

Gender differences on psychological factors in fibromyalgia: a systematic review on male's experience

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ABSTRACT

Objective. Fibromyalgia (FM) is highly prevalent in female gender. Scarce attention has been given to the exploration and description of this syndrome, from a psychological point of view, when occurring in males. The aim of the present study is to develop further knowledge, and to summarise the literature regarding subjective psychological experience, characteristics of symptoms presentation (both onset and development), and treatment options for FM in male patients, in order to highlight differences with FM in females.

Methods. All studies published between January 1993 and February 2020 using PubMed and PsycInfo were included, provided that they met the following criteria: 1) written in English; 2) original articles on studies with a longitudinal design; 3) prospective or retrospective, observational (analytical or descriptive), experimental or quasi-experimental, controlled or non-controlled studies. Reviews and non-original articles (i.e. editorials, Letters to the Editor, and book chapters) were not included. We utilised the following keywords: (male), (female), (fibromyalgia), combined with Boolean operators 'AND' and 'NOT'.

Results. We found an initial number of 55 papers. Duplicated records were excluded (n=13), as well as papers not focusing on male patients or not fulfilling inclusion criteria (n=25), narrowing the research to 17 papers.

Conclusion. FM male patients consider their masculine identity as inefficiently re-negotiated after symptoms' onset. FM males tend to endure pain for longer periods of time than females before seeking for treatment; bodily symptoms are prevalent with a compromised exploration of feelings about FM. Unfortunately, there is still pau-

city of evidence on clinical characterisation and treatment options when FM occurs in males. Moreover, no studies addressed the issue of the psychopharmacological/non-pharmacological management of males with FM and comorbid psychiatric syndromes.

Introduction

The term 'fibromyalgia' (FM) defines a chronic disease characterised by the presence of a widespread musculoskeletal pain with unknown and debated aetiology. FM also identified a syndrome, with chronic musculoskeletal pain as 'core manifestation', surrounded by a halo of several extra-skeletal symptoms. Thus, alongside with pain, patients describe muscle stiffness, sleep disturbances, asthenia, headache and cognitive impairment (1-5). FM prevalence in the general population ranges from 0.2 to 6.6% (6). The typical age at onset is bimodal, namely between 25 and 35 years, or between 45 and 55 years; FM is more represented in females than in males, with an approximate ratio of 9: 1 (7, 8). However, many authors pointed out that the lack of standardised diagnostic procedures for FM might have influenced the accuracy of epidemiological data and, at least in part, this might explain the variability of results (9). Currently, FM is classified as one of the most common rheumatological diseases, and one of the most frequent causes of chronic pain (10). The heterogeneity of FM clinical manifestations and the absence of a univocal definition contributed to the difficulties in defining a standard treatment for this syndrome (11, 12). Recently, attention has been devoted not only to FM physical manifestations, but also to its psychological characteristics, starting from a bio-psycho-social perspective (13-17).

Competing interests: none declared.

Personality profiles of FM patients are characterised by the inhibition of one's emotions and needs, by a '*strong propensity for self-sacrifice*' and by traumatic past experiences (18-20). Difficulties in recognising and expressing emotional states are common in FM patients, raising questions on diagnostic boundaries between FM and alexithymia (21-24) as well as between FM and the so-called '*type D personality*', mainly characterised by '*negative affect*' and '*severe socio-emotional inhibition*' (25-27). Studies of personality traits based on the 'Big Five Model' (28) showed low levels of awareness and extroversion, and high levels of neuroticism as the most common profiles in patients with FM. Considering the neuropsychological and cognitive profile associated with certain medical and psychophysical conditions induced by external causes may be useful in determining possible treatments (29, 30). Studies on cognitive variables in patients with FM found biased cognitive styles, control perception and coping strategies, with memory losses, fibro-fog, and tendency to catastrophise (31-35). More specifically, brooding and rumination were frequently associated with the severity of FM symptoms, and with the stress levels. These thinking styles were perceived as expensive both for the mind (as they persistently engage the working memory) and for the body (leading to an increased muscular tension) (36). Tendency to catastrophise pain is a risk factor for the development of anxiety and depression in FM patients, related to a maladaptive coping style, and to poor outcome (37-39). Furthermore, catastrophising absorbs subject's cognitive resources, leading to an increased pain perception (40). From a psychopathological point of view, several studies pointed to the association between FM and several psychiatric disorders as factors that might affect disability levels. Major depressive episodes are frequently comorbid (50-60% of patients with FM), as well as anxiety disorders, including Post-Traumatic Stress Disorder (PTSD) (20-60%) whose characteristics may change depending on the patient's gender (41-48). FM patients,

due to their high stress/anxiety levels related to pain, appear to be at risk of developing sexual dysfunction as well (49, 50). Bipolar spectrum disorders, Obsessive-Compulsive Disorder (OCD), and specific phobias are less frequently described in comorbidity with FM (4). Given the complexity of clinical FM presentation, the therapeutic approach encompasses pharmacological and non-pharmacological methodologies (51-53). Some non-pharmacological options are effective in reducing subjective discomfort and objective disability, such as mindfulness, music therapy, yoga, tai-chi, basic body awareness therapy, psycho-education and cognitive behavioural therapy (CBT) (54-58).

