Concomitant fibromyalgia in primary Sjögren's syndrome in the French ASSESS cohort: comparison of the ACR 1990 and ACR 2016 criteria, FiRST questionnaire and physician's opinion

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ABSTRACT

Objective. *Dryness, fatigue, and pain are classic symptoms in primary Sjögren's syndrome (pSS) but are also common in fibromyalgia (FM). We compared the characteristics of FM assessed by different criteria (American College of Rheumatology (ACR) 2016 and 1990 criteria), physician's opinion and Fibromyalgia Rapid Screening Tool (FiRST) questionnaire) in a cohort of patients with pSS.*

Methods. *Eight hospital departments tested 134 patients with pSS according to AECG criteria from the Assessment of Systemic Signs and Evolution in Sjögren's Syndrome (ASSESS) cohort.*

Results. *FM was present in 19%, 18%, 20%, and 29% of cases according to ACR 2016, ACR 1990 criteria, physician's opinion and the FiRST questionnaire, respectively. FM criteria-positive patients had higher EULAR SS Patient-Reported Index (ESSPRI) score, but not higher EULAR SS Disease Activity Index (ESSDAI) score. The objective measurements of dryness and the use of corticosteroids and immunosuppressive drugs did not differ between FM positive and negative patients. Regarding the ESSPRI dryness and fatigue subscale scores, depression and anxiety scores and the use of anxiolytics and antidepressants, the FiRST questionnaire exhibited a higher difference between positive and negative patients than ACR 2016 criteria. ACR 1990 and physician's opinion were somewhere in the middle. ACR 2016 exhibited moderate agreement with ACR 1990 (κ=0.52) and the physician's opinion (κ=0.60) and poor agreement with FiRST (κ=0.39).*

Conclusion. *The FM criteria identified pSS patients with higher ESSPRI scores but not higher ESSDAI systemic disease scores. Agreement between the different FM criteria was moderate, and the characteristics they described did not fully coincide.*

Introduction

Dryness of the mouth and eyes, fatigue, and joint pain are the triad of symptoms found in nearly 80% of patients with primary Sjögren's syndrome (pSS) (1), symptoms that are assessed using the European League Against Rheumatism (EULAR) SS Patient-Reported Index (ESSPRI). These symptoms are also common in fibromyalgia (FM), with one third of patients reporting dryness (2), making it difficult to discriminate between the symptoms of concomitant FM and pSS. FM is often associated with rheumatic and autoimmune diseases, such as rheumatoid arthritis (RA), spondyloarthritis (SpA), systemic lupus erythematosus (SLE) and systemic sclerosis (SSc) (3-5) and is observed in 12 to 31% of patients with pSS (5-10). FM is a factor in poor response to rheumatic treatment, particularly in RA and SpA (11, 12). Disease-modifying treatments are relatively ineffective in pSS, particularly regarding ESSPRI-defined dryness, fatigue and pain (1). It could be linked at least partly to an associated FM. Thus, as in SpA, it may be useful to consider concomitant FM when assessing the effectiveness of pSS treatment (12, 13).

Several criteria have been put forward to diagnose or classify FM. The American College of Rheumatology (ACR) 2016 criteria include fatigue, sleep disturbance, cognitive disorders, and somatic symptoms, and currently tend to replace the ACR 1990 criteria that mostly cen-

tred on tender points (2, 14). ACR 2016 criteria have been widely studied in primary FM but a good deal less in cases of co-occurrence with rheumatic diseases (14, 15). Because the 2016 criteria can be unwieldy in routine practice, other screening methods have been developed, such as the Fibromyalgia Rapid Screening Test (FiRST) questionnaire (16). This test is particularly useful because it is quick and easy and performs well in primary FM (16-20). Its performance has also been studied in FM concomitant with RA, SpA or systemic sclerosis, but not pSS (4, 21). The performance of the FM criteria may depend on the concurrent disease.

In the present study, we compared the characteristics of a subgroup of patients with concomitant FM and pSS from the French Assessment of Systemic Signs and Evolution in Sjögren's Syndrome (ASSESS) cohort based on the ACR 2016 and ACR 1990 criteria, FiRST questionnaire, and physician's clinical opinion.

