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How do women with CFS/ME rate quality and coordination of health care services? A cross-sectional study

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Quality of care, chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), general practice, specialist care, coordination of care, Norway.

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ABSTRACT

Objective: To test the association between self-rated health and self-rated degree of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), and CFS/ME patients' assessment of quality of primary care, specialist care, and coordination of care.

Design: Cross-sectional study.

Setting: Self-reported questionnaire data from women members of The Norwegian ME Association obtained in 2013.

Participants: 431 women with CFS/ME aged 16-73 years.

Main outcome measure: The participants' assessment of quality in primary care, specialist care, and in coordination of care (good/very good or poor/very poor). Main explanatory variables: self-rated health and self-rated degree of CFS/ME.

Results: Quality of care was rated poor/very poor by 60.6% in primary care, by 47.7% in specialist care, and by 71.2% regarding coordination of care. Poorer self-rated health increased the probability of rating quality poor/very poor in primary care (odds ratio [OR] 1.81, 95% confidence interval [CI] 1.36-2.41), and in specialist care (OR 1.38, CI 1.05-1.82), but not in coordination of care. The probability of reporting quality in primary care poor/very poor decreased with increasing number of GP visits during the previous year. Similar associations were observed for primary care in models where self-rated health was replaced by self-rated degree of CSF/ME. Those who had the same GP for three years or more were less likely to report primary care quality as poor/very poor (OR 0.61, CI 0.38-0.93).

Conclusions: A large proportion of women with CFS/ME rated quality of care poor/very poor in primary care, specialist care, and in coordination of care. The dissatisfaction was higher for primary care than for specialist care. Poorer self-rated health and a more severe CFS/ME were associated with lower quality scores in primary and specialist care, but not in coordination of care. Health care services, as assessed by patients with CSF/ME, do have a large potential for improvement.

STRENGHTS AND LIMITATIONS OF THIS STUDY

- Recruitment of a homogenous group of study participants from a patient organization is a study strength
- Use of a systematically tested questionnaire and an acceptable response rate is a strength
- There might be a selection bias due to recruitment form a patient organization, as well as a possible recall bias as for most questionnaire data, which are limitations of the study
- Validity and reliability of self-reported measures might be discussed, and in particular measures of disease severity and health
- The cross-sectional design precludes any causal interpretation

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INTRODUCTION

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), is a condition with an estimated prevalence around 1-2 per thousand [1]. It affects women more than men (70-85%), and etiology is unknown [2-4]. CFS/ME is characterized by its fluctuating nature, physical and mental fatigue, persistent post-exertional malaise, sleep disturbances, problems with concentration and memory, pain in muscles and joints, headache, and other symptoms related to cognitive, immune, and autonomous dysfunctions [5-7]. The terms CFS and ME are used interchangeably but mostly as a composite term, and the distinction between them is disputed [5]. In this paper we use the combined term CFS/ME as recommended by Norwegian health authorities and others [7, 8].

CFS/ME cannot be confirmed by specific tests, it is not localized to a specific part of the body, and it challenges the traditional distinction between psyche and soma [9]. In line with this, CFS/ME is a controversial condition and poses particular challenges in terms of diagnosis, therapy, and communication, to patients as well as to physicians [10].

CFS/ME patients often need coordinated health services from multiple providers over time. General practitioners (GPs) are their main contacts. All Norwegian citizens are provided a regular GP, and only 0,4 % of the population has chosen to remain outside GPs' lists [11]. Residents can change GP twice a year without justification. Together with universal tax-funding and gate-keeping, the list system provides strong incentives for personal continuity of care [12]. First line medical services including emergency clinics are run by the municipalities. Specialist services, consisting of hospitals and outpatient clinics, are run by regional health enterprises and are mainly owned by the state. Access is usually achieved by referrals from the GP (the gate-keeper role). GP and specialist outpatient visits are co-payed by adult patients.

Health care services, and GPs in particular, are well regarded in the population [13]. However, Norway's scores in the annual patient/population assessed international comparisons of health systems by the Commonwealth Fund (CWF) have been less than average in areas like general quality of care, waiting time for appointments, information sharing, communication, and coordination of services [14, 15]. Patient satisfaction, defined as "an individual's cognitive evaluation of, and emotional reaction to, his or her health-care experience" [16], is considered an important indication of overall quality in health care [17]. The Norwegian scores, thereby, indicate particular challenge areas for health care providers and policy makers.

Across health care settings, patient satisfaction or patients' assessment of health care quality is positively associated with better self-rated health and functional status, and inversely associated with the complexity of health problems [18-21]. In striving towards evidence based improvement of health care delivery, research on patient-based assessments of quality and coordination is informative, especially for groups with chronic diseases and long-

term needs of care from multiple providers, like CFS/ME. Moreover, this group of patients is interesting due to previously reported serious challenges in collaboration with health care services and providers, in line with the contested status of the condition [9].

The familiar measure self-rated health captures a comprehensive range of aspects and provides summative information about the individual's history of health and disease as well as the current level of health [22, 23]. However, a measure related to the disease of interest might provide a more specific indication of how disease severity and patients' ratings might be associated. Self-rated degree of CFS/ME might be such a measure. Solid evidence whether differences in health status might be associated with CFS/ME patients' assessment of quality has been lacking.

We embarked on this study for three main reasons. First, there is a need to explore how a challenging condition like CFS/ME relates to patients' evaluations. Second, we wanted to explore possible associations between quality assessment and health status as measured both by self-rated health and by a more specific measure related to CFS/ME. Third, we wanted to investigate CFS/ME patients' evaluation of primary and specialist care as separate entities, as well as their evaluation of coordination between care givers.

In the present study we had the opportunity to explore all of these aspects, which is relevant because it may influence GPs', specialists', patients', and policy makers' awareness of assessed quality, with possible consequences for clinical practice, communication, cooperation, health outcomes, and planning and organising of health services for CFS/ME patients.

Our aim was to study how CSF/ME patients rated health care quality and coordination of services, and to investigate whether self-rated health and self-rated degree of CFS/ME were associated with CFS/ME patients' assessment of quality in primary and specialist care, and in the coordination of care.

METHODS

Data

This cross-sectional study used postal survey data obtained in April and May 2013 from members of The Norwegian ME Association. Invitations were distributed by Norwegian Social Science Data Service (NSD) Web Survey to a total of 811 members with known email addresses (about 40 % of all members). Non-respondents were given one reminder.

Initially, we had no information about age or reasons for membership, thus, members were asked to refrain from participating if they were below 16 years or if they did not suffer from CFS/ME themselves (health professionals, parents, others). We do not know how many of the non-respondents were not eligible to participate, therefore, an exact response rate cannot be calculated. Other parts of this study have previously been published [24].

The questionnaire included information about demographic and socio-economic characteristics, health status including specific questions about symptoms, duration, severity, and treatment of their CFS/ME, and use of and experiences with health care services.

Participants

Women comprised 89,1 % of the 488 respondents. Due to low numbers, as well as to avoid overfitting [25] and a possible confounding effect of gender in the regression models, we excluded all men (53 respondents). We also excluded those who did not give information about gender (2 respondents) or age (2 respondents). This gave a net sample of 431 respondents (Figure 1).