All these data derive from studies on FM in females. Less attention has been devoted to FM exploration and description in males (59). Literature on disparity in painful stimulus processing between genders still reports controversial results. One of the hypotheses on the differences in pain modulation capacity between the two genders suggests that environmental pressures and social habits might have played a role during evolution. Thus, males, as '*hunters and warriors*', were more likely to experience acute pain from physical traumas and injuries than females; conversely, females had always faced prolonged visceral pain because of menstrual cycle and pregnancy, with a more efficient pain modulation on the mid/long term, as mediated by endorphin system (60, 61). According to this hypothesis, males might have more tolerance to acute pain, while females could have developed an attitude to manage chronic pain. However, it is also possible that females might be more used to report on severe and frequent pain than males, or on pain of longer duration than males (62). Studies are concordant in finding that the subjective perception of chronic pain is a clinically relevant variable when studying gender differences in subjective pain threshold and analgesia (63). Furthermore, psychological and social factors, such as stress levels, coping strategies, psychosocial attitude, and housing condition, contribute to gender

differences in pain perception and tolerance (64-67). It is still unclear whether these aspects might have a relevance in differentiating males and females with FM. Males with FM may present fewer tender points and less common '*hurt all over*', fatigue, morning fatigue, and irritable bowel syndrome, than female patients (68-71). It has been also observed that males with FM might be more frequently characterised than females by a sympathetic hyperactivity and a lower parasympathetic activity, together with an abnormal sympathovagal response during postural changes (72). Other studies suggested that '*a number of mechanisms may contribute to a different symptom expression in men and women with this spectrum of illness*', including a different heart rate variability (73).

Aim of this study was to systematically review available finding on males affected by FM, to highlight their psychological and clinical characteristics, and to enhance the available therapeutic options for this population of patients.

Methods

We adhered to the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA) guidelines in completion of this systematic review (74).

Protocol registration

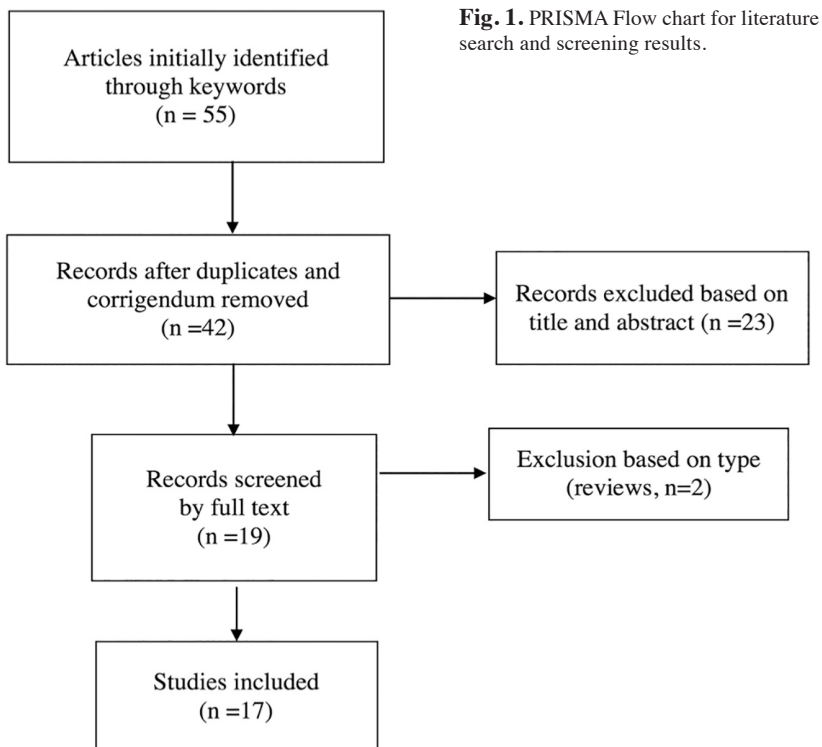
This systematic review is not included in a research protocol.

Eligibility criteria

Randomised clinical trials (RCTs), cross sectional, narrative and observational studies published in English up to January 2020 were included in our review. RCTs are regarded as the best evidence for treatment. However, based on a preliminary review of the literature, a lack of RCTs was expected. Thus, observational studies, case reports and series were included (Table II).

Literature search

All studies published between January 1993 and February 2020 using Pub Med and PsychInfo were included, provided that they met the following criteria: 1) written in English; 2) original articles on studies with a longitudinal design;



and 3) prospective or retrospective, observational (analytical or descriptive), experimental or quasi-experimental, controlled or non-controlled studies. Reviews and non-original articles (*i.e.*, editorials, Letters to the Editor, and book chapters) were not included.

Information source and search strategy

The literature search was designed and independently performed by two authors (RC and CC). We selected papers that presented a combination of at least two of the following terms in the abstract, namely: (male), (female), (fibromyalgia), combined with the Boolean operators 'AND' and 'NOT'. Inclusion criteria were as follows: the article must be in English and must contain quantitative or qualitative data about FM males.

We found fifty-five papers. After duplicates checking, thirteen papers were excluded (13/55; 23.6%). Twenty-three papers were excluded because not focused on FM or not in English (n=23/55; 41.8%). Moreover, two papers were excluded because reviews on other topics, methodological papers or editorials (n=2/55; 3.6%), thus leading to a final selection of seventeen papers

(17/55, 31%) (See PRISMA flow diagram for a detailed description).