Methods

Patients

ASSESS is a French prospective multicentre cohort. Fifteen internal medicine and rheumatology departments recruited 395 patients who met the American-European Consensus Group criteria for pSS between 2006 and 2009. These patients are prospectively followed for 20 years. Our study was approved by the institutional review board of Bichat Hospital, Paris, France, and the French Data Protection Authority. All participants gave written informed consent. Eight centres agreed to participate in our study.

Fulfilment of fibromyalgia criteria was assessed only once at the last follow-up visit in the ASSESS cohort between 2017 and 2019. Physicians were asked to state whether, they believed their patient had concomitant FM regardless of any classification criteria and met ACR 1990 criteria for that disease. The physicians then asked their patients to complete the FiRST and modified ACR 2010 questionnaires (22). The ACR 2016 criteria were used in the primary analysis of our study.

The following data were collected at inclusion in the ASSESS cohort: age, sex, time since pSS diagnosis, presence of anti-SS-A and anti-SS-B antibodies, Chisholm grade III or IV on salivary gland biopsy at diagnosis, Hospital Anxiety and Depression (HAD) scores, ESSPRI overall and subscale scores, Schirmer's test ≤5 mm/5 min, unstimulated whole salivary flow ≤0.1 mL/min and score on the EULAR SS Disease Activity Index (ESSDAI). At inclusion in the present study, we collected data on age, sex, time since diagnosis, ESSPRI overall and subscale scores, ESSDAI score, presence or history of lymphoma, erythrocyte sedimentation rate, C-reactive protein levels, gamma globulin levels, and use of corticosteroids, immunosuppressive medications, hydroxychloroquine, anxiolytics, and antidepressants. A significant change in ESSPRI between inclusion in ASSESS and inclusion in the study is defined as an increase or decrease ≥1 (23).

Statistical analyses

Statistical analyses were performed using Stata software, v. 15 (StataCorp, College Station, US). Continuous data were expressed as mean ± standard deviation or median [interquartile range] according to statistical distribution. Normality was assessed using the Shapiro-Wilk test. Independent groups were compared using chi-squared and Fisher's exact tests for categorical data and the Student's t-test or Mann-Whitney test for continuous variables. The homoscedasticity assumption was studied using Fisher-Snedecor's test. The tests were two-sided with α type I error set at 5%. As proposed by several authors (24, 25), we chose to report all individual *p*-values without applying any mathematical correction for the aforementioned tests comparing groups. Specific attention was given to the magnitude of differences and clinical relevance, particularly to compare relationships between the criteria for FM and symptoms. More precisely, the results were expressed using Hedges' effect-sizes (ES) and 95% confidence intervals, and were interpreted according to Cohen's recommendations (26) which defined ES bounds as small (ES: 0.2), medium (ES: 0.5) and large (ES: 0.8, "grossly perceptible and therefore large").

The agreement between FM criteria has been analysed, estimating the concordance rate (accuracy) and the Kappa agreement coefficient. The results have been studied in light of the usual rules defined in the literature (27, 28): 0–0.2 (negligible agreement), 0.2–0.4 (low/weak agreement), 0.4-0.6 (moderate agreement), 0.6–0.8 (substantial/good agreement) and >0.8 (excellent concordance). Sensitivity and specificity were estimated using 95% confidence intervals. A sensitivity analysis was carried out to guarantee the representativeness of our sample compared to the ASSESS study sample.

Results

Patient characteristics

The eight participating centres enrolled 134 of the 249 patients initially included in the ASSESS cohort. Compared to the 115 patients not included in the present study, our patients were younger at inclusion in the cohort (55.1±11.7 *vs*. 60.6±12.4 years; p=0.0005) and had slightly lower dryness scores (5.2±2.1 *vs*. 5.8±2.2; p=0.04). However, the groups were similar with regard to time since diagnosis, sex, ESSPRI overall score and fatigue and pain subscale scores, HAD anxiety and depression scores, prevalence of anti-SSA and anti-SSB antibodies or focus score ≥1, ESSDAI score, and prevalence of ESSDAI scores ≥5. The characteristics of our patients at inclusion in the FM substudy are given in Table I.

