromyalgia through an online survey administered with the help of the associations of patients distributed nationwide.
Design: cross-sectional study.
Method: A nationwide sample of 854 patients out of 1173 subjects enrolled was analyzed after the exclusion of comorbidities and incomplete answers. We additionally performed a post-hoc analysis comparing data of patients who entirely fulfilled the Fibromyalgia Research Criteria (FRC) (2011) for epidemiological studies with respect to those only partially responding to the FRC (FM-).
Results: Nearly 90% of subjects reported neuropathic pain, symptoms potentially indicative for small-fiber pathology, and at least 1 BPD, while 2 or more BPD was reported in 64.1% of cases. Phantom feelings of "heaviness", "constriction", and "swelling" were the most frequently self-reported perceptual distortions. BPD were significant correlated with symptoms potentially indicative for small-fiber pathology, neuropathic pain, disability, painful sites, and severity of fibromyalgia ($0.20<\tau\text{-b}<0.33$).
Conclusions: Our preliminary findings highlighted that the phenomenon of self-reported BPD in patients with fibromyalgia correlated with neuropathic symptoms. If these results will be confirmed in future studies BPD may be potentially considered as part of the clinical picture of fibromyalgia.

1. Introduction

Fibromyalgia (FM) severely affects the quality of life (Wolfe et al., 1990), especially for its psychological consequences, however, the variegated clinical picture of this syndrome sometimes has led clinicians to doubt its real existence to the extent of being considered a reflection of patients' anxiety or attention-seeking behaviour (McCabe et al., 2007). Despite this, recent studies have found that FM may underly a neuropathic condition affecting mostly the thin Aδ and C fibers, the so-called 'small fibers' (Grayston et al., 2019), especially in more severe phenotypes (Evdokimov et al., 2019). These findings have offered an additional hypothesis about the etiopathogenesis of this syndrome that seems to overcome the label of 'central sensitization syndrome' assigned to it until now (Cheng et al., 2018).

Noteworthy, clinical observations and preliminary reports suggest that, in addition to the classical symptoms, patients with FM report also body perception distortions (BPD) like "phantom" feelings of swollen hands and feet (*macrosomatoagnosia)* (Moseley, 2005), similarly to those complained by patients with Complex Regional Pain Syndrome (CRPS) (Lewis et al., 2007; Peltz et al., 2011) or musculoskeletal disorders (Viceconti et al., 2020; Tanaka et al., 2021). These symptoms have been defined as "*phantom*" because the perceived feelings can't be confirmed through the visual/manual inspection of the affected body parts. Patients often are aware of the perceptual conflict existing between what

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they feel and what their body parts look like, recognizing that no signs of swelling are obvious (McCabe et al., 2004). However, BPD seem so real that, for example, patients try to untie their shoes or they wear larger clothes to feel more comfortable with their "oversized" body parts (McCabe et al., 2009).

Pathological or experimentally-induced dysfunctions occurring in the peripheral nervous system may cause BPD (Pazzaglia et al., 2019; Vaso et al., 2014) as illusory perceptual phenomena of swelling, elongation, or shortening of the affected arms (Russell and Tsao, 2018). Interestingly, Paqueron et al. (2003) found that perceptual illusions coincided with the (partial or complete) loss of small-fibers functioning. Considering that body perception is generated from a sensory continuous flux of information from the periphery to the central nervous system (Medina and Coslett, 2010), an alteration in the way sensory signals converge from peripheral organs (for example, ectopic impulses or deafferentation) (Gandevia and Phegan, 1999; Paqueron et al., 2003) caused by lesions or dysfunctions in the peripheral nervous system (Vaso et al., 2014), may lead to altered and disordered sensory transmissions to the spinal cord and the brain, and should be considered as an alternative or concomitant plausible hypothesis accounting for phenomena of BPD.

Therefore, considering that about half of patients with FM presented a small-fiber pathology (Grayston et al., 2019) and showed abnormal C nociceptors activity (Serra et al., 2014), we hypothesize that this clinical population may also report BPD as a consequence of peripheral nerve dysfunctions.

To the authors' knowledge, there are no available data about the prevalence of BPD in patients with FM, nor concerning the relationship between small-fiber symptoms and neuropathic pain. This study aimed to explore the prevalence of self-reported BPD in a large cohort of Italian patients with FM distributed nationwide, studying the relationship with the neuropathic dimension of pain (more specifically with small-fiber neuropathic signs or symptoms - SFNS), and with clinical features of FM.

2. Materials and methods

2.1. Design

This study represents part of a larger project aimed at investigating the prevalence of neuropathic pain in patients with FM (Viceconti et al., 2021). We designed a web-based cross-sectional survey according to the Checklist for Reporting Results of Internet E-Surveys (CHERRIES) guidelines and the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) (Appendix 1). Ethical approval was obtained by the Liguria Regional Clinical Experimental Ethics Committee (protocol registration number 290/2018, approved on 29/07/2019).