Study selection

Two authors independently screened the resulting articles for their methodology and appropriateness for inclusion (RC and MM). All studies were summarised in Table I. Consensus discussion was used to resolve disagreements between reviewers.

Risk of bias assessment

Two reviewers (RC and MM) independently tried to assess the risk of selection, performance, detection, attrition, reporting, and other bias using the Cochrane risk of bias tool (75). However, an 'unclear risk of bias' emerged for all the selected papers, considering that insufficient data for a judgment were provided. A meta-analysis could not be conducted due to the small number of studies that met the inclusion criteria, as well as the heterogeneity in study designs, methods used, and samples collected.

Results

The main characteristics of the seventeen studies are summarised in Table I. Two studies were performed using a

randomised clinical trial methodology (2/17, 12%), five were narrative case studies (5/17, 29%), four were cross-sectional studies (4/17, 24%), two were case reports (2/17, 12%) and there was one element of the following types of articles: qualitative study, follow-up observational study, pre-post intervention and clinical observational study (1/17, 6% x 4).

The number of male subjects included in the studies ranged between 1 and 3757 (mean: 308.94±888.03), for a total of 5252 patients. The samples sizes and their compositions varied widely and depended on the characteristics of the setting. The adoption of outcome measures was largely inhomogeneous: we found 53 different instruments.

Data synthesis

Due to the lack of homogeneity among the resulting studies, a meta-analysis could not be performed. Studies widely varied in terms of how improvements were measured and on sample selection criteria.

Hence, this systematic review is presented as a narrative synthesis, according to three topics, namely: a) General and specific aspects of male FM psychology (8/17); b) Narrative approaches (5/17); c) Treatment options (4/17). In the first section, eight papers are included; two cross sectional studies (76, 77), two cross sectional/descriptive/correlation studies (78, 79), two RCT (80, 81), one observational (82), and one qualitative study (83). The second section contains five of the narrative case studies that emerged from our research (84-88). The last section gives an overview of the treatment options for males suffering from FM and includes a case report (89), a pre-post intervention study (90), a multiple patient case report (91), and a follow up observational study (92). For a more detailed description of the selected studies (Table II).

a) General and specific aspects of male FM psychology

Several psychological characteristics seem to significantly influence FM when occurring in males, such as comorbidity with anxiety and depression, low quality of life, sexual dysfunction,

Table I. Studies listed for publication per year.

No.	Authors	Year	Title
1	Culclasure <i>et al.</i>	1993	Post-traumatic stress disorder presenting as fibromyalgia.
2	Paulson <i>et al.</i>	2002	Men living with fibromyalgia-type pain: experiences as patients in the Swedish health care system.
3	Paulson <i>et al.</i>	2002 (b)	Struggling for a tolerable existence: the meaning of men's lived experiences of living with pain of fibromyalgia type.
4	Amital <i>et al.</i>	2006	Post-traumatic stress disorder, tenderness, and fibromyalgia syndrome: are they different entities?
5	Rutledge <i>et al.</i>	2007	Predicting high physical function in people with fibromyalgia.
6	Mac Farlane <i>et al.</i>	2009	Investigating the determinants of international differences in the prevalence of chronic widespread pain: evidence from the European Male Ageing Study.
7	Sadrediny <i>et al.</i>	2010	Sexual disorder improvement: a target or a way in treatment of fibromyalgia. A case report and brief review.
8	Yoshikawa <i>et al.</i>	2010	A comparison of quality of life, demographic and clinical characteristics of Brazilian men with fibromyalgia syndrome with male patients with depression.
9	Carbonell Baeza <i>et al.</i>	2011	Preliminary findings of a 4-month Tai Chi intervention on tenderness, functional capacity, symptomatology, and quality of life in men with fibromyalgia.
10	Carbonell Baeza <i>et al.</i>	2011 (b)	T'ai-Chi intervention in men with fibromyalgia: a multiple-patient case report.
11	Batmaz <i>et al.</i>	2012	Sexuality of men with fibromyalgia: what are the factors that cause sexual dysfunction?
12	Karper <i>et al.</i>	2013	Exercise effects on two men with fibromyalgia syndrome: an update.
13	Segura-Jiménez <i>et al.</i>	2016	Subgroups of fibromyalgia patients using the 1990 American College of Rheumatology criteria and the modified 2010 preliminary diagnostic criteria: the al-Ándalus project.
14	Sallinen <i>et al.</i>	2017	"I just want my life back!" - Men's narratives about living with fibromyalgia.
15	Muraleetharan <i>et al.</i>	2018	Understanding the Impact of Fibromyalgia on Men: Findings from a Nationwide Survey.
16	Sallinen <i>et al.</i>	2018	Memory gaps, lost words and crucial mistakes - Men's experiences of cognitive difficulties in fibromyalgia.
17	Sallinen <i>et al.</i>	2019	"I can't have it; I am a man. A young man!" - men, fibromyalgia and masculinity in a Nordic context.

and overlapping symptoms with Post-Traumatic Stress Disorder (PTSD).

