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Comparing the Impact of Symptoms and Health Care Experiences of People Who Have and Have Not Received a Diagnosis of Fibromyalgia: A Cross-Sectional Survey Within the PACFiND Study

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Objective. To compare the impact of symptoms and health care utilization of people diagnosed with fibromyalgia, people who fulfill the criteria but are not diagnosed, and people with chronic pain.

Methods. We recruited people who had participated in a previous population survey across Scotland and who reported some typical fibromyalgia symptoms or had received a diagnosis of fibromyalgia. Responses to a postal questionnaire were used to define mutually exclusive groups: people who had a fibromyalgia diagnosis, who met criteria for fibromyalgia, and who had chronic pain.

Results. Participants included 85 people with a diagnosis of fibromyalgia, 110 who met criteria for fibromyalgia, and 133 with chronic pain. The mean age across groups ranged 57–59 years, but the percentage female varied markedly: 86%, 64%, and 67%, respectively. Compared to those with chronic pain, participants with a fibromyalgia diagnosis were more likely to be out of employment due to health. An average of 3 years was needed to receive a fibromyalgia diagnosis, and more than half were diagnosed in secondary care (most commonly rheumatology). The fibromyalgia diagnosis and criteria groups were similar in terms of symptom impact, quality of life, and life satisfaction but were worse than the chronic pain group. Participants who had received a diagnosis of fibromyalgia reported the poorest health care experiences.

Conclusion. An urgent need exists for a model of care for fibromyalgia to ensure prompt diagnosis, access to evidence-based care, and long-term support, with the aim of improving function. The data suggest that diagnosis in men may be overlooked, and this finding warrants further study.

INTRODUCTION

Fibromyalgia represents one end of a spectrum of symptoms and is characterized by pain that is both chronic and widespread throughout the body, as well as additional symptoms such as cognitive dysfunction, fatigue, headaches, abdominal cramps, or depression (1). In population samples, prevalence ranges from approximately 2% to 5%, depending on the precise criteria used and the population studied (2,3). Prevalence is even higher in people with inflammatory musculoskeletal conditions; for example, patients with rheumatoid arthritis, psoriatic arthritis, and ankylosing spondylitis have been shown in a meta-analysis to have prevalence proportions of 21%, 18%, and 13%, respectively (4). The diagnosis of fibromyalgia can be particularly challenging when the clinical features (such as pain, fatigue, and tenderness on palpation) are shared with the coexisting condition (5).

There are no objective tests for the diagnosis of the condition, and this gap has an important consequence; a considerable time can be needed for the diagnosis to be made. A study based in UK general practice showed that, as a group, people who were

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SIGNIFICANCE & INNOVATIONS

- People whose symptoms meet the criteria for fibromyalgia have symptoms whose impact is very similar to those who have received a fibromyalgia diagnosis.
- The impact of symptoms in people who meet the criteria for fibromyalgia (or who have received a diagnosis) is much greater than in people with chronic pain.
- The diagnosis of fibromyalgia may be overlooked in men.
- Particular attention needs to be paid for improving work outcomes in those with fibromyalgia symptoms/diagnosis.

given the diagnosis of fibromyalgia had more frequently consulted over the 10 years prior to diagnosis, in comparison to people without such a diagnosis (6). Our recent review could find no evidence-based model of care for people with fibromyalgia (7). Challenges that were noted as specific to fibromyalgia included the view by some clinicians that fibromyalgia does not represent a "real" condition, and a lack of a specialty or professional group responsible for care of the condition. Moreover, some services do not routinely accept referrals of people with suspected fibromyalgia or do so only on the basis of excluding other treatable causes (8). The UK general practice–based study mentioned above found that the number of consultations decreased slightly after diagnosis, but within 3–5 years were higher than the time just before diagnosis (6).

As part of a comprehensive program of work on care of people with fibromyalgia, the Patient-Centered Care for Fibromyalgia: New Pathway Design study, we aimed to compare the impact of symptoms and health care utilization of people who had received a diagnosis of fibromyalgia, of people who met the criteria for fibromyalgia but had not received such a diagnosis, and of people who reported chronic pain but did not meet criteria for, or had not received a diagnosis of, fibromyalgia.

MATERIALS AND METHODS

In 2015, we undertook a population screening survey in advance of the MAmMOTH clinical trial. This was a trial that aimed to recruit people without chronic widespread pain but who were at risk of its development through having a combination of regional pain, sleep disturbance, reporting multiple somatic symptoms, and/or certain illness behaviors. Details of the trial and recruitment procedures have previously been published (9). The MAmMOTH screening survey was conducted across 3 health boards in Scotland (NHS Grampian, NHS Highland, and NHS Greater Glasgow and Clyde), and there was a total of 18,035 respondents. For the current study we contacted people who had responded to the MAmMOTH screening survey and 1) had not been invited to take part in the MAmMOTH Trial, 2) had given consent to be contacted for future health studies, and 3) had a fibromyalgia symptom score of at least 12 (of 31) (1) or had reported a diagnosis of fibromyalgia. The fibromyalgia symptoms score is the sum of scores on the Widespread Pain Index (WPI) and symptom severity scale of the 2011 fibromyalgia research criteria. The combination of the WPI and symptom severity was shown to be best at discriminating criteria-positive and criteria-negative subjects in the study leading to the 2011 fibromyalgia research criteria, and factor analysis had also shown the combined score to represent a single dimension on factor analysis (10).