2.2. Participants and setting

Patients were enrolled through associations (Appendix 2) of FM and rheumatic patients distributed nationwide that were contacted by phone and asked to share the survey via email with their associates. Subjects were considered eligible to participate in our survey if they: (a) aged more than 18 years old; b) declared to have received a clinical diagnosis of FM by a rheumatologist; (c) the clinical characteristics reported were in accordance with the self-administered Fibromyalgia Research Criteria (FRC) for epidemiological studies (Wolfe et al., 2011, 2016); (d) reported no neurological diseases and/or psychiatric comorbidities; (e) had a valid e-mail account; and (f) understood the Italian language. The FRC stated the positivity to FM (FM + group) if the following conditions were satisfied: (1) Widespread Pain Index (WPI) $\geq$7/19 pain sites and Symptom Severity Score (SSS) $\geq$5/12, or WPI between 3-6/19 and SSS $\geq$9/12 [25]; and (2) the presence of symptoms for at least 3 months. The sum of the WPI and SSS generated the 0–31 polysymptomatic distress score (PDS) that represents a measure of FM severity. By definition, FM criteria are fulfilled for a PDS score $\geq$12. However, some patients did not satisfy FRC even in the case of a PDS >12: this is, for e.g., in the case of a patient scoring 5 on WPI and 8 on SSS (PDS = 13). Thus, the FM-group represented patients having the typical FM symptoms across less painful areas with respect to the FM + group. In a post-hoc analysis, these patients were classified in the FM-subgroup, and they were compared to the FM + one. They were included in the analysis for 3 main reasons: (1) patients in this group may have achieved remission in symptoms; (2) they may have been erroneously diagnosed as having fibromyalgia; and (3) they may represent patients with less widespread pain with respect to FM + group, along with the continuum nature of FM symptoms distribution (Wolfe et al., 2013).

Taking into account that in previous similar surveys missing response range varied between 6% and 62% (Firestone et al., 2014; Horton et al., 2017; Salaffi et al., 2016), *a priori* we estimated a drop-out rate of 30%. Accordingly, 1100 invitations were sent to achieve a sample size of 800 participants at the end of the survey. We considered a conservative population proportion estimated at 50%, e.g. the rate of response to each item in the questionnaire is evenly distributed across the sample studied so that in a dichotomous item each category is chosen by 50% of the respondents. The application of the latter value to the formula estimating the size for a single population proportion produced a two-sided interval of 4.5 percentage points within which at 95.0% of confidence (95%CI) is included the true value and a relative standard error of 3.54.

2.3. Questionnaire development and pre-testing

A preliminary list of 43 questions coming from existing questionnaires and an *ad-hoc* created questionnaire (see the paragraph 'questionnaire implementation') was drafted and assessed by a panel of 6 professionals (all physiotherapists) of the research team experienced with clinical evaluation of FM and neuropathic pain assessment, and confidants with the design of electronic surveys. In addition, two patients with FM were involved in the panel aiming at increasing the quality of the design process and for face- and content-validity. The panel decided to exclude six questions from the preliminary version of the survey. The definitive list of questions used in the survey (Appendix 3-4) was the result of a unanimous agreement reached by the panel members on the accuracy of content, words, and structure of sentences (de Leeuw et al., 2008). Before adopting the final version of the survey it was pilot-tested in a convenience sample of 10 subjects with (n = 5) and without (n = 5) a diagnosis of FM to ascertain the clarity and relevance of the question proposed and to verify the time required for completion (Viceconti et al., 2021). The survey was administered using the online tool Survey Monkey (Survey-Monkey, Palo Alto, California, www.surveymonkey.com).

2.4. Questionnaire implementation

The survey was structured in three sections (A, B, and C). Section A included thirteen questions concerning: (a) demographic information; (b) time from the diagnosis and presence of comorbidities; previous or ongoing pharmacological therapies; and (c) multiple-choice questions extracted from the FRC that allowed the calculation of the WPI and the SSS. In section B (containing three items) we explored through *ad-hoc* created multiple choices questions about the phenomenon of BPD in body pats reported as painful by the patients. More in detail, with question 14 patients were asked to check 5 statements that referred to the presence of illusory perceptions of swelling, shrunken or asymmetrical body parts, and the feeling of constriction or heaviness across symptomatic areas. To highlight the concept that these perceptions are illusory, we specified that patients could recognize the difference between what they perceive without seeing or touching and what they feel by touching or looking at those body parts, and that to the visual inspection these body parts appear to be normal. To date, validated outcome measures of this kind of perceptual impairment are lacking,

therefore, the questions included in this section have been inspired by previous studies investigating body perceptual dysfunctions in other clinical conditions (Galer and Jensen, 1999; Lewis et al., 2007, 2010; Wand et al., 2016), adapted for patients with FM who complain of widespread symptoms, and modified based on the authors' clinical experience. Question 15 asked how long patients had experienced BPD while item 16 asked whether patients who reported BPD had referred this phenomenon to health professionals across different specialties.