Prevalence of fibromyalgia and characteristics of patients with fibromyalgia according to the ACR 2016 criteria

FM was diagnosed in 24 of 126 patients (19.0%) based on the ACR 2016 criteria. Compared to the 102 patients who did not meet ACR 2016 criteria for FM, the 24 patients who did meet the criteria had significantly higher scores on the ESSPRI overall and pain subscale scores, but not on dryness and fatigue subscale scores, and a lower prevalence of anti-SSA antibodies. Conversely, the ESSDAI scores, the prevalence of Schirmer's test ≤5 mm/5 min and unstimulated salivary flow ≤0.1 mL/min on ASSESS inclusion and the propor-

Table I. Characteristics of the 134th patients at inclusion in the present study.

Age, (years)	65.1 ± 11.7
Sex, females	128 (95.5)
Time since diagnosis (years), median [IQR]	15.2 [13 ; 19]
ESSPRI	5.9 ± 2
Dryness	6.7 ± 2.3
Fatigue	6.4 ± 2.4
Pain	4.8 ± 2.8
ESSDAI	4.0 ± 4.8
ESSDAI ≥5	37 (29.4)
Lymphoma	12 (9.1)
Erythrocyte sedimentation rate (mm/h), median [IQR]	15 [9 ; 30]
C-reactive protein (mg/L)	4.8 ± 11.8
Gammaglobulin (g/L), median [IQR]	13.2 [9.9 ; 16.4]
Corticosteroids	22 (16.7)
Immunosuppressive drugs	19 (14.4)
Hydroxychloroquine	41 (31.1)
Antidepressants	17 (12.9)
Anxiolytics	17 (12.9)

Data are given as mean ± SD or n (%) unless otherwise noted.
SD, standard deviation; IQR, interquartile range; ESSPRI, EULAR Sjögren's Syndrome Patient-Reported Index; ESSDAI, EULAR Sjögren's Syndrome Disease Activity Index.

Table II. Comparison of the characteristics of patients who did and did not meet ACR 2016 criteria for fibromyalgia.

	ACR 2016 + n=24	ACR 2016 -n=102	*p*-value
Age (years)	64.0 ± 14.3	65.1 ± 11.5	0.7
Age upon diagnosis (years)	46.2 ± 13.2	49.1 ± 12.1	0.3
Time since diagnosis (years), median [IQR]	15.6 [13.2; 19.5]	15.1 [13; 18.7]	0.6
Females	22 (91.7)	99 (97.1)	0.2
ESSPRI	6.8 ± 1.4	5.8 ± 2.0	0.006
Dryness	7.2 ± 1.7	6.4 ± 2.4	0.2
Fatigue	7.0 ± 2.0	6.4 ± 2.3	0.2
Pain	6.3 ± 2.2	4.6 ± 2.8	0.001
Change in ESSPRI score ≥1 (increase or decrease)	10 (47.62)	58 (65.91)	0.12
ACR 2016 fatigue score			
- 0	0	9 (9.2)	
- 1	0	29 (29.6)	0.0001
- 2	13 (54.2)	40 (40.8)	
- 3	11 (45.8)	20 (20.4)	
Symptom severity score (SSS, 0-12)	8.5 ± 1 .6	5.2 ± 2.8	<0.0001
Widespread pain index (WPI, 0-19)	10 ± 2.5	4.0 ± 3.0	<0.0001
Fibromyalgia severity score (FS, 0-31)	18.5 ± 3.6	9.2 ± 4.9	<0.0001
Unstimulated salivary flow≤0.01 mL/min	9 (39.1)	56 (57.1)	0.1
Schirmer's test≤5 mm	13 (61.9)	65 (71.4)	0.4
ESSDAI	4.5 ± 4.8	3.9 ± 4.9	0.6
ESSDAI ≥ 5	8 (36.4)	27 (27.3)	0.4
Lymphoma	2 (8.3)	9 (8.8)	1
HAD anxiety on ASSESS inclusion	10.0 ± 3.8	9.7 ± 4.1	0.7
HAD depression on ASSESS inclusion	6.7 ± 4.5	5.7 ± 3.5	0.3
Anti-SSA	11 (45.8)	72 (70.6)	0.03
Anti-SSB	7 (29.2)	42 (41.2)	0.3
Focus ≥ 1	21 (91.3)	82 (92.1)	1
ESR (mm/h) (n=87), median [IQR]	17 [8; 19]	14.5 [9; 31]	0.5
C-reactive protein (mg/L) (n=108)	4.8 ± 10.6	4.5 ± 12.5	0.7
Gammaglobulin (g/L) (n=107), median [IQR]	10.9 [9.5; 16.0]	13.3 [10.9; 16.1]	0.2
Corticosteroids	1 (4.2)	20 (19.6)	0.07
Immunosuppressive drugs	4 (16.7)	15 (14.7)	0.7
Corticosteroids or immunosuppressive drugs	4 (16.7)	30 (29.4)	0.3
Hydroxychloroquine	11 (45.8)	29 (29.4)	0.1
Antidepressants	6 (25.0)	10 (9.8)	0.08
Anxiolytics	3 (12.5)	14 (13.9)	1
Antidepressants or anxiolytics	8 (33.3)	19 (18.6)	0.1