The study questionnaire collected sociodemographic characteristics and quality of life using the 5-level EuroQol 5-domain instrument (EQ-5D-5L) (11) and a single question on global life satisfaction: "Overall how satisfied are you with your life nowadays?" with 7 responses ranging from "completely dissatisfied" to "completely satisfied" (as used in the UK Household Longitudinal Study "Understanding Society"). The impact of symptoms was measured by the Symptom Impact Questionnaire (revised) (SIQR) (12); this instrument is identical to the Fibromyalgia Impact Questionnaire except that it uses the word "symptoms" instead of "fibromyalgia." This questionnaire was more suitable for this study, as not all participants had fibromyalgia symptoms, and even if they did so, had not necessarily been diagnosed with fibromyalgia. Information was collected about work status, and in relation to those undertaking paid work, about absenteeism, work productivity (referred to as "presenteeism"), and activity impairment using the Work Productivity and Activity Impairment–General Health questionnaire (WPAI-GH) (13). Health care usage information was collected using specially created questions, and information on patient experience through the Patient Experience Questionnaire, which includes 13 structured questions (14).

Two patient partners supported the development of the survey questionnaire (providing feedback on the use of specific instruments and their order in the questionnaire, recommending additional answer options and changes in wording, and improving readability). They also gave feedback on relevant participant documents such as invitation, reminder letters, and the information sheet.

Statistical analysis. Survey respondents were categorized into mutually exclusive groups: those reporting a diagnosis of fibromyalgia, those with no diagnosis of fibromyalgia but who met the 2011 criteria (1), and those who did not report a diagnosis of fibromyalgia and did not meet the 2011 criteria but who reported chronic pain (pain lasting at least 3 months). Respondents who did not fall into any of these 3 groups were excluded from the current analysis.

Descriptive information is provided for variables using median (interquartile range [IQR]) or mean ± SD, depending on the distribution of individual variables. Multinomial or logistic regression were used to examine each demographic variable in

relation to group membership (fibromyalgia diagnosis, fibromyalgia criteria, chronic pain) and the relationships reported as multinomial odds ratios (MORs) or ORs with 95% confidence intervals (95% CIs). As an example, examining the relationship between sex and group membership, male was designated as the reference group and the fibromyalgia diagnosis group as the base outcome. Adjusted analyses took into account age, sex, NHS region, and level of deprivation, the latter of which was based on postcode of residence (quintiles based on the Scottish Index of Multiple Deprivation 2020 [www.gov.scot]). A similar analytical approach was used to examine the relationship with other musculoskeletal diagnoses, global life satisfaction, and work-related variables. Poisson regression with robust error variance was used to quantify the relationship between group and time since first seeing a health care professional. Estimated marginal mean differences with 95% CIs are reported. Linear regression with robust error variance was used to quantify the relationship between group membership and SIQR, WPAI-GH, and EQ-5D-5L, and those results are reported as mean differences. Among participants who said they had been in contact with their general practice surgery in the past 3 months, the difference in responses to the Patient Experience Questionnaire between groups was examined using multinomial logistic regression. If few participants gave a particular answer, response categories were combined for the regression analysis where the degree of difference between the categories was qualitatively perceived to be small, such as "very helpful" and "extremely helpful."

RESULTS

From respondents to the MAmMOTH screening survey, 824 fulfilled the eligibility criteria for the current study and were sent a postal questionnaire. A total of 421 returned a completed questionnaire; of these, 85 reported that they had been diagnosed with fibromyalgia (the fibromyalgia diagnosis group), 110 met research criteria for fibromyalgia but reported that they had not received such a diagnosis (fibromyalgia criteria group), while 133 reported chronic pain but neither met research criteria for, nor had received a diagnosis of, fibromyalgia (chronic pain group). The fibromyalgia symptom scores of these groups were 17 (IQR 13–22), 16 (IQR 14–19), and 11 (IQR 9–12), respectively. In comparison to the fibromyalgia diagnosis group, in whom