The European Male Ageing Study-EMAS gathered data from eight different European countries (Belgium, England, Poland, Estonia, Hungary, Italy, Spain and Sweden), on patients with FM and other chronic pain syndromes (76). The overall sample consisted of 3963 interviewed patients; 313 patients (7.8%) showed a chronic and widespread pain syndrome. The main aim of this study was to determine the prevalence of chronic pain, and its relationships with psychological and psychosocial variables. Overall, there were differences in widespread chronic pain amongst the different European countries, with a higher prevalence in Eastern Europe. Measurements were performed on pain, health and on the occurrence of stressful life events (SLEs). Chronic widespread pain showed a strong association with depressive symptoms, number of comor-

bidities and number of reported SLEs, as well as with psychosocial factors, such as adverse life events, and poor psychological health.

Yoshikawa *et al.* (78) assessed QoL in Brazilian males with FM compared to patients with unipolar depression (MDE). Fifty FM male patients and twenty patients with unipolar depression (MDE), aged between 18 and 72 years, underwent a medical screening with blood collection, physical examination, and thumb palpation for tender points. Psychological tests included anxious and depressive symptoms. FM patients were more compromised than MDE patients in general health physical function, role physical, body pain, and energy levels. The two groups were similar in depressive symptoms, but FM patients showed higher values in state-related anxiety. The relationships between sexual hormonal levels and stress have been also evaluated. The two groups (FM and MDE pa-

tients) did not differ on mean values of testosterone, even if lower levels of total testosterone were found in FM than in MDE patients.

The research group of Segura-Jiménez (81) investigated symptom profiles of FM patients, in a cross-sectional study on 913 Spanish patients (84 males: 9.2%), using a combination of the 1990, and 2010 diagnostic criteria. Measurements included levels of depression, anxiety, pain, catastrophising, cognitive tests, bioelectrical impedance, tender points' examinations, and physical fitness tests. This study confirmed the lack of standardisation of diagnostic processes, aetiology and treatments. Moreover, there was a further relevant variable, namely the heterogeneous nature of the FM patients' phenotypes. The authors proposed a phenotypic sub-typing, based on cases complexity, presence of specific symptoms, and treatments' response. The study showed that the combination of

1990 and 2010 criteria improved the accuracy of FM diagnosis as well as the characterisation of different FM phenotypes, according to symptoms presentation and severity.

Qualitative data from a national survey study by Muraleetharan *et al.* (83) showed that males with FM suffered for a relevant physical disability, together with depression and anxiety, deeply affecting subjective perception of QoL. Psychosocial variables were subjectively relevant: males complained about the loss of their ability to work, worsened by *'the load that society imposes on them through role-based social expectations'*, related to the stigma of having a syndrome *'typical of female gender'*.

Rutledge *et al.* (79) investigated potential relationships between physical function in FM patients and symptom status, psychosocial factors, and self-management. The aim was to understand the presence of variables with respect to normal levels of functioning. The strongest predictor of high physical function was *'being a man'*, followed by age and education: younger and better-educated people were more likely to be higher functioning. Engagement in exercise also represented a predictor of good physical function, together with not using relaxation methods and pain killer medications.

The systematic review regarding more specific psychological/syndrome aspects of FM in males revealed paucity of reports and findings. We found only three papers focused on the exploration of the potential phenomenological aspects and on the differentiation of the clinical presentation of FM in males.

The first one was a brief clinical observation, from Culclasure *et al.*, of a 47-year-old man with chronic muscle pain and generalised fatigue (82). The authors concluded that the patient was suffering for a PTSD, triggered by a prolonged traumatic experience as prisoner of war (7 years) in Vietnam. In this case, FM syndrome was interpreted as a possible presenting manifestation of a severe PTSD. Following the same research line, Amital *et al.* (77) investigated comorbidity between PTSD and FM syndrome, in a

sample of men with PTSD (n=55) compared with twenty patients with major depression (MDE), and 49 controls. PTSD was always induced by traumatic combat related events. A count of 18 tender points (TPs) was performed by thumb palpation, and patients were considered to have FM syndrome if they met the American College for Rheumatology (ACR) criteria for the definition of FM syndrome (*e.g.* widespread musculoskeletal pain with excess tenderness in at least 11 of 18 pre-defined anatomic sites). FM syndrome was overly represented in the sample of male patients with PTSD (49%), compared with patients with MDE (5%), and controls (FM absent). According to this study, PTSD in male patients was highly associated with FM syndrome, with a combined impact on the overall functioning and quality of life (QoL). It was unclear, though, whether PTSD precipitated the emergence of FM syndrome, or if these two disorders might coexist as independent comorbid syndromes, or, finally, if there was a substantial overlap between the two diagnostic entities.

Sexual function of male subjects with FM syndrome was investigated in a group of 37 sexually active men compared to 30 healthy controls (HC) (80). Both groups included married men between the ages of 20 and 50, who were sexually active during the last 4 weeks before study enrolment. Sexual function was assessed according to the international index of erectile function (IIEF) scoring system. Quality of life (QoL) was measured with the Short Form-36 quality of life questionnaire (SF-36 QoL). The occurrence of anxiety and depressive symptoms was assessed with the Hospital Anxiety and Depression Scale (HADS). The study was conducted with a cross-sectional design in two different sites. As expected, FM syndrome patients scored significantly higher in HADS, compared to HCSF-36 scores were lower in the FM group than in the HC group, as well as the five domains of the IIEF. Taken as a whole, findings from retrieved studies on the general and the specific psychological aspects of FM in males revealed three main findings:

a) males with FM showed a relevant burden of anxious and depressive symptoms having a significant impact on subjective perception of QoL; b) boundaries between PTSD and FM in males were often blurred by the overlap between these two realms, and by the co-occurrence, in a lifetime perspective, of FM, stressful life events, traumatic experiences, PTSD signs and symptoms, and chronic pain/fatigue whose relationships were far from being clarified; c) sexual functioning in FM males was a neglected topic, with a limited number of observations in this field, despite its relevance in terms of subjective well-being and self-esteem.

b) Narrative approaches

Narrative approach has been utilised to explore the meanings that FM male patients might give to their subjective experiences.