Section C, provided twenty-one items, including the Italian version of the Neuropathic Pain Symptoms Questionnaire (NPSI) and the multiple-choice question 27 that asked for the presence of 8 small-fiber neuropathic signs or symptoms (SFNS) typically referred by patients with small-fiber pathology and reported in the literature (Terkelsen et al., 2017; Themistocleous et al., 2014; Treister et al., 2017). The self-administered NPSI, through an 11-point numeric rating scale (recall period: last 24-h), asks for the presence of 10 different symptom descriptors that evaluate five different dimensions of neuropathic pain: burning spontaneous pain, pressing spontaneous pain, paroxysmal pain, evoked pain, and paresthesia/dysesthesia. The NPSI has a high test-retest reliability and good construct validity (Bouhassira et al., 2004; de Andrade et al., 2011; Matsubayashi et al., 2015; Padua et al., 2009; Sommer et al., 2011; Villoria et al., 2011) and has demonstrated its utility in phenotyping patients with neuropathic pain (Bouhassira et al., 2021).

Finally, we provided the Italian version of the Fibromyalgia Impact Questionnaire (FIQ) (Sarzi-Puttini et al., 2003). This questionnaire provides 10 questions concerning the condition subjectively perceived by the patients during the week before the administration and includes the assessment of the intensity of pain, fatigue, and stiffness. The total score ranges from 0 to 100, with higher scores indicating worse conditions. The FIQ has good psychometric properties for what concern the test-retest reliability and the internal consistency (Sarzi-Puttini et al., 2003).

2.5. Data collection procedure

The survey was administered between July 30, 2019 and September 14, September 2019. The invitation to complete the questionnaire was sent via email accompanied by the informed consent that was obtained before starting the survey. No incentives were offered to subjects that participated voluntarily. Forced response validation was adopted: participants were required to answer all the questions before submitting each question of the survey to reduce missing data (de Leeuw et al., 2008). The survey was designed to be completed at an average time of 12–15 min to improve the response rate as pointed out by guidelines. More details about the administration of the survey and data management are provided in the study of Viceconti et al. (2021).

2.6. Data analysis

Each questionnaire containing incomplete data or coarse errors was excluded from the analysis. We used Shapiro-Wilk tests and the visual inspection of the quantile-quantile (Q-Q) plots to check data normality. Descriptive statistics, with means and standard deviations (SD) (or median values and interquartile ranges - IQR - for data having a non-normal distribution), were used for demographic and clinical information. For questions containing multiple responses, we reported the absolute and relative frequency of every combination of answers provided by participants. Pain intensity and stiffness were extracted, respectively, from FIQ item-5 and item-8. A score >0 was originally described to detect neuropathic pain through the NPSI and for the Italian version of the questionnaire optimal cut-off values have not been studied. For these reasons, in order to mitigate a plausible excessive of the NPSI sensitivity in detecting neuropathic pain, we set a pre-defined arbitrary threshold at 10 points for avoiding overinflating the prevalence rates. The Welch's 2-sample *t*-test or the Mann-Whitney *U* test was used, respectively for continuous or ordinal variables, to compare variables between the FM+ and FM-subgroups in a post-hoc analysis, while the Chi-squared test with Yates's correction or the Fisher exact test was used to compare frequencies or nominal data. Kendall's τ-b was used for the correlations of BPD with demographic and clinical variables, while Kendall's τ-c test was adopted for correlation analysis between BPD, NPSI, SFNS, and comorbidities.

The alpha value for statistical significance was set at $P < 0.05$ for all statistical tests (indicated where appropriate). A custom-made script using R software *v3.4.1* was used for statistical analysis, with the adoption of the packages psych *v1.7.8* and ggplot2 *v3.0.0*.

3. Results

A total sample of 1173 subjects was enrolled through 7 associations of patients that accepted to receive our invitation by e-mail and forward it to their subscribers (the full list of associations is reported in Appendix 2). Six associations did not provide an answer to our invitation, also after some reminders. We analyzed 854 patients (72.8%) after the exclusion of 281 questionnaires (23.9%) due to incomplete or missing data. A flowchart (Fig. 1) details the enrolment process. The demographic characteristics of patients enrolled are summarized in Table 1.

The figure details the process for including/excluding subjects until the final analyzed sample was obtained.

Clinical variables are summarized in Table 2: most patients (n = 712; 83.4%) reported no comorbidities. Notably, 97.4% (n = 832/854) of patients reported an NPSI score ≥ of 10 that indicates a possible underlying neuropathic pain status in these patients. At the same time, 90.3% of subjects (n = 771/854) reported at least 1 SFNS, suggesting a possible specific involvement of the small-fibers.