Table 1. Sociodemographic characteristics of participants*

	Chronic pain		FM criteria		
	Value	MOR (95% CI)	Value	MOR (95% CI)	FM diagnosis
Sex					
Male	44 (33.1)	1 (Ref.)	40 (36.4)	1 (Ref.)	12 (14.1)
Female	89 (66.9)	0.33 (0.16, 0.68)	70 (63.6)	0.29 (0.14, 0.59)	73 (85.9)
Age, mean ± SD/MOR per 10 years	59.1 ± 13.8	1.12 (0.90, 1.41)	59.0 ± 10.6	1.12 (0.88, 1.41)	57.4 ± 11.4
NHS region					
Greater Glasgow and Clyde	43 (32.3)	1 (Ref.)	46 (41.8)	1 (Ref.)	27 (31.8)
Grampian	38 (28.6)	0.92 (0.46, 1.84)	29 (26.4)	0.65 (0.32, 1.33)	26 (30.6)
Highland	52 (39.1)	1.02 (0.53, 1.96)	35 (31.8)	0.64 (0.33, 1.26)	32 (37.7)
Current employment status					
Employed or self-employed	73 (54.9)	1 (Ref.)	38 (34.9)	1 (Ref.)	41 (48.2)
Retired	40 (30.1)	1.60 (0.78, 3.29)	28 (25.7)	2.16 (0.99, 4.70)	14 (16.5)
Not in paid employment, due to illness	15 (11.3)	0.30 (0.14, 0.63)	36 (33.0)	1.39 (0.72, 2.69)	28 (32.9)
Not in paid employment, not due to illness	5 (3.8)	1.40 (0.26, 7.56)	7 (6.4)	3.78 (0.74, 18.32)	2 (2.4)
Education (highest level)					
Secondary school	35 (26.5)	1 (Ref.)	34 (31.8)	1 (Ref.)	24 (28.2)
Apprenticeship	9 (6.8)	6.17 (0.73, 51.95)	5 (4.7)	3.52 (0.39, 32.16)	1 (1.2)
Further education or college	43 (32.6)	0.74 (0.38, 1.45)	47 (43.9)	0.83 (0.42, 1.62)	40 (47.1)
University degree	33 (25.0)	1.62 (0.72, 3.64)	17 (15.9)	0.86 (0.36, 2.07)	14 (16.5)
Further degree	12 (9.1)	1.37 (0.45, 4.16)	4 (3.7)	0.47 (0.12, 1.85)	6 (7.1)
Marital status					
Single	20 (15.2)	1 (Ref.)	23 (20.9)	1 (Ref.)	17 (20.2)
Married/living with partner	80 (60.6)	1.39 (0.66, 2.90)	49 (44.6)	0.74 (0.35, 1.55)	49 (58.3)
Widowed/divorced/separated	32 (24.2)	1.51 (0.64, 3.60)	38 (34.6)	1.56 (0.67, 3.62)	18 (21.4)
Area-based deprivation, quintiles					
1 (most deprived)	20 (15.0)	1.49 (0.60, 3.75)	28 (25.5)	3.25 (1.28, 8.25)	11 (12.9)
2	14 (10.5)	1.05 (0.40, 2.74)	16 (14.6)	1.86 (0.69, 4.98)	11 (12.9)
3	28 (21.1)	1 (Ref.)	18 (16.4)	1 (Ref.)	23 (27.1)
4	53 (39.9)	1.81 (0.87, 3.77)	30 (27.3)	1.60 (0.71, 3.62)	24 (28.2)
5 (least deprived)	18 (13.5)	0.92 (0.39, 2.21)	18 (16.4)	1.44 (0.58, 3.58)	16 (18.8)

* Values are the number (%) unless indicated otherwise. 95% CI = 95% confidence interval; FM = fibromyalgia; MOR = multinomial odds ratio; Ref. = reference.

Table 2. Impact on functional ability and quality of life across symptom/diagnosis groups*

	Chronic pain		FM criteria		
	Value	Mean difference/ MOR (95% CI)	Value	Mean difference/ MOR (95% CI)	FM diagnosis
Adjusted mean difference†					
SIQR, mean ± SD	36.9 ± 20.4	–20.67 (–26.61, –14.73)	57.4 ± 21.7	–2.16 (–8.43, 4.10)	57.2 ± 23.2
EQ-5D-5L, mean ± SD	0.63 ± 0.20	0.21 (0.13, 0.28)	0.39 ± 0.31	0.00 (–0.08, 0.07)	0.43 ± 0.28
Adjusted MOR, global life satisfaction†					
Completely dissatisfied	4 (3.0)	1.72 (0.24, 12.27)	9 (8.2)	2.71 (0.46, 16.12)	2 (2.4)
Mostly dissatisfied	11 (8.3)	1.02 (0.30, 3.49)	15 (13.6)	0.94 (0.30, 2.95)	10 (11.9)
Somewhat dissatisfied	12 (9.1)	0.52 (0.18, 1.54)	23 (20.9)	0.65 (0.25, 1.71)	24 (28.6)
Neither	13 (9.9)	1 (base)	19 (17.3)	1 (base)	13 (15.5)
Somewhat satisfied	30 (22.7)	3.37 (1.15, 0.90)	26 (23.6)	2.00 (0.70, 5.65)	11 (13.1)
Mostly satisfied	54 (40.9)	3.17 (1.21, 8.33)	16 (14.6)	0.67 (0.25, 1.82)	22 (26.2)
Completely satisfied	8 (6.1)	4.90 (0.82, 29.29)	2 (1.8)	0.83 (0.10, 6.91)	2 (2.4)

* Values are the number (%) unless indicated otherwise. 95% CI = 95% confidence interval; EQ-5D-5L = 5-level EuroQol 5-domain instrument; FM = fibromyalgia; MOR = multinomial odds ratio; SIQR = Symptom Impact Questionnaire (revised).
† Adjusted for age, sex, NHS region, and deprivation.

85.9% were female, significantly fewer members of other groups were female (fibromyalgia criteria group 63.6%; chronic pain group 66.9%) (Table 1). Age was similar across the 3 groups, with the mean varying between 57 and 59 years. There were few other important socioeconomic differences across groups, although notably, participants in the chronic pain group were significantly less likely to be out of employment due to their health than the fibromyalgia diagnosis group, with MOR 0.30 (95% CI 0.14, 0.63). The fibromyalgia diagnosis and criteria groups reported a time since first consulting a health professional with symptoms of 15 and 14 years, respectively, significantly longer than those with chronic pain (10 years; adjusted mean difference [md_{adj}] versus fibromyalgia diagnosis group –3.9 years [95% CI –7.4, –0.4]). The fibromyalgia diagnosis group reported that, on average, 3 years were needed to receive a diagnosis; 29 of 85 (34%) reported receiving a diagnosis in primary care, and 46 (54%) in secondary care, of which 22 were diagnosed by a rheumatologist, 7 by another clinical specialty, and 17 did not specify the specialty. Respondents were asked about other diagnoses: the fibromyalgia criteria group more commonly reported a diagnosis of rheumatoid arthritis (18%) or osteoporosis (19%) than either the fibromyalgia diagnosis group (12% and 13%, respectively) or chronic pain group (11% and 8%, respectively), although the differences were not statistically significant.