Variables

Participants were asked about their overall personal experiences with quality and coordination of follow-up and treatment in health care services, from their first contact because of CFS/ME, and until the time of the survey. The dependent variables were based on the three questions presented in Table 1. For an easier interpretation of logistic regressions all three variables were dichotomized by merging the original answering options into "good/very good" and "poor/very poor". Those who answered "not relevant" were excluded from the analyses.

Table 1. Female CFS/ME patients' assessment of quality in primary care, specialist care and in coordination of care (%)

	Very good		Good		Poor		Very poor		Not relevant	
	n	%	n	%	n	%	n	%	n	%
How would you describe the quality of the medical care you have received from primary care? (n=396)	30	7.6	114	28.8	132	33.3	108	27.3	12	3.0
How would you describe the quality of the medical care you have received from specialist care? (n= 392)	33	8.4	128	32.7	106	27.0	81	20.7	44	11.2
How do you think the services in the various parts of the support system are coordinated? (n=385)	2	0.5	61	15.8	152	39.5	122	31.7	48	12.5

The key independent variables were self-rated health and self-rated degree of CFS/ME. Self-rated health was obtained from the question "How would you assess your own health in general?" Response options were reduced from five original categories (very bad - bad - fair - good - excellent) to four by merging the good and excellent categories due to low numbers (five individuals reported excellent health). Self-rated degree of CFS/ME was obtained from the question "What degree of ME do you have as of today?" Four answering options were given; mild (about 50 % reduction in activity), moderate (housebound most of the day), severe (bedridden most of the day), and very severe (completely bedridden). This classification, defined by an international consensus panel [26], has been widely discussed in

the ME Association's membership magazine, and is well known to the members of the ME Association. We merged the severe and very severe categories due to low numbers (five individuals reported very severe disease).

Adjustment independent variables were age, education, duration of the current GP relation (GP duration), and number of GP visits the previous year (GP frequency). Age was grouped in 20-year age groups; however, age in years was used as a continuous variable in the regression models. Six original education categories were merged into four due to low numbers in the outermost groups (one individual with no education and three individuals with a PhD). GP duration was obtained from the question "Approximately for how long have you had your current GP?" Responses were dichotomised into 0-2 years and 3 years or more. GP frequency was obtained from the question "Approximately how many times have you seen your GP, another GP or visited an emergency clinic during the previous 12 months for issues related to your ME?" The answers were categorized into four levels; 0 visits, 1-4 visits, 5-9 visits, and 10 visits or more.

Analyses

Data was analysed by means of descriptive statistics and logistic regressions. Correlations were tested with Spearman's correlation coefficients.

We constructed two sets of multivariable regression models for each of the three dependent variables. The first set included the independent variables age, education, self-rated health, GP duration, and GP frequency, which were all introduced collectively into the model for trend analyses. In the second set of models self-rated health was replaced by self-rated degree of CFS/ME. First order interactions were tested by introducing interaction terms in the regression models.

We used 95% confidence intervals (CI) throughout the study. All analyses were accomplished using Stata, version 13.1.

Ethics

The study has been approved by the Norwegian Data Protection Official (id. 31784).

RESULTS

In total 488 members of the ME-association aged 16-73 years participated, constituting an overall estimated response rate of 60% (Figure 1). Due to non-response from non-eligible receivers and return of emails from email addresses that were not in use, the actual response rate is assumed to be higher. The 431 women constituting the final sample for analyses reported having the diagnoses ME (n=354), CFS (n=31) and/or post viral fatigue syndrome (n=70) (more than one diagnosis was possible).

Mean age of participants was 46.2 years. Most participants (61.8 %) had suffered from CFS/ME for 10 years or more. The highest percentage of people were aged 40-59 years, had university education, poor self-rated health, moderate degree of CFS/ME, a GP relation of 3 years or more, and 1-4 GP visits the previous year (Table 2). In the previous year, 92% of the

patients had visited primary health care services at least once for issues related to their CFS/ME (Table 2).

Table 2. Sample characteristics

	Total sample	
	n	%
Age	431	100.0
16-19	5	1.2
20-39	116	26.9
40-59	244	56.6
60+	66	15.3
Education	398	100.0
Primary	29	7.3
High school	128	32.1
University 1-4 years	156	39.2
University 5 years +	85	21.4
Self-rated health	399	100.0
Very good/excellent	44	11.0
Fair	83	20.8
Poor	205	51.4
Very poor	67	16.8
Degree of CFS/ME	396	100.0
Mild	88	22.2
Moderate	268	67.7
Severe/very severe	40	10.1
GP duration	398	100.0
0-2 years	144	36.2
3 years +	254	63.8
GP frequency*	368	100.0
0 visits	29	7.9
1-4 visits	159	43.2
5-9 visits	107	29.1
10+ visits	73	19.8

* Number of GP visits related to CFS/ME in the previous year

The quality of medical care was assessed poor/very poor by 60.6% in primary care, and by 47.7% in specialist care, whereas 71.2% of the participants regarded coordination between services as poor/very poor (Table 1).

In multivariable analyses with self-rated health in the model we found that poorer self-rated health increased the probability of reporting the quality as poor/very poor both in primary care and in specialist care (Table 3). For primary care, this association was modified by age, as it was stronger in higher ages (interaction term self-rated health x age, OR 1.03, CI

1.01-1.05). Furthermore, this association was modified by education (interaction term self-rated health x education OR 1.48, CI 1.07-2.04), indicating a stronger association in higher educational groups. We found no statistically significant associations between self-rated health and coordination of care (Table 3).

The probability of reporting quality of primary care as poor/very poor decreased with increasing number of GP visits during the previous year (Table 3). This association was modified by self-rated health, and was stronger among those in poorer health (interaction term GP frequency x self-rated health, OR 0.72, CI 0.54-0.96).

Table 3. Female CFS/ME patients' assessment of quality in primary care, specialist care, and coordination of care, according to self-rated health (multivariable logistic regressions)

	Probability of assessing quality in primary care bad/very bad n=348		**Probability of assessing quality in specialist care bad/very bad n=319**		**Probability of assessing quality in coordination of care bad/very bad n=309**	
	OR for trend	95% CI	OR for trend	95% CI	OR for trend	95% CI
Age in years	1.00	0.98-1.02	0.99	0.97-1.01	1.00	0.98-1.03
Education*	0.95	0.73-1.24	1.13	0.86-1.47	0.87	0.61-1.22
Self -rated health**	**1.81**	**1.36-2.41**	**1.38**	**1.05-1.82**	1.24	0.87-1.78
GP duration***	0.64	0.40-1.05	1.06	0.66-1.71	0.62	0.32-1.19
GP visits last year ****	**0.68**	**0.51-0.89**	0.95	0.73-1.25	1.16	0.82-1.65

OR odds ratio; CI confidence interval.
*Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.
** Self-rated health in four groups: 1=very good/excellent, 2=fair, 3=poor, 4=very poor.
*** GP duration: 0=0-2 years, 1=>3years.
**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.
Statistically significant findings are marked in bold.