Data are given as mean ± SD or n (%) unless otherwise noted.
ACR: American College of Rheumatology; CI: confidence interval; SD: standard deviation; IQR: interquartile range; ESR:erythrocyte sedimentation rate; ESSPRI: EULAR Sjögren's Syndrome Patient-Reported Index; ESSDAI: EULAR Sjögren's Syndrome Disease Activity Index; HAD: Hospital Anxiety and Depression scale; ASSESS: Assessment of Systemic Signs and Evolution in Sjögren's Syndrome cohort; NS: not significant.

tion of patients receiving corticosteroids and immunosuppressive drugs did not differ between the two groups. HAD anxiety and depression scores on inclusion in the ASSESS cohort and use of anxiolytics and antidepressants were similar in the two groups (Table II).

Prevalence of fibromyalgia and characteristics of patients with fibromyalgia according to the FiRST questionnaire, the ACR 1990 criteria and the physician's opinion

FM was diagnosed in 37 of 129 patients (28.7%) based on the FiRST questionnaire, in 23 of 130 patients (17.7%) based on the ACR 1990 criteria and in 26 of 132 patients (19.7%) according to the physician's opinion. Data were available on all four criteria in 121 patients. Comparison of the characteristics of patients who did and did not have FM according to the physician's opinion, the FiRST questionnaire, ACR 1990 and ACR 2016 criteria are given in the Supplementary Tables S1-S4.

We found a large difference in ESSPRI score between patients who were positive and negative on the FiRST questionnaire (ES =0.98 [0.55; 1.39]), ACR 1990 criteria (ES=0.94 [0.46; 1.42]), and the physician's opinion (ES=0.79 [0.34; 1.23]), but the difference was only moderate on ACR 2016 criteria (ES=0.53 [0.07; 0.99]; Figure 1). The results for the ESSPRI pain subscale were similar, with the difference in scores being large with FiRST (ES=1.07 [0.64; 1.49]), ACR 1990 criteria (ES=1.14 [0.64; 1.62]), and the physician's opinion (ES=1.01 [0.55; 1.46]) but moderate with ACR 2016 criteria (ES=0.66 [0.20; 1.13]). Regarding the dryness subscale, the difference in scores was moderate with FiRST (ES=0.57 [0.16; 0.97]) and ACR 1990 criteria (ES=0.52 [0.06; 0.10]), but small with the physician's opinion (ES=0.31 [-0.13; 0.75]) and ACR 2016 criteria (ES=0.24 [-0.22; 0.69]). As regards the fatigue subscale, the difference in scores was moderate with FiRST (ES=0.73 [0.32; 1.14]), ACR 1990 criteria (ES=0.49 [0.03; 0.96]), and the physician's opinion (ES=0.47 [0.03; 0.90]), and small with ACR 2016 criteria (ES=0.28 [-0.17; 0.74]). Regardless