Impact of symptoms. The impact of symptoms on health status was similar in both fibromyalgia diagnosis and criteria groups (SIQR mean score 57.2 and 57.4, respectively), significantly more than in the chronic pain group (SIQR mean score 36.9, md_{adj} versus fibromyalgia criteria group –20.67 [95% CI –26.61, –14.73]) (Table 2). This difference in impact was also reflected in the poorer quality of life of the fibromyalgia groups

Table 3. Impact of symptoms on paid work across groups*

	Chronic pain		FM criteria		
	Value	OR_{adj}/coefficient (95% CI)	Value	OR_{adj}/coefficient (95% CI)	FM diagnosis
OR_{adj}†					
Stopped work due to symptoms	25 (18.8)	0.28 (0.15, 0.53)	43 (39.8)	0.77 (0.42, 1.41)	36 (42.3)
Stopped work due to					
Pain	20 (15.00)	0.28 (0.14, 0.56)	39 (35.5)	0.82 (0.44, 1.54)	30 (35.3)
Fatigue	18 (13.5)	0.21 (0.11, 0.42)	35 (31.8)	0.62 (0.33, 1.17)	34 (40.0)
Mental health/stress	8 (6.0)	0.26 (0.10, 0.65)	23 (20.9)	1.02 (0.47, 2.19)	16 (18.8)
Other	4 (3.0)	0.29 (0.08, 1.05)	11 (10.0)	1.10 (0.39, 3.09)	8 (9.4)
WPAI coefficient†					
Absenteeism, median (IQR) % (n = 117)	0 (0–0)	–5.75 (–17.43, 5.93)	0 (0–9.9)	–6.12 (–21.67, 9.44)	0 (0–19.0)
Presenteeism, median (IQR) % (n = 116)	30 (20–50)	–9.14 (–17.43, 5.93)	40 (30–70)	0.21 (–13.68, 14.10)	50 (30–60)
Activity impairment, median (IQR) % (n = 319)	40 (30–65)	–16.86 (–23.80, –9.91)	70 (60–80)	3.41 (–3.48, 10.31)	60 (40–80)

* Values are the number (%) unless indicated otherwise. 95% CI = 95% confidence interval; FM = fibromyalgia; IQR = interquartile range; OR = odds ratio; WPAI = Work Productivity and Activity Impairment.
† Adjusted for age, sex, NHS region, and deprivation.

(EQ-5D-5L mean score: fibromyalgia diagnosis group 0.43 and fibromyalgia criteria group 0.39) compared to the chronic pain group (0.63, md_{adj} versus fibromyalgia diagnosis group 0.21 [95% CI 0.13, 0.28]). There was no difference in reported global life satisfaction between the fibromyalgia groups, but those with chronic pain were considerably more likely to report one of the "satisfied" categories (fibromyalgia diagnosis 41,7%; fibromyalgia criteria 40.0%; chronic pain group 69.7%).

There was a substantial impact on paid work for people with a diagnosis of fibromyalgia or who met the criteria for fibromyalgia (Table 3); 42% and 40%, respectively, had stopped work due to their health as compared with 19% in the chronic pain group (OR_{adj} stopped work versus fibromyalgia diagnosis group 0.28 [95% CI 0.15, 0.53]). Those in the fibromyalgia diagnosis group who had stopped work due to their health had last undertaken work a median of >10 years previously (131 months [IQR 26–192]), and in the fibromyalgia criteria group the median was over 8 years (99 months [IQR 35–241]). The symptoms of pain, fatigue, and mental health/stress were commonly mentioned by people in the fibromyalgia criteria and fibromyalgia diagnosis groups as contributing to stopping work. Among those still working and over all of the groups, absenteeism was relatively uncommon. The median percentage of time lost in the past 7 days was 0% (IQR 0–1), but with a mean of 11%, reflecting the fact that a small proportion of respondents were recording high rates of absenteeism, while the median level of presenteeism (when working) was 30% (IQR 20–50). Results from linear regression showed no consistent difference between the fibromyalgia criteria and

Table 4. Participants' reported health care experiences with primary care in the past 3 months*