Similar overall associations were observed for primary health care in models where self-rated health was replaced by self-rated degree of CSF/ME (Table 4), although slightly weaker. In this model, those who had the same GP for three years or more were less likely to report the quality of primary care as poor/very poor. The effect modifications in the self-rated health model were not replicated in this model. No significant associations in quality reports were observed for specialist care or coordination of care (Table 4).

Table 4. Female CFS/ME patients' assessment of quality in primary care, in specialist care, and in coordination of care according to self-rated degree of CFS/ME (multivariable logistic regressions)

	Probability of assessing quality in primary care bad/very bad n=346		**Probability of assessing quality in specialist care bad/very bad n=317**		**Probability of assessing quality in coordination of care bad/very bad n=308**	
	OR for trend	95% CI	OR for trend	95% CI	OR for trend	95% CI
Age in years	1.00	0.98-1.02	0.98	0.96-1.01	1.00	0.98-1.03
Education*	1.05	0.81-1.37	1.18	0.90-1.54	0.86	0.61-1.22
Degree of CFS/ME**	**1.68**	**1.10-2.57**	1.46	0.95-2.24	1.52	0.87-2.67
GP duration***	**0.61**	**0.38-0.99**	1.00	0.62-1.60	0.61	0.32-1.17
GP visits last year****	**0.70**	**0.54-0.93**	0.94	0.72-1.24	1.14	0.80-1.64

OR odds ratio; CI confidence interval.
*Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.
** Degree of ME in 4 groups: 1=mild, 2=moderate 3=severe/very severe.
*** GP duration: 0=0-2 years, 1=>3years.
**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.
Statistically significant findings are marked in bold.

There were no strong correlations (defined as rho >0.5) between any of the independent variables in any of the models. We found a modest correlation between self-rated health and self-rated degree of CFS/ME (rho=0.5067) but these variables were not both included in any model.

DISCUSSION

We found that a poorer self-rated health, as well as a more severe degree of CFS/ME, increased the probability of poor/very poor quality scores in primary care, as reported by women with CFS/ME. Similar scores were found for self-rated health and specialist care, although weaker. Coordination of care was assessed bad/very bad by most of the study participants, regardless of self-rated health and self-rated degree of CFS/ME. Frequent GP visitors were less likely to report the quality in primary care as bad/very bad compared to those who visited less frequently.

CFS/ME patients' assessment of health care services in relation to self-rated degree of CFS/ME is largely unknown, as this measure has hardly been used in previous studies. Self-rated health is a more commonly used but less specific measure as it refers to general health and not specifically CFS/ME-related health. Our finding that poorer self-rated health was associated with lower quality scores is in line with most previous studies across diagnoses and health care settings [18-21, 27]. It is worth noting that similar findings were slightly weaker for self-rated degree of CFS/ME compared to self-rated health, indicating that self-rated

health encompasses a wider range of issues and complexity of health problems [19, 22, 23], even for patients with a complex and challenging condition like CFS/ME. This is also confirmed by a no more than modest correlation between these two variables.

Quality in primary care was more likely reported low by patients with a shorter GP relation and less frequent GP visits. These variables might be intertwined. Previous studies have suggested that continuity of GP care is associated with higher patient satisfaction [24], and that people in poorer health are more likely to have shorter GP relationships [12]. Some of these patients might suffer from ailments that do not fit into specific diagnoses, thus generating dissatisfaction and a search for another GP [27]. In the present study 36.2 % reported a short duration of their GP relation, indicating that CFS/ME patients might replace their GPs to a higher extent than the general population [12]. In line with our findings, others have reported a positive GP assessment to be associated with increased frequency of attendance [27]. This might indicate that once patients have found an understanding GP, they consider GP visits beneficial and therefore visit more frequently.

In an international comparison of patient-evaluated GP care in 10 European countries, 76 % of Norwegian patients viewed care as good/excellent, a score below the study average [28]. Only 36.4 % of our study participants viewed GP care as good/very good. Despite differences between the studies, both strongly indicate that CFS/ME patients are less satisfied than the general population. This is reinforced by the fact that there are only women in our study, as women in general are more satisfied with health care than men [29]. Regarding the notion that communication and the GP-patient relationship are important tools in the treatment of CFS/ME, this is a cause for concern.

Slightly more than half of the participants reported the quality of specialist care as good/very good. A Swedish study of outpatient care in all hospital specialties found that on average more than 80 % were satisfied [29], which largely contradicted our findings. Hence, similarly to primary care CFS/ME patients seem to be less satisfied than patients in general. This is not surprising, since quality of health care is often regarded lower by people in poorer health [18, 19, 21].

We found that quality scores for specialist care were better than for primary care. Previous research report that many GPs are constrained by the scientific uncertainty of CFS/ME [30], unconfident with diagnosing and treating the condition [31], and worried that the label of CFS/ME might be potentially harmful to the patient [32]. Patients with CFS/ME or other medically unexplained conditions on the other hand, have reported feeling belittled, stigmatized, distrusted, rejected and ignored by their doctors, and that their moral character and the reality of their symptoms are questioned [33, 34]. These aspects might partly explain the low quality scores in primary care. We reported in a previous paper that patients do value referrals [24], which is in line with patients' reports that specialist services provide acknowledgment of their ailments, treatment, better handling of daily life issues, and improved dialogue between professionals [35]. However, CFS/ME patients often have to struggle for a referral [36, 37], which might affect the relationship with their GP. Patients in Europe have evaluated GP care more positively in countries without gate-keeping [28], and the Norwegian gate-keeping system could explain some of the low primary health care scores.

Quality in coordination of care was rated poor/very poor by 71.2 % of the patients, with no significant differences according to the independent variables in the study. This is in line with experiences of patients with complex health care needs internationally [38] and with the Norwegian scores in the CWF international comparisons [14, 15]. Despite the Norwegian Coordination Reform (2012), aiming to facilitate better coordination in health care [39], challenges seem large in this field.

Are self-rated health and self-rated degree of CFS/ME to be regarded as measures of individual characteristics or measures of health outcome? This is worth some reflection considering the controversies of this condition, where communication between patient and doctor is regarded as an important treatment tool. Most participants in our study reported poor health and a moderate to severe degree of CFS/ME, thus indicating a complex health situation. The individual-characteristic perspective may indicate that a bad quality of care is influenced by the patients' receptiveness of the offered care, which in turn may lead to blaming the patient herself for a possible bad quality. On the other hand, the outcome perspective might indicate that a bad quality of care is influenced by the health care providers' inability to handle patients and their condition, which in turn may lead to blaming the doctor and health care services for a possible bad quality. In real life these perspectives might be intertwined. However, according to professional ethics and health care laws [40], the doctor is the one responsible for quality in the medical encounter, regardless of patient characteristics and issues raised. Considering health status as (at least partly) a measure of outcome, it is not surprising that poorer self-rated health and a more severe degree of CFS/ME was associated with lower quality scores. In line with this we assume that patients emphasize the outcome perspective. This notion might also be underpinned by the result that the low quality scores for primary and specialist services both were associated with poorer health status, whereas coordination of care was not.