of the criteria considered, the difference between patients who were positive and negative was very small with the ESSDAI, Schirmer's test ≤5 mm/5 min, unstimulated whole salivary flow ≤0.1 mL/and small with anti-SSA and anti-SSB antibodies, erythrocyte sedimentation rate, C-reactive protein, gamma globulin levels, and use of corticosteroids, immunosuppressive medications, and hydroxychloroquine (Suppl. Tables S1-S4). For the HAD depression score at inclusion in the ASSESS cohort, the difference was large with ACR 1990 criteria (ES=0.92 [0.43; 1.41]), moderate with FiRST (ES=0.71 [0.28; 1.14]) and the physician's opinion (ES=0.69 [0.23; 1.14]), and small with ACR 2016 criteria (ES=0.28 [0.19; 0.75]). For the HAD anxiety score, the difference was moderate with FiRST (ES=0.51 [0.08; 0.93]), small with both ACR 1990 criteria (ES=0.30 [-0.18; 0.78]) and the physician's opinion (ES=0.29 [-0.16; 0.74]), and very small with ACR 2016 criteria (ES=0.07 [-0.40; 0.54]). The difference between patients who were positive and negative was also small with anxiolytics and antidepressants regardless of the criteria considered.

Agreement between fibromyalgia criteria

Forty-six (38%) patients had at least one positive criteria, 28 (23%) at least two, 23 (19%) at least three and 11 (9%) had all four positive criteria (Fig. 2). ACR 2016 criteria exhibited moderate agreement with ACR 1990 criteria (κ=0.52; concordance=85%) and the physician's opinion (κ=0.60; concordance=87%) and poor agreement with FiRST questionnaire (κ=0.39; concordance=77%). FiRST exhibited moderate agreement with ACR 1990 criteria (κ=0.55; concordance=83%) and the physician's opinion (κ=0.53; concordance=82%). Agreement was very good between ACR 1990 criteria and the physician's opinion (κ=0.84; concordance=95%). Table III shows the sensitivity and specificity of the four criteria depending on whether the gold standard was ACR 2016 or ACR 1990 criteria. All four criteria had good specificity but varying sensitivity. ACR 2016 criteria had the poorest sensitivity when ACR 1990 criteria was the gold standard. FiRST had the poorest specificity regardless of the gold standard.

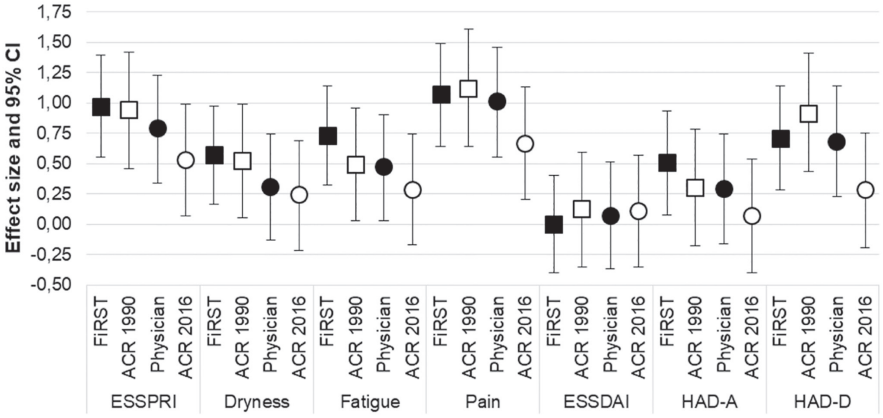

Fig. 1. Effect sizes of the FiRST questionnaire, ACR 1990 criteria, physician's opinion, and ACR 2016 criteria.
FiRST: Fibromyalgia Rapid Screening Tool; ACR: American College of Rheumatology; ESSPRI: EULAR Sjögren's Syndrome Patient-Reported Index; ESSDAI: EULAR Sjögren's Syndrome Disease Activity Index; HAD-A: Hospital Anxiety and Depression-Anxiety subscale; HAD-D: Hospital Anxiety and Depression-Depression subscale; CI: confidence interval.