As shown in Table 3, most of the patients (42%), reported no

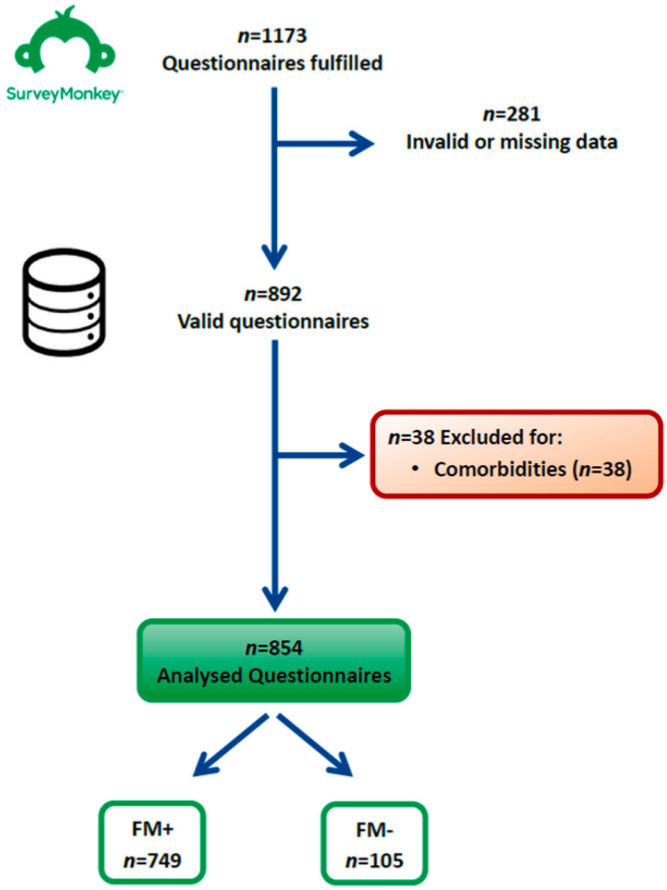

Fig. 1. Enrollment process flow-chart.

Table 1
Demographic-related information for patients enrolled in the study.

	n = 854	
	Values	95%CI
Gender, n (%)		
Female	803 (94.0)	92.2–95.5
Male	51 (6.0)	4.5–7.3
Age, Years (%)		
Mean (SD)	48.1 (10.5)	47.3–48.8
Ranges (min.-max.)	18.0–80.0	–
Italian Region, n (%)		
North	470 (55.0)	51.6–58.4
Centre	132 (15.5)	13.1–18.1
South	145 (17.0)	14.5–19.7
Islands	107 (12.5)	10.4–15.0
Education, n (%)		
PhD	12 (1.4)	0.8–2.5
Bachelor's degree	238 (27.9)	25.0–31.0
High school	462 (54.1)	50.7–57.5
Secondary school	139 (16.3)	13.9–19.0
Primary school	3 (0.3)	0.1–1.1

number of participants (n); percentage (%); minimum value (min.); maximum value (max.); Standard Deviation (SD); 95% confidence interval (95%CI).

assumption of any drugs. Among the patients that declared drugs assumption, the 2 active principles most frequently taken were duloxetine (19.5%) and cyclobenzaprine (15.3%).

3.1. Prevalence of body perception distortions

As reported in Table 2 the median number of BPD reported by patients was 2 (range: 0–5; IQR: 2). The percentages of patients that reported at least one BPD was 90.3% (n = 771/854), while 9.7% (n = 83/854) of subjects reported no episodes of BPD.

As reported in Table 4, the three most frequently reported BPD were phantom feelings of 'heaviness' (74.9%), 'constriction' (50.8%), and 'swelling' (47.2%). Fig. 2 shows the frequencies of single and multiple responses provided by patients enrolled.

As detailed in Table 5 in 62.5% of cases, patients with BPD reported to have talked about the presence of these phenomena with specialist physicians, and in 48.5% of cases with general practitioners.

Results show as percentages of individual single choices and a combination of responses. A: Body parts perceived as "swollen"; B: Body parts perceived as "asymmetrical"; C: Body parts perceived as "shrunk"; D: Body parts perceived as "heavy"; E: Body parts perceived as "constricted"; F: no BPD.

3.2. Between-groups differences in post-hoc analysis

On the base of scores obtained from the administration of the FRC, we considered in post-hoc analysis two subgroups of patients, FM+ (n = 749) and FM- (n = 105). As shown in Table A1 (Appendix 6), no significant between-groups differences were found for gender, age, and regional provenience ($P > 0.05$), while the educational level was higher in the FM-group ($P > 0.001$). More details about the clinical characteristics of both sub-groups are shown in Table A2 (Appendix 6).

Table A3 (Appendix 6) reported the prevalence rates of BPD for the two sub-groups. At least 1 BPD was found in 67.3% 92.3% of FM + patients (n = 691/749) and in 76.2% (n = 80/105) of FM-ones ($P < 0.001$). Fig. A1 and Fig. A2(Appendix 5) show the frequencies of single and multiple responses provided by patients, respectively, in the FM+ and FM-groups. Table A4(Appendix 6) shows the difference between sub-groups after post-hoc analysis of the distributions for the categories of professionals to whom patients have previously talked about the presence of their BPD.