Paulson, Norberg and Danielson (84) analysed 14 narrative interviews. The most relevant themes were: *'feeling afraid of being looked upon as being a whiner'*, *'feeling hopeless'*, and *'feeling neglected by others'*. As expected, most of the participants reported a high number of medical consultations and an excessive use of painkillers, associated to antidepressant or corticosteroids, with no relief and a burden of side effects. Psychotherapies were also described, with no clinically relevant results. Males with FM tended to endure pain for long periods of time before seeking treatment, also because they described relevant difficulties in sharing their thoughts and experiences with health care professionals. Bodily events were the focus of their negative thoughts. The same sample was analysed by Paulson *et al.* (85) that used a phenomenological hermeneutic interpretation inspired by Ricoeur. Experiencing the body as an *'obstruction'* and *'being a different man'* or *'striving to endure'* represented the main interlaced themes of the analysis. Intriguingly, sub-themes encompassed *'living with a reluctant body'*, *'living day by day with body in pain'*, *'not being the same man as earlier'*, and *'not being really understood'*.

Recently, the Swedish and Finnish re-

search groups on FM used a qualitative narrative approach as methodological frame (86-88). According to these studies, males with FM tended to look at their experiences with an illness-driven point of view and tried to develop new strategies to manage their social life and work. The analysed samples were small: five FM male patients in the first study (86) and eight in the second one (87). The aim of the first study was to investigate the content of patients' narratives and their dialogue mode, namely 'how patients expressed their illness experiences'. According to this qualitative approach, males with FM acquired a new perspective on their lives, social roles and works. However, not all patients with FM were able to adjust their activities and to adapt to their illness, with a significant impairment of one's dignity, integrity, and future perspectives perception, thus leading to an incomplete role transition.

The second paper was focused on the interplay between illness and gender. Males with FM tended to look at their future as lacking in well-being, subjective health, and personal goals' achievement capacity. Consequently, they sensed their masculine identity as re-negotiable because of FM symptoms. Moreover, males considered the transition from a 'strong and powerful body' to a 'disabled' one as a fundamental aspect of their suffering. Male patients with FM also described a wide range of cognitive problems, from mild to severe, such as memory issues, problem-solving difficulties, and impaired verbal fluency. The 'dyscognition' experienced by males with FM seemed to have a substantial negative impact on their daily lives, work, and social relationships.

In summary, narrative approach revealed some interesting topic regarding the subjective perception and description of FM in males, even if the generalisability of these data was limited, considering the small sample sizes and the emphasis on subjective dimensions difficult to rate in an objective manner. However, narrative approach highlighted the more relevant fears of males with FM, and their subjective suffering mainly related to the perceived menace

to their masculinity, and the perception of an enduring lost healthy-self condition, thus limiting their abilities and social-interpersonal role. Moreover, it is noteworthy the description of cognitive problems, that need to be addressed as a field of therapeutic intervention. We strongly believe that, despite the well-known limitations of narrative approach, it would be important to continue the search for the 'meaning of pain' in these patients, because it could help the clinicians to better understand subjective experiences and to target the development of 'tailored' communication strategies.

c) Treatment options

Four papers were selected, three were case report and one was a 4-month study. One of the three case reports was on a sample of six Spanish men, subsequently re-analysed and described in a four-month study on nine patients (90, 91). The two papers were on the same sample, considering that the statistical analyses of the second paper were performed on six patients out of nine initially enrolled.

The first case report (89) was on a 41-year-old outpatient evaluated in Iran. He was diagnosed with FM and treated with amitriptyline (25 mg twice/day) + fluoxetine (20 mg/day), with no significant improvement during a 6-month follow-up. The complete remission of FM signs and symptoms occurred when the patient changed his sexual behaviours, by accepting his homosexual desires that he had repressed for his entire life.

The second case report (multiple case report) was on a group of six patients with FM treated with T'ai-Chi for a period of 4 months (90), then re-analysed in a second paper (91). The purpose of both observations was to determine the effectiveness of a 4-month T'ai Chi intervention on tenderness and functional capacity. Tenderness was measured using a standard pressure algometer; functional capacity was assessed by the 30-second chair stand, chair sit and reach, 8-foot up and go, and 6-minute walk tests. The impact of FM signs and symptoms was measured with the fibromyalgia impact questionnaire (FIQ).

According to the first observation, T'ai Chi intervention improved body flexibility and promoted positive changes in algometer scores, total score of FIQ, and dimensions of physical function, anxiety, and depression in four patients out of six. No statistical analyses were performed in the first paper. In the second paper, some statistical analysis was performed and, basically, confirmed the first report's results. However, given the small sample size, especially if compared with the number of variables analysed, the generalisability of finding was extremely limited.