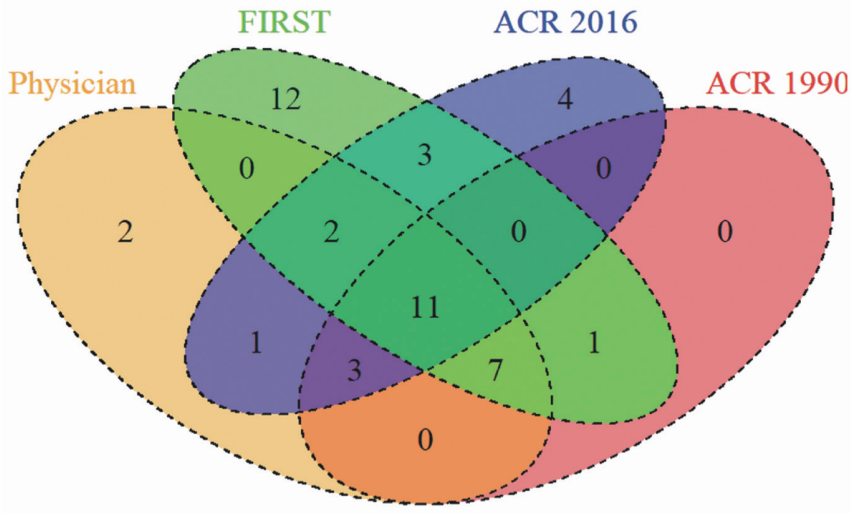

Fig. 2. Patients with at least one positive criteria for fibromyalgia (n= 46).
Overlap between the four criteria of fibromyalgia in the 121 patients: the American College of Rheumatology (ACR) 2016, the ACR 1990, the FiRST questionnaire and the physician.

Discussion

Our study of a multicentre cohort of patients with pSS shows that approximately 20% met the criteria for FM. FM criteria-positive patients had higher ESSPRI scores, but not higher or lower ESSDAI systemic disease scores. However, agreement between the criteria was moderate to poor, and the characteristics of the subgroups of patients identified by the criteria did not entirely overlap. These findings are in line with series in the literature, which have reported prevalence values of 12–31% (5-10). FM criteria-positive patients had higher ESSPRI scores, but their ESSDAI score did not differ from the score of those who were negative. Choi *et al*. reported similar findings, FM based on ACR 2010 criteria, was associated with higher ESSPRI and more severe depression but ESSDAI score did not differ significantly (8). A study conducted on the Spanish SJOGRENSER cohort, reported that patients with positive ACR 1990 criteria had higher ESSPRI score, higher dryness, fatigue and pain subscale scores, more constitutional symptoms but not higher ESSDAI score than patients who were negative (9). In our study, the difference in the ESSPRI subscale scores

Table III. Sensitivity and specificity of the FiRST questionnaire, ACR 1990 criteria, physician's opinion, and ACR 2016 criteria according to the gold standard used to define fibromyalgia.

	Gold standard: ACR 1990 criteria Sensitivity/specificity [95% CI]	Gold standard: ACR 2016 criteria Sensitivity/specificity [95% CI]
FiRST questionnaire	0.86 [0.65; 0.97] / 0.83 [0.74; 0.90]	0.67 [0.45; 0.84] / 0.79 [0.70; 0.87]
ACR 1990 criteria	- -	0.58 [0.37; 0.78] / 0.92 [0.84; 0.96]
Physician's opinion	0.96 [0.77; 1] / 0.95 [0.89; 0.98]	0.71 [0.49; 0.87] / 0.91 [0.83, 0.96]
ACR 2016 criteria	0.64 [0.41; 0.83] / 0.90 [0.82; 0.95]	- -

FiRST: Fibromyalgia Rapid Screening Tool; ACR: American College of Rheumatology.

between patients who did and did not have FM depended on the classification criteria used. As expected, the pain subscale was higher in the FM group regardless of the FM criteria. Regarding dryness and fatigue, the FiRST questionnaire exhibited a high difference between positive and negative patients, whereas the ACR 2016 criteria did not find any difference concerning these two items. The ACR 1990 criteria and physician's opinion were somewhere in the middle. Our findings were the same with respect to depression and anxiety scores and the use of anxiolytics and antidepressants. Anti-SSA antibodies were slightly less prevalent in positive patients based on ACR 1990 and 2016 criteria and on the physician's opinion, but not in those who were positive on the FiRST questionnaire. Objective measurements of dryness on ASSESS inclusion did not differ between positive and negative FM patients regardless of the FM criteria. ESSPRI score, which has been proven to be an essential tool for assessing pSS, was significantly affected by concomitant FM, although the effect varied depending on the FM criteria used. ACR 2016 criteria seems to focus more on pain and, thus is more specific to FM, whereas FiRST gives greater consideration to other symptoms, such as fatigue, dryness, anxiety, and depression. Unexpectedly, the ACR 1990 criteria lay somewhere in the middle, even though they are supposed to assess only pain.