Table 2
Clinical-related information for patients enrolled in the study.

Clinical Variables	n = 854	
	Values	95%CI
Comorbidities, n (%):		
None	712 (83.4)	80.7–85.8
Non-active cancer	40 (4.7)	3.4–6.3
Rheumatoid Arthritis	73 (8.5)	6.8–10.6
Diabetes	34 (4.0)	2.8–5.6
FM diagnosis (months):		
Median (IQR)	36 (45)	–
Ranges (min.-max.)	1–400	–
WPI, median (IQR)	11 (7)	–
SSS, median (IQR)	9 (3)	–
PDS, median (IQR)	20 (8)	–
Symptoms Duration, months:		
Median (IQR)	48 (70)	–
Range (min.-max.)	1–564	–
Pain Intensity (last week), 0–10 NRS:		
Median (IQR)	7.0 (3)	–
Stiffness (last week), 0–10 NRS:		
Median (IQR)	8.0 (3)	–
Disability, FIQ (0–100):		
Median (IQR)	68.6 (21.0)	–
SFNS (0–8):		
Median (IQR)	2.0 (4.0)	–
Neuropathic Pain, NPSI:		
Median (IQR)	54.0 (34)	–
Occurrence of Neuropathic Pain:		
NPSI ≥10, n (%)	832 (97.4)	96.1–98.4
SFNS:		
None, n (%)	83 (9.7)	7.8–11.9
≥1, n (%)	771 (90.3)	88.1–92.2
≥3, n (%)	399 (46.7)	43.3–50.1
≥5, n (%)	117 (13.7)	11.5–16.2
≥7, n (%)	20 (2.3)	1.4–3.6
BPD		
Median (IQR)	2.0 (2.0)	–
Range (min.-max.)	0–5	–
Duration of BPD		
Median (IQR)	36.0 (42.0)	–
Range (min.-max.)	0–500	–

number of participants (n); percentage (%); minimum value (min.); maximum value (max.); Interquartile Range (IQR); 95% confidence interval (95%CI); Neuropathic Pain Symptom Inventory (NPSI), range from 0 (no neuropathic pain symptoms) to 100 (worst neuropathic pain symptoms); Fibromyalgia Impact Questionnaire (FIQ), range from 0 (no impact on health status) to 100 (maximum impact on health status); 0–10 Numeric Rating Scale (0–10 NRS), range from 0 (no pain) to 10 (worst pain as possible); Widespread Pain Index (WPI), range from 0 (none painful areas) to 19 (maximum number of painful areas); Symptoms Severity Score (SSS), range from 0 (low symptoms severity) to 12 (high symptoms severity); Polysymptomatic Distress Scale (PDS), range from 0 (low fibromyalgia severity) to 31 (high fibromyalgia severity), Small Fibers Neuropathic Symptoms (SFNS), range from 0 (no neuropathic symptoms) to 8 (maximum number of neuropathic symptoms); Body Perception Distortions (BPD), range from 0 (no body perception distortions) to 5 (maximum number of body perception distortions).

Table 3
Pharmacological treatments.

	n = 854	
	Values	95%CI
Drugs, n (%)		
No drugs assumption	359 (42.0)	38.7–45.4
Duloxetine	167 (19.5)	17.0–22.4
Cyclobenzaprine	131 (15.3)	13.0–18.0
Pregabalin	113 (13.2)	11.1–15.7
Amitriptyline	90 (10.5)	8.6–12.8
Tramadol	82 (9.6)	8.1–12.5
Cannabinoids	81 (9.5)	7.6–11.7
Fluoxetine	27 (3.2)	2.1–4.6

number of participants (n); percentage (%); 95% confidence interval (95%CI).

Table 4
Prevalence of body perception distortions.

Body Perception Distortions of Painful/Stiff body parts	n = 854	
	Values	95%CI
Occurrence of BPD: n (%)		
0	83 (9.7)	7.8–11.9
≥1	771 (90.3)	88.1–92.2
≥2	547 (64.1)	60.7–67.3
≥3	246 (28.8)	25.8–32.0
≥4	54 (6.3)	4.8–8.2
BPD typology: n (%)		
Body parts perceived as "heavy"	640 (74.9)	71.9–77.8
Body parts perceived as "constricted"	434 (50.8)	47.4–54.2
Body parts perceived as "swollen"	403 (47.2)	43.8–50.6
Body parts perceived as "asymmetrical"	123 (14.4)	12.1–16.9
None	83 (9.7)	7.8–11.9
Body parts perceived as "shrunk"	31 (3.6)	2.5–5.1

Legends: number of participants (n); percentage (%); Body Perception Distortions (BPD), range from 0 (no body perception distortions) to 5 (maximum number of body perception distortions).