A particular strength of the study was the recruitment of study participants from a patient organization since it is detached from sites of care, with the aim to enable patients to describe their experiences without fear of how this information might affect their relation to health care providers. We used a well-designed systematically tested questionnaire, and the response rate was acceptable. By studying a specific group of patients we have been able to interpret our findings according to a relatively homogenous group.

This study had some limitations. Our sample may not fully represent women with CFS/ME. First, there might be a selection regarding membership of the patient organization. Survivor bias may be a part of this, indicating that the healthiest patients will not demand membership to the same extent as those in poorer health [41]. On the other hand, the most seriously affected members might have refrained from participating because of disease severity. The direction of a possible selection bias from these factors is not obvious. A general population satisfaction study reported that non-respondents were overrepresented in groups with lower satisfaction [29], indicating that a possible selection bias might have skewed our study in the direction of better satisfaction scores than would otherwise be found. However, female CFS/ME patients might differ from this population [34]. Second, the distribution of e-mail addresses might have been skewed, for instance towards younger members with higher education. However, since 93 % of Norwegian households have access to the internet [42] we

find it unlikely that this have influenced our results to a significant degree. Third, our sample had lower age and higher education than the Norwegian average [43]. A possible skewness regarding these variables might be connected, since younger individuals will not have completed their education.

In questionnaire data there is always a potential for recall bias, particularly regarding minor events and distant past, usually leading to underreporting. While, some studies indicate that doctors hold the opinion that CFS/ME patients often exaggerate the severity of their ailments [34]. Anyway, it is difficult to judge whether overreporting or underreporting might be present in our data. The validity of self-reported data on disease severity may be disputable per se, but in the case of CFS/ME, where no objective tests are diagnostic or suitable to evaluate disease development and severity, self-rated degree of ME might actually be a strong contender as the golden standard for describing disease severity.

Self-rated health and self-rated degree of CFS/ME describes the health status at the time of the survey, whereas participants assess the quality of their care from the first onset of their symptoms. This might be a problem, in particular if self-rated health and self-rated degree of CFS/ME, with its fluctuating nature, are considered individual characteristics. However, we have argued that patients most likely consider these variables as outcome measures, and the difference in observation time might thus lack significant impact. Besides, self-rated assessments of health status might not be solely limited to the current status of health or CFS/ME [22, 23]. The questionnaire wording may draw in this direction for self-rated health, as it emphasizes health status more generally ("How would you assess your own health in general?"). However, for self-rated degree of CFS/ME the current situation is emphasized to a greater extent ("What degree of ME do you have as of today?"). All in all, it remains unclear whether this might have affected the assessments. Most likely it has not had any significant impact on our main results.

The cross sectional study design implicates that no causal relationships can be established. For future research we would recommend a longitudinal design investigating factors relevant to patients' quality assessment over time.

CONCLUSIONS

We concluded that a large proportion of women with CFS/ME rated the quality of their care poor or very poor for primary care, specialist care, and coordination of care. The dissatisfaction was higher for primary care than for specialist care, and even higher for coordination of care. Poorer self-rated health and a more severe degree of CFS/ME were associated with lower quality scores, particularly in primary care services, but were not associated with coordination between services. The findings indicate that quality in health care services, as assessed by patients with CFS/ME, do have a significant potential for improvement. In order to achieve this, health care services must recognise and acknowledge the voice of the users, which is a foundational concept in all medical practice as well as a precondition for congruence in doctor-patient relationships and shared decision-making. This

is particularly important when the consultation is likely to provide neither an explanation nor a remedy.

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COMPETING INTERESTS

The authors declare that they have no competing interests.

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FIGURES

Figure 1

Flow chart of study participants.

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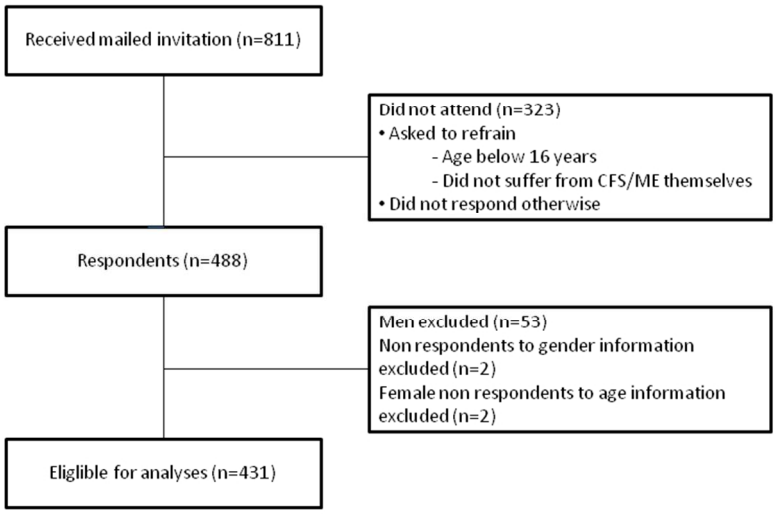

Flow chart of study participants
254x190mm (96 x 96 DPI)

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How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services? A cross-sectional study

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How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services?

A cross-sectional study

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How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services?

A cross-sectional study

ABSTRACT

Objective: To test the association between self-rated health and self-rated degree of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), and CFS/ME patients' assessment of quality of primary care, specialist care, and coordination of care.

Design: Cross-sectional study.

Setting: Self-reported questionnaire data from women members of The Norwegian ME Association obtained in 2013.

Participants: 431 women with CFS/ME aged 16-73 years.

Main outcome measure: The participants' assessment of quality in primary care, specialist care, and in coordination of care (good/very good or poor/very poor). Main explanatory variables: self-rated health and self-rated degree of CFS/ME.

Results: Quality of care was rated poor by 60.6% in primary care, by 47.7% in specialist care, and by 71.2% regarding coordination of care. Poorer self-rated health increased the probability of rating quality in primary care poor, particularly among women 40 years and over (odds ratio [OR] 2.38, 95% confidence interval [CI] 1.63-3.49), women with university education (OR 2.57, CI 1.68-3.94), and less frequent GP visits (OR 2.46, CI 1.60-3.78). Poorer self-rated health increased the probability of rating quality poor in specialist care (OR 1.38, CI 1.05-1.82), but not in coordination of care. A more severe CFS/ME was associated with higher probability of rating quality in primary care poor (OR 0.61, CI 0.38-0.93). Frequent visitors and those with a long GP relationship were less likely to report primary care quality as poor.

Conclusions: A large proportion of women with CFS/ME rated quality of care poor/very poor in primary care, specialist care, and in coordination of care. The dissatisfaction was higher for primary care than for specialist care. Overall, poorer self-rated health and a more severe CFS/ME were associated with lower quality scores in primary and specialist care, but not in coordination of care. Health care services, as assessed by women with CFS/ME, do have a large potential for improvement.

STRENGHTS AND LIMITATIONS OF THIS STUDY

- Recruitment of a homogenous group of study participants from a patient organization is a study strength
- Use of a systematically tested questionnaire and an acceptable response rate is a strength
- There might be a selection bias due to recruitment from a patient organization, as well as a possible recall bias as for most questionnaire data, which are limitations of the study
- Validity of self-reported measures might be discussed, and in particular measures of disease severity and health
- The cross-sectional design precludes any causal interpretation

How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services?