	Chronic pain		FM criteria		
	Value	MOR_{adj} (95% CI)†	Value	MOR_{adj} (95% CI)	FM diagnosis
How easy did you find it to get the care you needed when you wanted it?					
Very easy	16 (21.6)	1.07 (0.39, 2.93)	13 (17.6)	0.60 (0.23, 1.60)	7 (14.0)
Fairly easy	36 (48.7)	–	26 (35.1)	–	27 (54.0)
Not very easy	14 (18.9)	1 (base)	18 (24.3)	1 (base)	10 (20.0)
Difficult	5 (6.8)	0.92 (0.22, 3.89)	9 (12.2)	1.33 (0.36, 4.97)	5 (10.0)
Very difficult	3 (4.1)	–	8 (10.8)	–	1 (2.0)
Were you given clear and understandable information about symptoms and care?					
Yes, definitely	3 (43.2)	1.68 (0.73, 3.88)	22 (30.1)	1.04 (0.44, 2.47)	14 (28.0)
Yes, to some extent	34 (45.9)	1 (base)	43 (58.9)	1 (base)	39 (58.0)
No	8 (10.8)	–	8 (11.0)	–	7 (14.0)
Were you involved as much as you wanted in decisions about symptoms and care?					
Yes, definitely	41 (55.4)	1.31 (0.61, 2.85)	32 (43.2)	0.87 (0.40, 1.89)	22 (44.9)
Yes, to some extent	26 (35.1)	1 (base)	38 (51.4)	1 (base)	19 (38.8)
No	7 (9.5)	–	4 (5.4)	–	8 (16.3)
Were you given the opportunity to talk about worries and fears with regard to symptoms?					
Yes, definitely	41 (55.4)	2.18 (0.79, 6.06)	27 (36.5)	2.46 (0.79, 7.63)	18 (36.0)
Yes, to some extent	17 (23.0)	1.20 (0.41, 3.51)	37 (50.0)	3.91 (0.41, 3.51)	18 (36.0)
No	16 (21.6)	1 (base)	10 (13.5)	1 (base)	11 (22.0)
Had no worries or fears	0 (0.0)	–	0 (0.0)	–	3 (6.0)
Did the different people treating and caring for you work well together to give you the best possible care?					
Yes, always	37 (50.0)	1.96 (0.84, 4.58)	27 (36.5)	1.16 (0.49, 2.74)	15 (30.0)
Yes, sometimes	23 (31.1)	1 (base)	31 (41.9)	1 (base)	23 (46.0)
No, never	6 (8.1)	–	8 (10.8)	–	4 (8.0)
Can't remember	1 (1.4)	0.99 (0.29, 3.41)	2 (2.7)	0.81 (0.23, 2.83)	3 (6.0)
Not relevant	7 (9.5)	–	6 (8.1)	–	5 (10.0)

(Continued)

Table 4. (*Cont'd*)

	Chronic pain		FM criteria		
	Value	MOR_{adj} (95% CI)†	Value	MOR_{adj} (95% CI)	FM diagnosis
Did health care staff offer family, carers, friends opportunity to be involved in decisions about symptoms and care?					
Yes, always	12 (16.9)	1.67 (0.51, 5.47)	6 (8.6)	2.94 (0.93, 9.27)	2 (4.1)
Yes, sometimes	5 (7.0)	–	16 (22.9)	–	4 (8.2)
No, never	28 (39.4)	1 (base)	25 (35.7)	1 (base)	20 (40.8)
No family etc. available	11 (15.5)	1.19 (0.37, 3.78)	12 (17.1)	1.34 (0.42, 4.24)	8 (16.3)
Didn't want involved	15 (21.1)	0.91 (0.34, 2.46)	11 (15.7)	0.87 (0.30, 2.53)	15 (30.6)
Did health care staff respond to your individual needs?					
Yes, at all times	34 (47.2)	2.03 (0.78, 5.27)	19 (26.0)	0.87 (0.33, 2.30)	15 (30.0)
Yes, most of the time	20 (27.8)	1.37 (0.51, 3.65)	28 (38.4)	1.43 (0.57, 3.60)	16 (32.0)
Yes, some of the time	13 (18.1)	1 (base)	18 (24.7)	1 (base)	16 (32.0)
No, never	5 (6.9)	–	8 (11.0)	–	3 (6.0)
Did health care staff do everything they could to make you feel physically comfortable?					
Yes, at all times	35 (48.6)	2.93 (1.02, 8.43)	30 (41.1)	1.90 (0.68, 5.28)	16 (32.0)
Yes, most of the time	26 (36.1)	2.10 (0.74, 6.01)	27 (37.0)	1.50 (0.55, 4.10)	18 (36.0)
Yes, some of the time	6 (8.3)	1 (base)	11 (15.1)	1 (base)	9 (18.0)
No, never	5 (6.9)	–	5 (6.8)	–	7 (14.0)
Did you have confidence and trust in health care staff?					
Yes, always	41 (55.4)	2.02 (0.91, 4.46)	34 (47.2)	1.53 (0.69, 3.38)	18 (36.0)
Yes, sometimes	28 (37.8)	1 (base)	29 (40.3)	1 (base)	24 (48.0)
No	5 (6.8)	–	9 (12.5)	–	8 (16.0)
How helpful has your care been in dealing with the problems you sought help for?					
Extremely helpful	13 (17.8)	1.92 (0.74, 4.97)	13 (17.8)	1.24 (0.48, 3.19)	4 (8.0)
Very helpful	25 (34.3)	–	18 (24.7)	–	10 (20.0)
Helpful	19 (26.0)	1 (base)	23 (31.5)	1 (base)	17 (34.0)
A little helpful	11 (15.1)	0.64 (0.23, 1.80)	14 (19.2)	0.53 (0.20, 1.45)	16 (32.0)
Not at all helpful	5 (6.9)	–	5 (6.9)	–	3 (6.0)
Have health services helped you to better understand and manage your health?					
Yes, definitely	22 (29.7)	1.36 (0.55, 3.35)	17 (23.3)	2.61 (0.98, 6.97)	5 (10.0)
Yes, to some extent	36 (48.7)	–	45 (61.6)	–	31 (62.0)
No	16 (21.6)	1 (base)	11 (15.1)	1 (base)	14 (28.0)
Overall, were you treated with kindness and understanding?					
Yes, always	55 (75.3)	1.32 (0.57, 3.07)	52 (71.2)	1.22 (0.53, 2.80)	35 (67.3)
Yes, sometimes	17 (23.3)	1 (base)	19 (26.0)	1 (base)	17 (32.7)
No	1 (1.4)	–	2 (2.7)	–	0 (0.0)
Overall, did health care staff keep you informed what would happen next?					
Yes, completely	35 (47.3)	6.13 (1.65, 22.76)	31 (42.5)	2.18 (0.73, 6.53)	16 (31.4)
Yes, to some extent	35 (47.3)	5.07 (1.40, 18.35)	32 (43.8)	1.93 (0.67, 5.61)	21 (41.2)
No	4 (5.4)	1 (base)	10 (13.7)	1 (base)	14 (27.5)