3.3. Correlation analyses

BPD were correlated with all clinical variables in the whole sample of the patients (Table 6). In particular, the strength of correlation was 'moderate' (Lee, 2016; Richardson, 2010; Walker, 2003) with SFNS (τ-b = 0.33; $P < 0.01$) and NPSI ($\tau = 0.29$; $P < 0.01$), and 'small to moderate' with PDS ($\tau = 0.28$; $P < 0.01$); WPI ($\tau = 0.26$; $P < 0.01$), SSS ($\tau = 0.22$; $P < 0.01$), and with FIQ ($\tau = 0.20$; $P < 0.01$). In a post-hoc analysis, we didn't find any significant differences in correlation coefficients between the subgroups FM+ and FM-.

BPD were not significantly correlated (τ-c = 0.01, $P > 0.05$) with the presence of comorbidities (diabetes, non-active cancer, and rheumatoid arthritis), while both the NPSI and the SFNS were only poorly correlated (NPSI: τ-c = 0.06, $P < 0.01$; SFNS: τ-c = 0.05, $P < 0.01$). Considering also the low prevalence of comorbidities referred by our patients (83.4%), we can hypothesize that the presence of comorbidities may have possibly played a limited influence on the prevalence of BPD.

4. Discussion

To the authors' knowledge, this study is the first conducted to investigate the prevalence of self-reported body perception distortions (BPD) in a large sample of patients with FM, studying the correlation with neuropathic features and other clinical data. Our main findings revealed that among the sample analyzed, a large portion of the sample reported a minimum of 1 BPD. This phenomenon appears to be significantly correlated with the severity of FM. In fact, a higher prevalence rate of BPD was found in the FM + sub-group that represents patients affected by more severe symptoms with respect to those in FM-. By contrast, symptoms duration seems only to minimally interfere with the prevalence of BPD. Disorders of body perception were also significantly correlated with neuropathic pain and, especially, with symptoms potentially indicative of small-fiber pathology, with correlation coefficients that were greater in the FM + group than in FM-. We found a significantly higher mean scoring for neuropathic pain in FM + concerning FM-, as well as a higher number of SFNS. These findings suggest that the phenomenon of BPD in these patients may be partially explained by a potential underlying neuropathic status and that, conceivably, the pain syndrome may be potential of neuropathic origin in these patients. This hypothesis is supported in the literature by evidence that highlights the presence of neuropathic conditions in about half of patients with FM (Grayston et al., 2019), as well as by clinical (Pazzaglia et al., 2019; Vaso

Table 5
Categories of professionals to whom patients reported the presence of Body Perception Distortions.

	FM+ (n= 771) [a]	
	Values	95%CI
Professionals: n (%)		
Specialist Physicians	482 (62.5)	59.0-65.9
General Practitioners	374 (48.5)	44.9-52.1
None	118 (15.3)	12.8-18.0

Number of participants (n); percentage (%); 95% confidence interval (95%CI).
[a] subjects with body perception distortions.

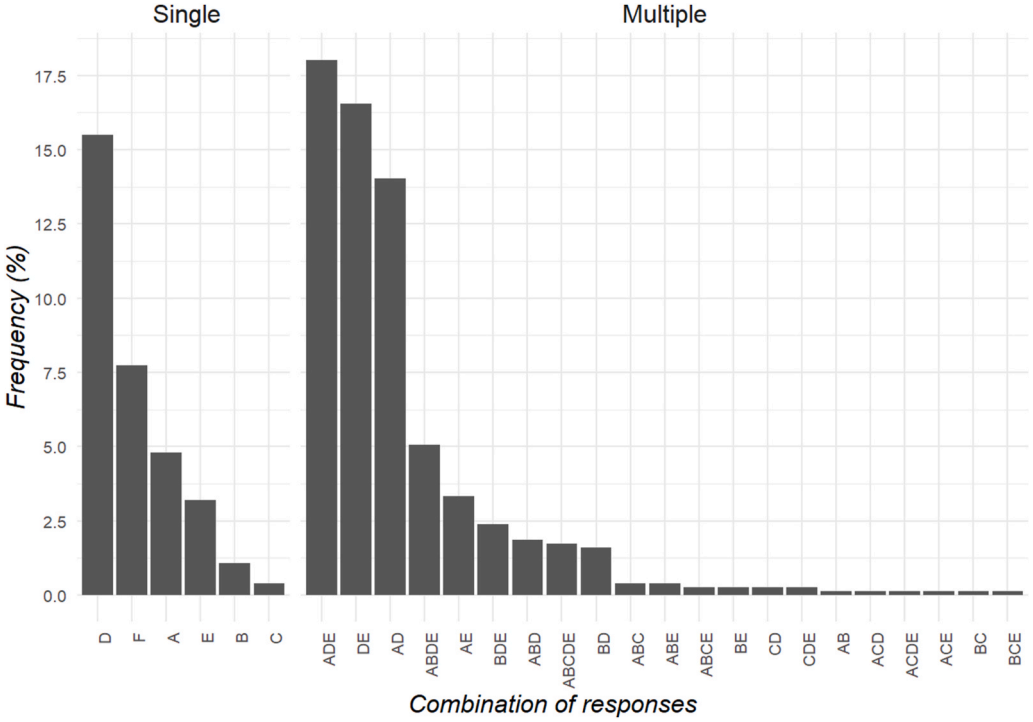

Fig. 2. Body perception distortions: frequencies of single and multiple responses.