The case report of Karper (92) on two men with FM syndrome was an update of a previously published report on the same subjects (93). Patients were followed-up for a total of 4 years to assess the effects of a university-based exercise program, consisting of a walking warm up for approximately 15 minutes, followed by light-dumbbell weight training and other callisthenic type exercises. Patients were then evaluated every 6 months on the quarter mile walking capacity test, the 30-second arm curl muscle strength/endurance test, the handgrip dynamometer strength test, the 30-second wall push-up muscle strength/endurance test, the 30-second chair stand muscle strength/endurance test. Psychosocial functioning and FM symptoms were collected using a 10-point analog scale (1 = extremely negative response and 10 = extremely positive response), developed by Karper to rate upper body pain, lower body pain, fatigue, brain fog, restorative sleep, perceived stress, perceived depression, perceived quality of life, anger at FM (pain), and fear of FM (pain) once per week. Both patients seemed to be stable. Patient 1 slowed in walking (8 seconds) and his arm strength had weakened by four repetitions in 30 seconds compared to the previous observations. Improvement was sustained when comparing the other three measures of physical fitness over time. The psychosocial ratings remained the same on six dimensions and improved slightly on four. Grip strength of patient 2 reduced considerably when comparing average data. Improvement was shown in com-

Table II. Characteristics of the selected studies.

No.	Study design	Sample (n)	Males (n)	Mean age ± Std Dev (Or Range)	Intervention	Outcome measures	Main findings
1	Clinical observational study	1	1	47	Evaluation of chronic muscle pain and generalised fatigue.	Clinical records	Similarities between FM symptoms and PTSD ¹ .
2	Narrative case study	14	14	48 ± NA ²	Narrative interviews were conducted, and a content analysis was performed on results.	Narrative interview	Themes constructed from the structural analysis of the interview: Feeling afraid of being looked upon as being a whiner, feeling like a guinea pig, feeling hopeful, feeling neglected and feeling no recovery.
3	Narrative case study	14	14	47 ± NA	Narrative interviews were conducted, and a phenomenological hermeneutic interpretation was used to analyse results.	Narrative interview	Themes and subthemes constructed from the structural analysis of the interview: Experiencing the body as an obstruction (living with a reluctant body, living day by day with body in pain), Being a different man (not being the same man as earlier, not being really understood) and Striving to endure (living as normally as possible, searching for alleviation, having to nurture hope).
4	Cross-sectional study	124 of which :-55 PTSD -20 MDD ³ -49 controls	124	49,7 ± 7,5	Cross sectional evaluation focusing on clinical expressions of FM signs among PTSD patients.	Sleep History Questionnaire (SHQ), Sheehan Disability Scale, The Short-Form Health Survey 36 (SF-36), Clinician-Administred PTSD Scale (CAPS), Clinical Global Impression Scale (CGI), Hamilton Depression Rating Scale (HADS), FM tenderness assessment	High association between FM and PTSD (49% of the PTSD sample fulfilled the ACR Criteria for FM), specifically in degree and impact characteristics.
5	Cross-sectional descriptive correlational study	2580	93.8% of F	47.2 ± NA	Evaluation of relationships among specific personal characteristics, symptoms, and functional status, along with the effect of self-management strategies on functional status.	Fibromyalgia Association Questionnaire (NFAQ), Physical Ability Impact scale (PAI)	Significant explanatory variables of high physical function were identified in 'being men', 'greater education', 'younger age', 'lower intensity fatigue', 'spasticity', 'balance problems', 'not using pain medications', 'using aerobic or strength training exercise', and 'not using relaxation methods'.
6	Cross-sectional study	3757	3757	NA	Analysis from a major epidemiological study being conducted among men in eight countries of Europe (EMAS) to compare the prevalence of chronic widespread pain among middle-aged and elderly men across several countries.	Beck Depression Inventory (BDI), Questionnaire for pain experience (ad hoc), Brugha 12-event inventory.	Chronic widespread pain in eastern Europe related to higher levels of adverse psychosocial factors, as well as to the influence of poorer psychological health and physical morbidities
7	Case report	1	1	41	To explore the relationship of sexual disorder with FM by treating the subject with psychoeducation on sexual tendencies.	Fibromyalgia Impact Questionnaire (FIQ), clinical interview.	Single case evidence for efficacy of sexual disorder improvement in the treatment of refractory FM
8	Cross-sectional descriptive correlational study	50 FM 20 MDD	70	43.96, FM 44.35, MDD ± NA	To measure the impact of FM on QoL ⁴ and determine whether male patients experience impaired adrenal and gonadal androgen secretion	Clinical assessment, SF-36, Visual analog scale, Spielberger State-Trait Anxiety Inventory (STAI), Beck Depression Inventory (BDI), Blood collections, radioimmunoassay.	Scores were significantly lower in all the SF-36 domains in patients with FM as compared with depressive patients. No significant differences were observed in hormones' mean concentration. Male patients with FM experienced worse QoL than those with depression.
9	Pre-post intervention	6	6	52.3 ± 9.3	To define the effects of a 4-month Tai Chi intervention on tenderness, functional capacity, symptomatology, and QoL.	Assessment of the tender-points count, 30-second blind flamingo, 30-second chair stand, anthropometry, chair sit and reach, back scratch, 8 feet up and go, handgrip strength, 6-minute walking tests, Fibromyalgia Impact Questionnaire (FIQ), SF-36, HADS, Vanderbilt Pain Management Inventory (VPMI)	Significant improvement after the intervention period for the chair sit and reach test (lower body flexibility) was found. Improvement was maintained after the detraining period.
10	Multiple-patient case report	6	6	52.3 ± 9.3	To define the effectiveness of a 4-month Tai Chi intervention on pain, functional capacity, and the impact of the FM in men.	Same as above	Improved body flexibility and positive changes in algometer score, aerobic capacity, agility-dynamic balance, total score of FIQ, and physical function, anxiety and depression dimensions.