A cross-sectional study

INTRODUCTION

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), is a condition with an estimated prevalence around 1-2 per thousand [1]. It affects women more than men (70-85%), and etiology is unknown [2-4]. CFS/ME is characterized by its fluctuating nature, physical and mental fatigue, persistent post-exertional malaise, sleep disturbances, problems with concentration and memory, pain in muscles and joints, headache, and other symptoms related to cognitive, immune, and autonomous dysfunctions [5-7]. The terms CFS and ME are used interchangeably but mostly as a composite term, and the distinction between them is disputed [5]. In this paper we use the combined term CFS/ME as recommended by Norwegian health authorities and others [7, 8].

CFS/ME cannot be confirmed by specific tests, it is not localized to a specific part of the body, and it challenges the traditional distinction between psyche and soma [9]. In line with this, CFS/ME is a controversial condition and poses particular challenges in terms of diagnosis, therapy, and communication, to patients as well as to physicians [10].

CFS/ME patients often need coordinated health services from multiple providers over time. General practitioners (GPs) are their main contacts. All Norwegian citizens are provided a regular GP, and only 0,4 % of the population has chosen to remain outside GPs' lists [11]. Residents can change GP twice a year without justification. Together with universal tax-funding and gate-keeping, the list system provides strong incentives for personal continuity of care [12]. First line medical services including emergency clinics are run by the municipalities. Specialist services, consisting of hospitals and outpatient clinics, are run by regional health enterprises and are mainly owned by the state. Access is usually achieved by referrals from the GP (the gate-keeper role). GP and specialist outpatient visits are co-payed by adult patients.

Health care services, and GPs in particular, are well regarded in the population [13]. However, Norway's scores in the annual patient/population assessed international comparisons of health systems by the Commonwealth Fund (CWF) have been less than average in areas like general quality of care, waiting time for appointments, information sharing, communication, and coordination of services [14, 15]. Patient satisfaction, defined as "an individual's cognitive evaluation of, and emotional reaction to, his or her health-care experience" [16], is considered an important indication of overall quality in health care [17]. The Norwegian scores, thereby, indicate particular challenge areas for health care providers and policy makers.

Across health care settings, patient satisfaction or patients' assessment of health care quality is positively associated with better self-rated health and functional status, and inversely associated with the complexity of health problems [18-21]. In striving towards

evidence based improvement of health care delivery, research on patient-based assessments of quality and coordination is informative, especially for groups with chronic diseases and long-term needs of care from multiple providers, like CFS/ME. Moreover, this group of patients is interesting due to previously reported serious challenges in collaboration with health care services and providers, in line with the contested status of the condition [9].

The familiar measure self-rated health captures a comprehensive range of aspects and provides summative information about the individual's history of health and disease as well as the current level of health [22, 23]. However, a measure related to the disease of interest might provide a more specific indication of how disease severity and patients' ratings might be associated. Self-rated degree of CFS/ME might be such a measure. Solid evidence whether differences in health status might be associated with CFS/ME patients' assessment of quality has been lacking.

We embarked on this study for three main reasons. First, there is a need to explore how a challenging condition like CFS/ME relates to patients' evaluations. Second, we wanted to explore possible associations between quality assessment and health status as measured both by self-rated health and by a more specific measure related to CFS/ME. Third, we wanted to investigate CFS/ME patients' evaluation of primary and specialist care as separate entities, as well as their evaluation of coordination between care givers.

In the present study we had the opportunity to explore all of these aspects, which is relevant because it may influence GPs', specialists', patients', and policy makers' awareness of assessed quality, with possible consequences for clinical practice, communication, cooperation, health outcomes, and planning and organising of health services for CFS/ME patients.

Our aim was to study how CFS/ME patients rated health care quality and coordination of services, and to investigate whether self-rated health and self-rated degree of CFS/ME were associated with CFS/ME patients' assessment of quality in primary and specialist care, and in the coordination of care.

METHODS

Data

This cross-sectional study used postal survey data obtained in April and May 2013 from members of The Norwegian ME Association. Invitations were distributed by Norwegian Social Science Data Service (NSD) Web Survey to a total of 811 members with known email addresses (about 40 % of all members). Non-respondents were given one reminder.

Initially, we had no information about age or reasons for membership, thus, members were asked to refrain from participating if they were below 16 years or if they did not suffer from CFS/ME themselves (health professionals, parents, others). We do not know how many of the non-respondents were not eligible to participate, therefore, an exact response rate cannot be calculated. Other parts of this study have previously been published [24].

The questionnaire included information about demographic and socio-economic characteristics, health status including specific questions about symptoms, duration, severity, and treatment of their CFS/ME, and use of and experiences with health care services.

Participants

Women comprised 89.1 % of the 488 respondents. Due to low numbers, as well as to avoid overfitting [25] and a possible confounding effect of gender in the regression models, we excluded all men (53 respondents). We also excluded those who did not give information about gender (2 respondents) or age (2 respondents). This gave a net sample of 431 respondents (Figure 1).

Variables

Participants were asked about their overall personal experiences with quality and coordination of follow-up and treatment in health care services, from their first contact because of CFS/ME, and until the time of the survey. The dependent variables were based on the three questions presented in Table 1. For an easier interpretation of logistic regressions all three variables were dichotomised by merging the original answering options into "good/very good" and "poor/very poor". Those who answered "not relevant" were excluded from the analyses.

Table 1. Female CFS/ME patients' assessment of quality in primary care, specialist care and in coordination of care (%)

	Very good	Good	Poor	Very poor	Not relevant
	n (%)	n (%)	n (%)	n (%)	n (%)
How would you describe the quality of the medical care you have received from primary care? (n=396)	30 (7.6)	114 (28.8)	132 (33.3)	108 (27.3)	12 (3.0)
How would you describe the quality of the medical care you have received from specialist care? (n= 392)	33 (8.4)	128 (32.7)	106 (27.0)	81 (20.7)	44 (11.2)
How do you think the services in the various parts of the support system are coordinated? (n=385)	2 (0.5)	61 (15.8)	152 (39.5)	122 (31.7)	48 (12.5)

The key independent variables were self-rated health and self-rated degree of CFS/ME. Self-rated health was obtained from the question "How would you assess your own health in general?" Response options were reduced from five original categories (very poor – poor - fair - good - excellent) to four by merging the good and excellent categories due to low numbers (five individuals reported excellent health). Self-rated degree of CFS/ME was obtained from the question "What degree of ME do you have as of today?" Four answering options were given; mild (about 50 % reduction in activity), moderate (housebound most of the day), severe (bedridden most of the day), and very severe (completely bedridden). This

classification, defined by an international consensus panel [26], has been widely discussed in the ME Association's membership magazine, and is well known to the members of the ME Association. We merged the severe and very severe categories due to low numbers (five individuals reported very severe disease).

Adjustment independent variables were age, education, duration of the current GP relation (GP duration), and number of GP visits the previous year (GP frequency). Six original education categories were merged into four due to low numbers in the outermost groups (one individual with no education and three individuals with a PhD). GP duration was obtained from the question "Approximately for how long have you had your current GP?" Responses were dichotomised into 0-2 years and 3 years or more. GP frequency was obtained from the question "Approximately how many times have you seen your GP, another GP or visited an emergency clinic during the previous 12 months for issues related to your ME?" The answers were categorized into four levels; 0 visits, 1-4 visits, 5-9 visits, and 10 visits or more.