Table 6
Correlation analysis between body perception distortions and demographic/clinical variables.

Outcome Measures	BPD *Correlation coefficient for Kendall's τ-c*			
	Entire Sample	FM+	FM-	P [b]
Age	*−0.03*	*−0.02*	*−0.05*	*0.78*
Symptoms duration	*0.12* [a]	*0.12* [a]	*0.52* [a]	*0.001*
BPD duration	*0.17* [a]	*0.15* [a]	*0.27* [a]	*0.23*
NPSI	0.29 [a]	0.27 [a]	*0.27* [a]	*1.00*
FIQ	0.20 [a]	0.17 [a]	*0.10*	*0.50*
SFNS	0.33 [a]	0.30 [a]	*0.22* [a]	*0.42*
PAIN 0-10 NRS	*0.15* [a]	*0.11* [a]	0.14	0.77
STIFFNESS 0-10 NRS	*0.16* [a]	*0.13* [a]	*−0.07*	*0.06*
WPI	*0.26* [a]	*0.22* [a]	*0.08*	*0.17*
SSS	*0.22* [a]	*0.17* [a]	*0.12*	*0.63*
PDS	*0.28* [a]	*0.25* [a]	*0.22* [a]	*0.76*

p-value of tests for comparisons between correlation coefficients of subgroups FM+ and FM- (P); Neuropathic Pain Symptoms Inventory (NPSI), range from 0 (no neuropathic pain symptoms) to 100 (worst neuropathic pain symptoms); Fibromyalgia Impact Questionnaire (FIQ), range from 0 (no impact on health status) to 100 (maximum impact on health status); Numeric Rating Scale (0–10 NRS), range from 0 (no pain/stiffness) to 10 (worst pain/stiffness as possible); Small Fibers Neuropathic Symptoms (SFNS), range from 0 (no neuropathic symptoms) to 8 (maximum number of neuropathic symptoms); Widespread Pain Index (WPI), range from 0 (none painful areas) to 19 (maximum number of painful areas); Symptoms Severity Score (SSS), range from 0 (low symptoms severity) to 12 (high symptoms severity); Polysymptomatic Distress Scale (PDS), range from 0 (low fibromyalgia severity) to 31 (high fibromyalgia severity); Body Perception Distortions (BPD), range from 0 (none body perception distortions) to 5 (maximum number of body perception distortions).

[a] <0.01.

[b] Fisher's r to Z transformation.

et al., 2014) and experimental data (Paqueron et al., 2003; Walsh et al., 2015), in which BPD have been found when the peripheral neural function was impaired. For these reasons, we think that this hypothesis would deserve further well-designed clinical and neurophysiological studies.

The perception of own's body dimensions, contours, and ownership is usually taken for granted. Therefore, when patients perceive 'bizarre' body distortions, beyond the pain, they may experience negative feelings about their bodies as a consequence. Indeed, the illusory experience of distortions in body perception may be detrimental to the quality of life, social interactions, and, overall, mental health (Lewis et al., 2007; Longo 2015). For these reasons, complaints regarding the alteration in body perception should not be undervalued by health care providers to which patients with FM report their symptoms. Our study did not provide specific questionnaires to assess the psychological impact of BPD in patients that reported their presence. Nevertheless, perceptual dysfunctions could be reasonably considered as having a potential impact both on the overall severity of FM symptoms and its psychological dimension, as highlighted by the association between BPD and both PDS and SSS that have been correlated with higher levels of anxiety and depression (Wolfe et al., 2015). Notably, patients reported the presence of BPD to specialist physicians and general practitioners in more than half of the cases. Thus, in these circumstances, they have presumably reputed that these phenomena constituted elements of clinical relevance for their health status, as well as other symptoms like pain, stiffness, and cognitive difficulties. Unfortunately, the nature of our study prevents us to know what kind of information these patients received and how health professionals manage the communication about this particular clinical condition. For this reason, we suggest conducting future qualitative studies administering interviews both to patients and health professionals.

On the other hand, we found a weak correlation between BPD and pain and stiffness. It is not surprising that BPD were weakly correlated with pain intensity. In fact, by definition, BPD are non-painful distorted feelings related to the body (Russell and Tsao, 2018). Beyond the pure terminological definition, our findings seem to indicate that body and painful perceptions could be two different domains that follow a different clinical course in patients with FM.