* Values are the number (%) unless indicated otherwise. 95% CI = 95% confidence interval; FM = fibromyalgia; MOR_{adj} = adjusted multinomial odds ratio.
† Adjusted for age, sex, NHS region, and deprivation.

diagnosis groups, but there was less impact among people with chronic pain across absenteeism, presenteeism, and difference in activity impairment, the latter of which was statistically significant (versus fibromyalgia diagnosis group coefficient –16.86 [95% CI –23.80, –9.91]).

Health care consultations and experiences. Respondents in the fibromyalgia criteria group were most likely to report a primary care or hospital consultation in the previous 3 months (67% and 48%, respectively) followed by the fibromyalgia diagnosis group (61% and 42%, respectively), and then the chronic pain group (57% and 34%, respectively), although the differences between groups were not statistically significant. Admission to hospital in the past 3 months was uncommon: 6%, 2%, and 4%, respectively. When asked about their experiences of health care, people in the fibromyalgia diagnosis group reported fewer positive experiences compared with the chronic pain group, with the fibromyalgia criteria group usually reporting levels of positive experience between these 2 groups (Table 4).

People who had received a diagnosis of fibromyalgia were significantly less likely, in comparison to those in the chronic pain group, to endorse the idea that "health care staff had done everything possible to make [them] feel physically comfortable" (fibromyalgia diagnosis 32%, fibromyalgia criteria 41%, chronic pain 49%) and "that [they] were kept informed about what would happen next in their health care" (31%, 43%, 47%, respectively). They were also less likely to agree with the statement "that (they) definitely had confidence and trust in health care staff" (36%, 47%, 55%, respectively) and "that [their] care was very or extremely helpful in dealing with the problems [they] sought help for" (28%, 43%, 52%, respectively). Both the fibromyalgia diagnosis group (28%) and fibromyalgia criteria group (30%) were less likely to agree with the statement "that [they] had been given clear and understandable information about [their] symptoms and care" compared with the chronic pain group (43%).

DISCUSSION

In this population survey, we found that people whose symptoms met the criteria for fibromyalgia, but who had not received a diagnosis, had symptoms with an impact (including on work) that was very similar to that in people who had received such a diagnosis. In both cases, those with and without a diagnosis who fulfilled the criteria had symptoms that were having a greater impact than were the symptoms of people with chronic pain. People with a diagnosis of fibromyalgia reported more negative experiences of health care.

There are some methodologic issues to consider in interpreting the results. First, while the sampling frame was population-based for the original MAmMOTH screening survey, people were only invited to take part in the current study if they had responded to the MAmMOTH screening questionnaire and had indicated a willingness for future contact. Further, to be eligible for the current study, participants needed to have told us either that they had received a diagnosis of fibromyalgia or that they had a fibromyalgia symptom score of at least 12 (of a maximum of 31). This score is relatively high, so that those people in the chronic pain group who participated in the study were at the most severe end of chronic pain and may have displayed at least some features typical of people with fibromyalgia. As a result, the chronic pain group here is less typical of the broader group of chronic pain patients and potentially less different from the fibromyalgia groups. Therefore, the differences we have seen between the chronic pain and both fibromyalgia groups in this study are very likely to be an underestimate of the true differences.

Second, while the fibromyalgia criteria group met research criteria and reported that they had not received this diagnosis, they may well have had fibromyalgia. People in the fibromyalgia criteria group were more likely to report other musculoskeletal diagnoses, and the criteria for fibromyalgia are valid even in the context of another diagnosis (1). Certainly, the similarity was striking between the fibromyalgia criteria and the fibromyalgia diagnosis groups across the domains studied. Third, important health care experiences (beyond patient satisfaction and patient-reported outcomes) and unmet needs/service gaps were quantified. To the best of our knowledge, this is the first quantitative survey using a patient-reported experience measure in relation to fibromyalgia/chronic pain.