Analyses

Data was analysed by means of descriptive statistics and logistic regressions. Correlations were tested with Spearman's correlation coefficients.

We constructed two sets of multivariable regression models for each of the three dependent variables. The first set included the independent variables self-rated health, age, education, GP duration, and GP frequency, which were introduced collectively into the model. In the second set of models self-rated health was replaced by self-rated degree of CFS/ME. We performed dummy-analyses as well as trend analyses. Since some of the groups were small, and there was no significant lack of linearity, we chose to report the trend analyses exclusively. We tested first order interactions by introducing interaction terms in the regression models. Where interactions were present, we performed stratified analyses accordingly.

We used 95% confidence intervals (CI) throughout the study. All analyses were accomplished using Stata, version 13.1.

Ethics

The study has been approved by the Norwegian Data Protection Official (id. 31784).

RESULTS

In total 488 members of the ME-association aged 16-73 years (mean age 46.2 years) participated, constituting an overall estimated response rate of 60% (Figure 1). Due to non-response from non-eligible receivers and return of emails from email addresses that were not in use, the actual response rate is assumed to be higher. The 431 women constituting the final sample for analyses reported having the diagnoses ME (n=354), CFS (n=31) and/or post viral fatigue syndrome (n=70) (more than one diagnosis possible).

Most participants (61.8 %) had suffered from CFS/ME for 10 years or more. The highest percentage of people were aged 40-59 years, had university education, poor self-rated

health, moderate degree of CFS/ME, a GP relation of 3 years or more, and 1-4 GP visits the previous year (Table 2). In the previous year, 92% of the patients had visited primary health care services at least once for issues related to their CFS/ME (Table 2).

Table 2. Sample characteristics

	Total sample	
	n	%
Age	431	100.0
16-19	5	1.2
20-39	116	26.9
40-59	244	56.6
60+	66	15.3
Education	398	100.0
Primary	29	7.3
High school	128	32.1
University 1-4 years	156	39.2
University 5 years +	85	21.4
Self-rated health	399	100.0
Very good/excellent	44	11.0
Fair	83	20.8
Poor	205	51.4
Very poor	67	16.8
Degree of CFS/ME	396	100.0
Mild	88	22.2
Moderate	268	67.7
Severe/very severe	40	10.1
GP duration	398	100.0
0-2 years	144	36.2
3 years +	254	63.8
GP frequency*	368	100.0
0 visits	29	7.9
1-4 visits	159	43.2
5-9 visits	107	29.1
10+ visits	73	19.8

* Number of GP visits related to CFS/ME in the previous year

The quality of medical care was assessed poor/very poor by 60.6% in primary care, and by 47.7% in specialist care, whereas 71.2% of the participants regarded coordination between services as poor/very poor (Table 1).

In multivariable analyses we found that the associations between primary care quality assessments and self-rated health were modified by age (interaction term between self-rated

health and age, p-value (p)=0.010), education (interaction term between self-rated health and education, p=0.016), and GP frequency (interaction term between GP frequency and self-rated health, p=0.025), indicating a stronger association between poorer quality assessment and poorer self-rated health among women in higher age, with higher education, and less frequent GP visits. In analyses stratified by age, education, and GP frequency, these associations were statistically significant only in women 40 years and over, in women with university education, and in women who visited their GP four times or less during the previous year (Table 3). Also, quality in primary care was more likely reported poor/very poor by women with a shorter GP relation if they had university education or less frequent GP visits the previous year (Table 3).

Table 3. Female CFS/ME patients' probability of assessing quality poor/very poor in primary care, according to self-rated health (multivariable regression analyses stratified by age, education, and GP frequency).

	Age		Education		GP frequency	
	16-39 years (n=103)	40 years and over (n=245)	Primary/high school (n=137)	University (n=211)	0-4 visits (n=177)	5+ visits (n=171)
	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)
Self-rated health*	1.35 (0.85-2.15)	**2.38 (1.63-3.49)**	1.28 (0.85-1.94)	**2.57 (1.68-3.94)**	**2.46 (1.60-3.78)**	1.34 (0.90-1.99)
Age in years	-	-	1.00 (0.97-1.03)	0.98 (0.95-1.01)	1.02 (0.98-1.05)	0.99 (0.96-1.02)
Education**	1.26 (0.80-1.98)	0.73 (0.52-1.04)	-	-	0.67 (0.45-1.02)	1.33 (0.92-1.95)
GP duration***	0.52 (0.22-1.19)	0.70 (0.38-1.30)	0.88 (0.42-1.85)	**0.49 (0.25-0.97)**	**0.43 (0.19-0.95)**	0.93 (0.50-1.75)
GP frequency ****	0.83 (0.52-1.32)	**0.62 (0.44-0.88)**	**0.55 (0.36-0.84)**	0.75 (0.51-1.10)	-	-

OR odds ratio; CI confidence interval.

* Self-rated health in four groups: 1=very good/excellent, 2=fair, 3=poor, 4=very poor.

** Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.

*** GP duration: 0=0-2 years, 1=>3years.

**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.

Statistically significant findings are marked in bold.

Poorer self-rated health increased the probability of reporting the quality as poor/very poor in specialist care, whereas we made no significant findings regarding coordination of care (Table 4).

Table 4. Female CFS/ME patients' probability of assessing quality poor/very poor in specialist care and coordination of care, according to self-rated health (multivariable logistic regressions)

	Specialist care (n=319)	**Coordination of care (n=309)**
	OR for trend (95% CI)	OR for trend (95% CI)
Self -rated health*	**1.38 (1.05-1.82)**	1.24 (0.87-1.78)
Age in years	0.99 (0.97-1.01)	1.00 (0.98-1.03)
Education**	1.13 (0.86-1.47)	0.87 (0.61-1.22)
GP duration***	1.06 (0.66-1.71)	0.62 (0.32-1.19)
GP frequency ****	0.95 (0.73-1.25)	1.16 (0.82-1.65)

OR odds ratio; CI confidence interval.

* Self-rated health in four groups: 1=very good/excellent, 2=fair, 3=poor, 4=very poor.

** Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.

*** GP duration: 0=0-2 years, 1=>3years.

**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.

Statistically significant findings are marked in bold.

In models where self-rated health was replaced by self-rated degree of CFS/ME, we found that a more severe CFS/ME increased the probability of rating quality in primary care poor/very poor (Table 5). In this model, those who had the same GP for three years or more, and those who visited more frequently, were less likely to report the quality of primary care as poor/very poor. The effect modifications in the self-rated health model were not replicated in this model. No significant associations in quality reports were observed for specialist care or coordination of care (Table 5).