The prevalence of BPD in our sample appears to be slightly lower than that found in CRPS patients (48%–84%) (Förderreuther et al., 2004; Galer and Jensen, 1999; Reinersmann et al., 2012), even though an accurate comparison between studies is not possible due to two main issues: 1) different outcome measures have been adopted to study both neuropathic pain and BPD; 2) pain in patients with CRPS affects mainly hands and feet. Therefore, the phenomenon of altered body perception may represent a distinct entity in these two cohorts of patients, and this may also explain a different correlation with pain levels.

The FM-group, concerning FM+, showed the lowest prevalence rates for BPD, neuropathic pain, and SFNS, as well as weaker strength of association between these variables. It is possible to hypothesize that the FM-group represents a phenotype of patients characterized by less severe degree and spread of symptoms with fewer neuropathic features of symptoms (Viceconti et al., 2021) and lower frequency of BPD.

If confirmed in future studies, the alterations of body perception may be considered as one of the dimensions involved in the heterogeneous clinical presentation of FM, on a par with pain perception, stiffness, cognitive symptoms, and disability. Especially, the relationship between BPD and small-fiber symptoms deserves further investigations through the adoption of appropriate diagnostic tests (e.g., skin punch biopsy and microneurography) (Devigili et al., 2008; Serra et al., 2014). If this link will be confirmed, the identification of BPD could lead to the suspicion of underlying small-fiber pathology and may help clinicians in managing patients with appropriate diagnostic-therapeutic pathways (Devigili et al., 2008). In line with recent evidence, it should be considered that more than a half of patients with FM may have an undiagnosed small-fiber pathology (Grayston et al., 2019) and, noteworthy, detecting this condition may be clinically relevant because sometimes is possible to find an underlying cause, e.g. diabetes (Freeman, 2014) or pre-diabetes (Kirthi et al., 2021) and to suggest appropriate treatments.

Moreover, if the association between BPD and psychological distress will be proved, it may represent a specific area of therapeutic intervention, for example through targeted informative and educational strategies, especially for those medical categories like specialist physicians and general practitioners to whom more often patients with FM seem to report BPD phenomena.

4.1. Strengths and limitations

The descriptive nature of this study prevented us from drawing any conclusions about the causality effect between BPD and the clinical variables considered. The absence of a clinical or neurophysiological evaluation did not allow the confirmation of the neuropathic origin of symptoms reported by patients and to ascertain the exclusion of medical conditions like diabetes (Freeman, 2014) and pre-diabetes (Kirthi et al., 2021) that may contribute at least in part to the neuropathic symptoms recorded in our survey. However, we explicitly asked patients for a known diagnosis of diabetes and only 4% of them reported this comorbidity.

We explicitly required in both the email invitation and consent form to participate in our study only in presence of a previous diagnosis of FM obtained by a rheumatologist. However, we cannot reliably be sure that all included subjects have followed this requirement. To overcome this potential limitation, we administered the self-reported FRC (Wolfe et al., 2011) that have shown good results with respect to the 2010-ACR criteria administered by physicians (Wolfe et al., 2016).

The arbitrary adoption of a 10-points cut-off for the NPSI may have underestimated the prevalence rate for neuropathic pain in our sample. Nevertheless, estimating the prevalence of neuropathic pain in patients

with FM was not the primary goal of this study for which readers may refer to Viceconti et al., (2021). Considering the lack of a consensus about the meaning of the strength of correlations and the different approaches adopted within the various research areas and specialties (Akoglu 2018), cautions should be taken in interpreting the categorical levels of associations.

We only asked participants about their educational level and not their income and race: both factors may result as confounding factors and these data should be collected in future epidemiological studies. Finally, a major part of the respondents lived in North Italy. Considering that, the perception of pain may be influenced by the social context, we must take into account that the sample of patients was unbalanced under this point of view, therefore were non-representative of the Italian population and this could limit the interpretation of the data. Nevertheless, this study provides preliminary findings on the phenomenon of BPD in a large sample of Italian patients with FM and suggests a working hypothesis for further studies on the potential role played by SFNS.

5. Conclusions

Distortions of the body were perceived by nearly two-thirds of patients with fibromyalgia who completed our online survey. Body parts are more commonly perceived as “heavy”, “constricted” or “swollen”. This distorted perceptual phenomenon was significantly correlated with the severity of fibromyalgia, the number of painful sites, and mostly with neuropathic pain and symptoms potentially indicative of small-fiber pathology.

In light of these preliminary findings, self-reported body distortions should be carefully investigated by clinicians and potentially considered as part of the clinical presentation of fibromyalgia. The phenomena of body perception disorders in patients with fibromyalgia deserve further investigation.

Authors contributions

All authors of this discussed the results and commented on the manuscript.

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Data availability

The data underlying this article will be shared at a reasonable request to the corresponding author.

Declaration of competing interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.msksp.2022.102570.

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Further reading

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