No.	Study design	Sample (n)	Males (n)	Mean age \pm sd (Or Range)	Intervention	Outcome measures	Main findings
11	RCT	37 FM 30 controls	67	37.2 \pm 8.4 FM 35.4 \pm 6.1 controls	To evaluate sexual function in male patients with FM and to observe the relationship between the sexual dysfunction and widespread pain, psychological status and QoL.	HADS, International index of erectile function scoring system (IIEF), SF-36.	Widespread pain, disturbed QoL and advancing age seemed to have a greater impact on sexual function than psychological state.
12	Follow up observational study	2	2	Patient 1: 65 Patient 2: 74	To report how the previously described FM program and exercise protocol (Karper, 2007) have affected the same two men (who were subjects of the previous report) over the past 4 years.	Quarter mile walking capacity test (a modified form of the 880 yard walk test; Osness <i>et al.</i> , 1990), 30-second arm curl muscle strength/endurance test (Osness <i>et al.</i> , 1990), handgrip dynamometer strength test (Stoelting, Chicago, IL, or Country Technology, Inc., Gay Mills, WI), 30-second wall push-up muscle strength/endurance test (developed by the author and a graduate student for use with people who have FM), and the 30-second chair stand muscle strength/ endurance test (Rikli & Jones, 1999)	Both these men still functioned at approximately the same levels (physically and psychosocially) as reported in 2007.
13	RCT	N= 913 (Controls= 285 FM= 628)	84	Controls: 49.1 \pm 10.1 FM: 51.7 \pm 8.1	To investigate symptom profiles in subsets of FM patients according to the subgroups created from the satisfaction of the 1990 American College of Rheumatology (ACR) diagnostic criteria (1990c) and/ or the modified 2010 ACR preliminary diagnostic criteria (m-2010c).	Algomerty, The modified 2010 ACR preliminary criteria, The Mini Mental State Examination (MMSE), Portable eight-polar tactile-electrode impedancimetre (InBody R20, Biospace, Seoul, Korea), Pain Visual Analogue Scale (PVAS), The Chronic Pain Self-efficacy Scale, The Pain Catastrophising Scale, The Revised Fibromyalgia Impact Questionnaire (FIQR), The Spanish version of the Multidimensional Fatigue Inventory (MFI-S), SF-36, BDI-II, STAI, The Functional Senior Fitness Test Battery, The chair sit-and-reach test, The back scratch test, The 8-foot up-and-go, The 30-s chair stand test, The handgrip strength test, The arm curl test, The 6-min walk test	Differences were consistent across all study outcomes showing that the subgroup fulfilling both diagnostic criteria had the worst profile of all. The subgroup fulfilling the m-2010c only had a worse profile than the subgroup fulfilling the 1990c only and presented similar but slightly better results than those fulfilling both diagnostic criteria.
14	Narrative case study	5	5	43 \pm 10.09	To explore how men with FM manage their life and work with a narrative approach.	Narrative interview (three-phase interview method inspired by Rosenthal)	Results were divided in two main models such as: 'Adjusting life to match the illness' and 'Being imprisoned by pain'. Results suggested that adjusting one's activities might help to manage symptoms and to support work ability in many cases. For some patients the experience of feeling healthy or pain free might be 'nothing but a fading memory'.
15	Qualitative study	1,163	925	Between 45 and 64	To understand why FM is underdiagnosed in men by examining multiple impacts of FM regarding interactions in society and the U.S. health system.	Qualitative survey	Thematic analyses of survey responses suggested that men with FM had negative experiences with physical and mental health, QoL, relationships, and careers.
16	Narrative case study	8	8	47 (Range: 24-61) \pm NA	To explore the impact of perceived cognitive problems on daily life in men with fibromyalgia.	Narrative interview (three-phase interview method inspired by Rosenthal)	Themes constructed from the structural analysis of the interview: 'Story of focusing on one thing at a time', 'Story of having memory gaps' and 'Story of losing cognitive work ability'. Men in this study experienced a wide number of cognitive problems, in which severity varied from minor to severe, and frequency from occasional to constant. Dyscognition experienced by men with FM had a substantial negative impact on their daily lives, work and social relations.
17	Narrative case study	8	8	47 (Range: 24-61) \pm NA	To elucidate the interplay between illness and gender by exploring life-stories of men who suffer from fibromyalgia.	Narrative interview (three-phase interview method inspired by Rosenthal)	Masculine identity of the participants was re-negotiated by comparisons to other men and to life before symptom onset, and by discussing expectations and beliefs of how men should act in contemporary societies. The transition from experiencing a strong, active and reliable body to experiencing a painful, vulnerable and helpless body was perceived as fundamental.

¹PTSD: post-traumatic stress disorder; ²NA: not available; ³MDD: major depression disorder; ⁴QoL: quality of life.

parison with the other three measures of physical fitness. On psychosocial symptom ratings, he scored as previously.