Table 5. Female CFS/ME patients' probability of assessing quality poor/very poor in primary care, specialist care, and coordination of care, according to self-rated degree of CFS/ME (multivariable logistic regressions)

	Primary care (n=346)	**Specialist care (n=317)**	**Coordination of care (n=308)**
	OR for trend (95% CI)	OR for trend 95% CI	OR for trend 95% CI
Degree of CFS/ME*	**1.68 (1.10-2.57)**	1.46 (0.95-2.24)	1.52 (0.87-2.67)

Age in years	1.00 (0.98-1.02)	0.98 (0.96-1.01)	1.01 (0.98-1.03)
Education**	1.05 (0.81-1.37)	1.18 (0.90-1.54)	0.86 (0.61-1.22)
GP duration***	**0.61 (0.38-0.99)**	1.00 (0.62-1.60)	0.61 (0.32-1.17)
GP frequency****	**0.70 (0.54-0.93)**	0.94 (0.72-1.24)	1.14 (0.80-1.64)

OR odds ratio; CI confidence interval.
* Degree of ME in 3 groups: 1=mild, 2=moderate, 3=severe/very severe.
** Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.
*** GP duration: 0=0-2 years, 1=>3years.
**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.
Statistically significant findings are marked in bold.

There were no strong correlations (defined as rho >0.5) between any of the independent variables in any of the models. We found a modest correlation between self-rated health and self-rated degree of CFS/ME (rho=0.5067) but these variables were not both included in any model.

DISCUSSION

We found that primary and specialist care quality was rated as poor/very poor by 60.6 and 47.7 % of study participants, respectively. Poorer self-rated health increased the probability of poor/very poor quality scores both in primary and specialist care. In primary care, these findings were statistically significant among women 40 years and over, among women with higher education, and among women who visited their GP four times or less during the previous year. A more severe CFS/ME was associated with higher probability of rating primary care, but not specialist care, poor. Coordination of care was assessed poor/very poor by most of the study participants, regardless of self-rated health and self-rated degree of CFS/ME. Overall, frequent visitors and those with a long GP relationship were less likely to report poor primary care quality.

CFS/ME patients' assessment of health care services in relation to self-rated degree of CFS/ME is largely unknown, as this measure has hardly been used in previous studies. Self-rated health is a more commonly used but less specific measure as it refers to general health and not specifically CFS/ME-related health. Our finding that poorer self-rated health was associated with lower quality scores confirms with most previous studies across diagnoses and health care settings [18-21, 27]. It is worth noting that similar findings were slightly weaker for self-rated degree of CFS/ME compared to self-rated health, indicating that self-rated health encompasses a wider range of issues and complexity of health problems [19, 22,

23], even for patients with a complex and challenging condition like CFS/ME. This is also confirmed by a no more than modest correlation between these two variables.

Overall, quality in primary care was more likely reported low by patients with a shorter GP relation and less frequent GP visits. These variables might be intertwined. Previous studies have suggested that continuity of GP care is associated with higher patient satisfaction [24], and that people in poorer health are more likely to have shorter GP relationships [12]. Some of these patients might suffer from ailments that do not fit into specific diagnoses, thus generating dissatisfaction and a search for another GP [27]. In the present study, 36.2 % reported a short duration of their GP relation, indicating that CFS/ME patients might replace their GPs to a higher extent than the general population [12]. In line with our findings, others have reported a positive GP assessment to be associated with increased frequency of attendance [27]. This might indicate that once patients have found an understanding GP, they consider GP visits beneficial and therefore visit more frequently.

In an international comparison of patient-evaluated GP care in 10 European countries, 76 % of Norwegian patients viewed care as good/excellent, a score below the study average [28]. Only 36.4 % of our study participants viewed GP care as good/very good. Despite differences between the studies, both strongly indicate that CFS/ME patients are less satisfied than the general population. This is reinforced by the fact that there are only women in our study, as women in general are more satisfied with health care than men [29]. Regarding the notion that communication and the GP-patient relationship are important tools in the treatment of medically contested conditions like CFS/ME [30], this is a cause for concern.

Slightly more than half of the participants reported the quality of specialist care as good/very good. A Swedish study of outpatient care in all hospital specialties found that on average more than 80 % were satisfied [29], which largely contradicts our findings. Hence, similarly to primary care, CFS/ME patients seem to be less satisfied than patients in general. This is not surprising, since quality of health care is often regarded lower by people in poorer health [18, 19, 21].

We found that quality scores for specialist care were better than for primary care. Previous research report that many GPs are constrained by the scientific uncertainty of CFS/ME [31], unconfident with diagnosing and treating the condition [32], and worried that the label of CFS/ME might be potentially harmful to the patient [33]. Patients with CFS/ME or other medically unexplained conditions on the other hand, have reported feeling belittled, stigmatized, distrusted, rejected and ignored by their doctors, and that their moral character and the reality of their symptoms are questioned [34, 35]. These aspects might partly explain the low quality scores, especially in primary care where doctors are likely to be more skeptical towards the CFS/ME diagnosis than those who have specialized in dealing with them [36]. We reported in a previous paper that patients do value referrals [24], which is in accordance with patients' reports that specialist services provide acknowledgment of their ailments, treatment, better handling of daily life issues, and improved dialogue between

professionals [37]. However, CFS/ME patients often have to struggle for a referral [38, 39], which might affect the relationship with their GP. Patients in Europe have evaluated GP care more positively in countries without gate-keeping [28], and the Norwegian gate-keeping system could explain some of the low primary health care scores.

Quality in coordination of care was rated poor/very poor by 71.2 % of the patients, with no significant differences according to the independent variables in the study. This corresponds to experiences of patients with complex health care needs internationally [40] and with the Norwegian scores in the CWF international comparisons [14, 15]. Despite the Norwegian Coordination Reform (2012), aiming to facilitate better coordination in health care [41], challenges seem large in this field.

Are self-rated health and self-rated degree of CFS/ME to be regarded as measures of individual characteristics or measures of health outcome? This is worth some reflection considering the controversies of this condition, where communication between patient and doctor is regarded as an important treatment tool [30]. Most participants in our study reported poor health and a moderate to severe degree of CFS/ME, thus indicating a complex health situation. The individual-characteristic perspective may indicate that a poor quality of care is influenced by the patients' receptiveness of the offered care, which in turn may lead to blaming the patient herself for a possible poor quality. On the other hand, the outcome perspective might indicate that a poor quality of care is influenced by the health care providers' inability to handle patients and their condition, which in turn may lead to blaming the doctor and health care services for a possible poor quality. In real life these perspectives might be intertwined. However, according to professional ethics and health care laws [42], the doctor is the one responsible for quality in the medical encounter, regardless of patient characteristics and issues raised. Considering health status as (at least partly) a measure of outcome, it is not surprising that poorer self-rated health and a more severe degree of CFS/ME was associated with lower quality scores. In line with this we assume that patients emphasize the outcome perspective. This notion might also be underpinned by the result that the low quality scores for primary and specialist services both were associated with poorer health status, whereas coordination of care was not.

A particular strength of the study was the recruitment of study participants from a patient organization since it is detached from sites of care, with the aim to enable patients to describe their experiences without fear of how this information might affect their relation to health care providers. We used a well-designed systematically tested questionnaire, and the response rate was acceptable. By studying a specific group of patients we have been able to interpret our findings according to a relatively homogenous group.