From an epidemiologic perspective, fibromyalgia lies at one end of the pain spectrum in terms of the role of etiologic factors. While both physical trauma and psychosocial factors are important, the latter are relatively more important, for example, than for pain that is more localized and has been present for a shorter time (15). The data on symptom impact and health service–related factors also suggest that patients with fibromyalgia lie at one end of a spectrum. While the concept of primary and secondary fibromyalgia is no longer considered appropriate, physicians increasingly recognize that fibromyalgia is commonly comorbid with a range of other musculoskeletal conditions (4,16) and that management of, for example, the comorbid inflammatory condition is not sufficient to manage fibromyalgia symptoms (17). This study provides evidence that the impact of fibromyalgia symptoms is similar in those patients who meet the criteria, whether they have been diagnosed or not. Indeed the coexistence of another condition may make a diagnosis of fibromyalgia less likely and therefore presumably also decrease appropriate management for fibromyalgia (18).

One aspect in which people with a diagnosis and those who met the criteria differed markedly was with respect to sex. Most clinical studies of fibromyalgia report a very high ratio of female: male cases, often of the order of 10:1, but population studies show a much lower ratio, and specifically with more recent criteria the ratio is close to 1 (2). This higher ratio is explained partly by female patients reporting a greater likelihood of consulting but is also likely due to the perception of fibromyalgia as a

predominantly female disease and thus possible reticence to make the diagnosis in men (19). Wolfe et al (2) have previously highlighted this issue, which they concluded was partly related to biased sampling in some studies, and specifically noted a much lower sex ratio in studies using unbiased sampling strategies. This important issue needs further study to clarify the reasons for the discrepancy and thus allow diagnosis and appropriate management for everyone who needs it.

The information on health and care provides important information in terms of areas of unmet need. First, there is the greater impact of fibromyalgia on work, and the important role of pain, fatigue, and mental health in having to give up work. A meta-analysis of 2,757 people in 4 trials of pregabalin in fibromyalgia demonstrated that a reduction in days lost from work was importantly related to improvement in pain (20), while in a study of 301 patients with fibromyalgia in clinics throughout Spain, high levels of fatigue were an independent predictor of temporary work disability (21). Patients with musculoskeletal conditions have highlighted a lack of support from their rheumatologist in enabling them to remain in work (22). Putting in place services to enable greater work stability needs to be a priority, and promising approaches are being evaluated (23). Further effective approaches to management of fibromyalgia, such as exercise and cognitive behavior therapy, have specifically been shown to be beneficial for the symptoms that patients identify as the barriers to remaining in work (24,25). However, many people with fibromyalgia probably do not have access to such therapies.

Previously, we reported that people with fibromyalgia described that, after diagnosis, there were no clear "next steps" (7). The current study confirms this finding: approximately 1 in 4 people who had received a diagnosis of fibromyalgia reported that health care staff had not informed them about what would happen next in their health care. Despite a care pathway having been proposed for chronic widespread pain (including fibromyalgia) as a result of work undertaken by the British Pain Society, there has been no meaningful uptake of this pathway (26). Both the management guidelines and the proposed care pathway acknowledge the key role of primary care, and thus consideration as to the facilitators and barriers for effecting such a change needs to be a key issue. This consideration could include increasing awareness of what exists (diagnostic criteria and management recommendations, including evidence-based therapies) and providing ongoing support (such as sharing information and planning care for fibromyalgia) among primary care staff. The Royal College of General Practitioners in the UK has a wide range of toolkits for conditions and scenarios but not for fibromyalgia (Clinical Toolkits: rcgp.org.uk). In a study that included 809 primary care physicians from 8 countries across Europe (including the UK), the Americas, and Asia, 60% stated that making the diagnosis was somewhat or very difficult, and just over half were not aware of the American College of Rheumatology classification criteria (27).

In summary, this study highlights the importance of developing models of care for people with fibromyalgia to ensure a prompt diagnosis and access to evidence-based approaches that have been demonstrated to improve outcomes, while ensuring ongoing support. Although certain components of a model of care exist, consideration needs to be given about how these are integrated, identifying gaps and prioritizing activities to ensure that any proposed model of care is taken up. Specifically, this work has highlighted the fact that the impact of fibromyalgia symptoms is very similar between those with a diagnosis and those without a diagnosis but who meet the criteria, that there needs to be awareness of the condition in men, and in terms of management, that a focus needs to be made on additional outcomes, such as work, to facilitate people who wish to do so remaining in employment.

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AUTHOR CONTRIBUTIONS

All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors approved the final version to be submitted for publication. Dr. Macfarlane had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study conception and design. Doebl, Hollick, Beasley, Choy, Macfarlane.
Acquisition of data. Doebl, Beasley.
Analysis and interpretation of data. Beasley, Macfarlane.

REFERENCES

1. Wolfe F, Clauw DJ, Fitzcharles MA, Goldenberg DL, Häuser W, Katz RS, et al. Fibromyalgia criteria and severity scales for clinical and epidemiological studies: a modification of the ACR preliminary diagnostic criteria for fibromyalgia. J Rheumatol 2011;38:1113–22.
2. Wolfe F, Walitt B, Perrot S, Rasker JJ, Häuser W. Fibromyalgia diagnosis and biased assessment: sex, prevalence and bias. PLoS One 2018;13:e0203755.
3. Jones GT, Atzeni F, Beasley M, Flüß E, Sarzi-Puttini P, Macfarlane GJ. The prevalence of fibromyalgia in the general population: a comparison of the American College of Rheumatology 1990, 2010, and modified 2010 classification criteria. Arthritis Rheumatol 2015;67:568–75.
4. Duffield SJ, Miller N, Zhao S, Goodson NJ. Concomitant fibromyalgia complicating chronic inflammatory arthritis: a systematic review and meta- analysis. Rheumatology (Oxford) 2018;57:1453–60.
5. Macfarlane GJ, Pathan E, Siebert S, Packham J, Gaffney K, Choy E, et al. AxSpA patients who also meet criteria for fibromyalgia: identifying distinct patient clusters using data from a UK national register (BSRBR-AS). BMC Rheumatol 2019;3:19.
6. Hughes G, Martinez C, Myon E, Taïeb C, Wessely S. The impact of a diagnosis of fibromyalgia on health care resource use by primary care patients in the UK: an observational study based on clinical practice. Arthritis Rheum 2006;54:177–83.