Taken as a whole, finding in the considered field was subject to several limitations. The first was that all selected papers were case reports. Moreover, only two papers addressed the question of a longer-term follow-up. Furthermore, the number of instruments used for the assessment of physical, psychological, and psychosocial characteristics of men with FM was too wide and inhomogeneous, especially when compared with the small samples analysed. However, it was interesting that the available studies were mainly focused on treatments other than the psychopharmacological approach, raising questions on the distance between clinical practice and research studies.

Discussion

Summary of evidence

Findings from our systematic review confirm the initial hypothesis of a paucity of empirical evidence both on FM psychological/clinical characterisation and treatment options in males, as already pointed out by previous research that stressed out bias aspects in the diagnosis process (94, 95). The importance of a different approach both on psychological and pharmacological intervention when FM is occurring in the two genders is the main finding of this review, since that males and females are facing FM symptoms through allegedly different biological and psychological structures, and different coping strategies (63).

Unfortunately, studies on this topic showed several and methodological limitations. The overall strength of the available evidence was poor, with a substantial lack of consensus and inconclusive outcomes on available treatment options. Although we have attempted to follow rigorous criteria in the process of assessment of studies included, there were several limitations affecting the generalisability of results. Most studies were conducted at a single centre, with treatment-specific biases arising from small sample sizes, differences in study protocol, and clinical

rather than statistical interpretation of results. These limitations could be interpreted as related to the difficulties in conducting well-designed research studies in male populations with FM, given the unbalanced male/female ratio in this specific area. Other limitations included recruitment difficulties due to the resistance in males when asked to describe their subjective suffering, the diagnostic limitations due to the heterogeneity of clinical FM presentation, the ethical issues in dealing with 'non-conventional' treatments, and the challenges in follow-up, as in other chronic pain conditions (96, 97, 86, 88).

Therefore, the small number of available findings and the variable reporting style of the reviewed studies reduced the summary analysis to a descriptive interpretation. In addition, due to the variability in design and methodological features, the quality assessment of the studies included was challenging and likely biased.

No studies addressed the problematic issue of the psychopharmacological and non-pharmacological management of patients with FM and comorbid psychiatric syndromes, with no specific treatment options offered.

Most studies focused on the importance that should be given towards morbidities such as depressive symptoms and anxiety in chronic pain, as well as on the influence of adverse psychosocial factors and poorer psychological health (98-101). Furthermore, physical function, vitality, general health, and body pain resulted to be more compromised in FM males than in depressed patients. Despite a theoretical model supporting an equal approach between FM and comorbid psychiatric syndromes, such as depression or PTSD, the detection of psychiatric syndromes in FM patients led to a primacy of the 'psychiatric' manifestations, thus limiting the interest for systematic studies in this area. We believe that, despite all the disappointing findings of FM treatment, research should highlight the need for a better understanding of psychopathological features when comorbid with FM, raising questions on the usefulness of a combined systematic approach.

We could speculate that the search for

more effective forms of treatment for FM in males should begin with a closer examination of the factors that make FM specifically difficult to study and to treat. We believe that it would be helpful to refine more accurate definitions of FM phenotypes and their treatment resistance. It is unclear if a distinction can be made between such resistance and the natural history of the disease, at least in its current clinical manifestations with available treatments. As a consequence, a recommendation concerning the clinical challenge of improving both short- and long-term treatments of FM male patients is to draft therapeutic trials with instruments that are able to detect the complete range of clinical manifestations of this polymorphic condition.

In summary, even if most of the available studies used a qualitative narrative approach as methodological frame, we believe that we could highlight some take home messages from available data. First of all, reviewed studies showed how males with FM tended to perceive a future without well-being and subjective health in a more catastrophic way than females with FM. Masculine identity appears as inefficiently re-negotiated after the onset of FM symptoms, leading to an enduring condition of perceived inadequacy and to a loss of the psychosocial role, more severe than observed in females with FM. This finding seems to be related not only to the biological characteristics of males, but also to the introjections of cultural models/stereotypes that consider 'inappropriate for a real man' to be affected by a chronic 'psychosomatic' condition of 'uncertain aetiology' (84, 86, 87). The adoption of such stereotypes has a severe side effect: males with FM tend to avoid medical treatments and care, with a feeling of 'lost healthy self', deeply interfering with self-esteem, not even expressed, and not accepted. Therefore, males with FM often decide to endure pain for long periods before seeking for a treatment (102). Finally, males with FM tend to enhance the bodily symptom dimension, and to consider less relevant the exploration/expression of feelings about the illness and its con-

sequences, leading to a completely different approach to FM when compared with female patients.

We strongly believe that several aspects should be highlighted and faced in future research in this field. Here, some point that should be considered:

- Males with FM tend to show anxious and depressive symptoms having a significant impact on subjective perception of QoL and pain more than observed in females with FM;
- Sexual impairment in males with FM needs much more attention, because of its relevance with subjective well-being and self-esteem;
- Narrative approaches to FM in males may help the clinician with the development of 'tailored' communication strategies;
- There is paucity of long term follow up studies on pharmacological and non-pharmacological treatment options, which makes it impossible, at present, to report on specific and targeted therapeutic strategies for males with FM;
- There is a need for instruments focused on the assessment of physical, psychological, and psychosocial characteristics of males with FM. Future studies should offer better-matched interventions targeted to the specific features of this disorder in males.

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