In our study, the agreement between the FM criteria was moderate to poor. The ACR 2016 criteria exhibited moderate agreement with ACR 1990 criteria and had good specificity but poor sensitivity. In our study, the ACR 2016 criteria were filled in by the patient, and Wolfe *et al*. reported that discordance is possible between the physician-reported 2010 and patient-reported 2011 criteria (29). Results similar to ours have been observed based on ACR 2011 criteria in another study of concomitant FM in RA and systemic sclerosis. Specificity was 0.90 and sensitivity 0.50 and 0.56, respectively (4). On primary FM, comparable specificity was observed in a review of 14 studies, but median sensitivity was better (0.86) (14). The FiRST questionnaire had moderate agreement with ACR 1990 and the physician's opinion. In studies of primary FM that have used ACR 1990 criteria as the gold standard, the FiRST questionnaire has a reported sensitivity of 0.86 to 0.92 and specificity of 0.55 to 0.87 depending on how the control population was defined (16-20). In studies of concomitant FM, the agreement between FiRST and ACR 1990 criteria has been poor in RA (κ=0.25 and 0.28), SpA (κ=0.34 and 0.35), and systemic sclerosis (κ=0.51) (4, 21). Its specificity has been good (0.80 to 0.87), but its sensitivity has been reported to be poorer than in our study (0.44 to 0.73). In our study, the FiRST questionnaire exhibited surprisingly poor agreement with ACR 2016 criteria even though the 2016 criteria place particular emphasis on fatigue, feelings of depression, and cognitive difficulties, and do not include tender points. In RA and systemic sclerosis, Perrot *et al*. found that the FiRST questionnaire has similar sensitivity to ours (0.61 and 0.79) but better specificity (0.86 and 0.88) when their gold standard was the ACR 2011 criteria, which do not factor in generalised pain (4). The physician's opinion was highly concordant with the ACR 1990 criteria. That said, the physicians filled in the ACR 1990 criteria at the same time that they formed their opinion, whereas the FiRST questionnaire and ACR 2016 criteria were filled in by the patient without the physician knowing the results when they made their assessment. Thus, the physician's opinion and ACR 1990 criteria were not independent criteria, so bias is possible.

According to some authors, primary and concomitant FM diagnosed on the basis of ACR 1990 criteria or the Polysymptomatic Distress scale are the same condition (2, 30). Yet, the FM criteria seem to be more sensitive, specific, and concordant in primary FM than in concomitant FM in rheumatic diseases. Such discordance may arise because the rheumatic symptoms overlap with the FM symptoms, or because FM-like symptoms actually result from the rheumatic disease rather than from any concomitant FM. The symptoms of FM and pSS are very similar, and FM criteria have difficulty discriminating between them. One third of FM patients report dryness (2). The triad of dryness, fatigue and limb pain characterises the pSS but also non-immune mediated entities named dry eye and mouth (DEMS) or sicca asthenia polyalgia syndrome (SAPS) which are considered as FM equivalents (31, 32). The Fibromyalgia Severity score or Polysymptomatic Distress scale were found to be >9 in our ACR 2016 criteria-negative patients, but between 3 and 4 in the general population and 6.6 in patients with RA or osteoarthritis (14). Fatigue is very prevalent in patients with pSS but does not seem to result from concomitant FM (33, 34).

Our study's main limitation was the small sample size. This hampered our comparison of characteristics between patient groups based on FM criteria. The main advantage of this study was its multicentre nature, which limited data collection bias by ensuring that more investigators were involved.

Conclusion

Our study showed that FM classification criteria identified a subgroup of pSS patients with higher ESSPRI scores but without higher or lower ESSDAI systemic disease scores. FM should be considered when assessing pSS patients. However, the FM criteria did not exhibit good agreement, as FM and rheumatic symptoms may overlap.

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