7. Doebl S, Macfarlane GJ, Hollick RJ. "No one wants to look after the fibro patient." Understanding models, and patient perspectives, of care for fibromyalgia: reviews of current evidence. Pain 2020;161:1716–25.
8. Shir Y, Fitzcharles MA. Should rheumatologists retain ownership of fibromyalgia? J Rheumatol 2009;36:667–70.
9. Macfarlane GJ, Beasley M, Scott NW, Chong H, McNamee P, McBeth J, et al. Maintaining musculoskeletal health using a behavioural therapy approach: a population-based randomised controlled trial (The MAmMOTH study). Ann Rheum Dis 2021;80:903–11.
10. Wolfe F. New American College of Rheumatology criteria for fibromyalgia: a twenty-year journey. Arthritis Care Res (Hoboken) 2010;62: 583–4.
11. Herdman M, Gudex C, Lloyd A, Janssen M, Kind P, Parkin D, et al. Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). Qual Life Res 2011;20:1727–36.
12. Bennett RM, Friend R, Jones KD, Ward R, Han BK, Ross RL. The Revised Fibromyalgia Impact Questionnaire (FIQR): validation and psychometric properties. Arthritis Res Ther 2009;11:R120.
13. Reilly MC, Zbrozek AS, Dukes EM. The validity and reproducibility of a work productivity and activity impairment instrument. Pharmacoeconomics 1993;4:353–65.
14. Fitzpatrick R, Graham C, Gibbons E, King J, Flott K, Jenkinson C. Development of new models for collection and use of patient experience information in the NHS–PRP 070/0074. Picker Institute Europe and University of Oxford. 2014.
15. Clauw DJ. Fibromyalgia: a clinical review. JAMA 2014;311:1547–55.
16. Atzeni F, Cazzola M, Benucci M, Di Franco M, Salaffi F, Sarzi-Puttini P. Chronic widespread pain in the spectrum of rheumatological diseases. Best Pract Res Clin Rheumatol 2011;25:165–71.
17. Macfarlane GJ, MacDonald RI, Pathan E, Siebert S, Gaffney K, Choy E, et al. Influence of co-morbid fibromyalgia on disease activity measures and response to tumour necrosis factor inhibitors in axial spondyloarthritis: results from a UK national register. Rheumatology (Oxford) 2018;57:1982–90.
18. Fitzcharles MA, Perrot S, Häuser W. Comorbid fibromyalgia: a qualitative review of prevalence and importance. Eur J Pain 2018;22:1565–76.
19. Katz JD, Mamyrova G, Guzhva O, Furmark L. Gender bias in diagnosing fibromyalgia. Gend Med 2010;7:19–27.
20. Straube S, Moore RA, Paine J, Derry S, Phillips CJ, Hallier E, et al. Interference with work in fibromyalgia: effect of treatment with pregabalin and relation to pain response. BMC Musculoskelet Disord 2011;12:125.
21. Rivera J, Esteve-Vives J, Vallejo MA, Rejas J, Grupo ICAF. Factors associated with temporary work disability in patients with fibromyalgia. Rheumatol Int 2011;31:1471–7.
22. Hollick RJ, Stelfox K, Dean LE, Shim J, Walker-Bone K, Macfarlane GJ. Outcomes and treatment responses, including work productivity, among people with axial spondyloarthritis living in urban and rural areas: a mixed-methods study within a national register. Ann Rheum Dis 2020;79:1055–62.
23. Carruthers EC, Rogers P, Backman CL, Goldsmith CH, Gignac MA, Marra C, et al. "Employment and arthritis: making it work": a randomized controlled trial evaluating an online program to help people with inflammatory arthritis maintain employment (study protocol). BMC Med Inform Decis Mak 2014;14:59.
24. Bernardy K, Klose P, Busch AJ, Choy EH, Häuser W. Cognitive behavioural therapies for fibromyalgia. Cochrane Database Syst Rev 2013; 2013:CD009796.
25. Sosa-Reina MD, Nunez-Nagy S, Gallego-Izquierdo T, Pecos-Martín D, Monserrat J, Álvarez-Mon M. Effectiveness of therapeutic exercise in fibromyalgia syndrome: a systematic review and meta-analysis of randomized clinical trials. Biomed Res Int 2017;2017:2356346.
26. Lee J, Ellis B, Price C, Baranowski AP. Chronic widespread pain, including fibromyalgia: a pathway for care developed by the British Pain Society. Br J Anaesth 2014;112:16–24.
27. Choy E, Perrot S, Leon T, Kaplan J, Petersel D, Ginovker A, et al. A patient survey of the impact of fibromyalgia and the journey to diagnosis. BMC Health Serv Res 2010;10:102.

APPENDIX A: PACFiND STUDY INVESTIGATORS

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