This study had some limitations. Our sample may not fully represent women with CFS/ME. First, there might be a selection regarding membership of the patient organization. Survivor bias may be a part of this, indicating that the healthiest patients will not demand membership to the same extent as those in poorer health [43]. On the other hand, the most

seriously affected members might have refrained from participating because of disease severity. The direction of a possible selection bias from these factors is not obvious. A general population satisfaction study reported that non-respondents were overrepresented in groups with lower satisfaction [29], indicating that a possible selection bias might have skewed our study in the direction of better satisfaction scores than would otherwise be found. However, female CFS/ME patients might differ from this population [35]. Second, the distribution of e-mail addresses might have been skewed, for instance towards younger members with higher education. However, since 93 % of Norwegian households have access to the internet [44] we find it unlikely that this have influenced our results to a significant degree. Third, our sample had lower age and higher education than the Norwegian average [45]. A possible skewness regarding these variables might be connected, since younger individuals will not have completed their education.

In questionnaire data there is always a potential for recall bias, particularly regarding minor events and distant past, usually leading to underreporting. While, some studies indicate that doctors hold the opinion that CFS/ME patients often exaggerate the severity of their ailments [35]. Anyway, it is difficult to judge whether overreporting or underreporting might be present in our data. The validity of self-reported data on disease severity may be disputable per se, but in the case of CFS/ME, where no objective tests are diagnostic or suitable to evaluate disease development and severity, self-rated degree of ME might actually be a strong contender as the golden standard for describing disease severity.

Self-rated health and self-rated degree of CFS/ME describes the health status at the time of the survey, whereas participants assess the quality of their care from the first onset of their symptoms. This might be a problem, in particular if self-rated health and self-rated degree of CFS/ME, with its fluctuating nature, are considered individual characteristics. However, we have argued that patients most likely consider these variables as outcome measures, and the difference in observation time might thus lack significant impact. Besides, self-rated assessments of health status might not be solely limited to the current status of health or CFS/ME [22, 23]. The questionnaire wording may draw in this direction for self-rated health, as it emphasizes health status more generally ("How would you assess your own health in general?"). However, for self-rated degree of CFS/ME the current situation is emphasized to a greater extent ("What degree of ME do you have as of today?"). All in all, it remains unclear whether this might have affected the assessments. Most likely it has not had any significant impact on our main results.

The cross sectional study design implicates that no causal relationships can be established. For future research we would recommend a longitudinal design investigating factors relevant to patients' quality assessment over time.

CONCLUSIONS

We concluded that a large proportion of women with CFS/ME rated the quality of their care poor or very poor for primary care, specialist care, and coordination of care. The dissatisfaction was higher for primary care than for specialist care, and even higher for coordination of care. Poorer self-rated health and a more severe degree of CFS/ME were associated with lower quality scores, particularly in primary care services, but were not associated with coordination between services. The findings indicate that quality in health care services, as assessed by patients with CFS/ME, do have a significant potential for improvement. In order to achieve this, health care services must recognise and acknowledge the voice of the users, which is a fundamental value in all medical practice as well as a precondition for congruence in doctor-patient relationships and shared decision-making. This is particularly important when the consultation is likely to provide neither an explanation nor a remedy.

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CONTRIBUTORSHIP STATEMENT
Both the authors contributed to the design and conduct of the study. OSL designed the questionnaire, provided funding and collected the data. AHH undertook the statistical analyses and drafted the manuscript. OSL contributed with major improvements and critical revisions. Both the authors approved the final version for publication.

COMPETING INTERESTS
The authors declare that they have no competing interests.

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DATA SHARING STATEMENT
No additional data available.

FIGURES
Figure 1

Flow chart of study participants.

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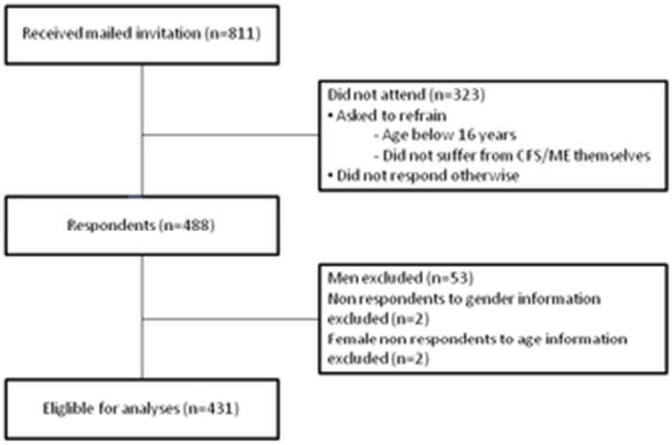

Figure 1. Flow chart of study population.
36x27mm (300 x 300 DPI)

STROBE Statement—Checklist of items that should be included in reports of ***cross-sectional studies***

	Item No	Recommendation
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found
Introduction		
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported
Objectives	3	State specific objectives, including any prespecified hypotheses
Methods		
Study design	4	Present key elements of study design early in the paper
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group
Bias	9	Describe any efforts to address potential sources of bias
Study size	10	Explain how the study size was arrived at
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding
		(*b*) Describe any methods used to examine subgroups and interactions
		(*c*) Explain how missing data were addressed
		(*d*) If applicable, describe analytical methods taking account of sampling strategy
		(*e*) Describe any sensitivity analyses
Results		
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed
		(b) Give reasons for non-participation at each stage
		(c) Consider use of a flow diagram
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders
		(b) Indicate number of participants with missing data for each variable of interest
Outcome data	15*	Report numbers of outcome events or summary measures
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included
		(*b*) Report category boundaries when continuous variables were categorized
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses

Discussion		
Key results	18	Summarise key results with reference to study objectives
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence
Generalisability	21	Discuss the generalisability (external validity) of the study results
Other information		
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based

*Give information separately for exposed and unexposed groups.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

BMJ Open

How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services? A cross-sectional study

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How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services?

A cross-sectional study

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How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services?

A cross-sectional study

ABSTRACT

Objective: To test the association between self-rated health and self-rated degree of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), and CFS/ME patients' assessment of quality of primary care, specialist care, and coordination of care.

Design: Cross-sectional study.

Setting: Self-reported questionnaire data from women members of The Norwegian ME Association obtained in 2013.

Participants: 431 women with CFS/ME aged 16-73 years.

Main outcome measure: The participants' assessment of quality in primary care, specialist care, and in coordination of care (good/very good or poor/very poor). Main explanatory variables: self-rated health and self-rated degree of CFS/ME.

Results: Quality of care was rated poor by 60.6% in primary care, by 47.7% in specialist care, and by 71.2% regarding coordination of care. Poorer self-rated health increased the probability of rating quality in primary care poor, particularly among women 40 years and over (odds ratio [OR] 2.38, 95% confidence interval [CI] 1.63-3.49), women with university education (OR 2.57, CI 1.68-3.94), and less frequent GP visits (OR 2.46, CI 1.60-3.78). Poorer self-rated health increased the probability of rating quality poor in specialist care (OR 1.38, CI 1.05-1.82), but not in coordination of care. A more severe CFS/ME was associated with higher probability of rating quality in primary care poor (OR 0.61, CI 0.38-0.93). Frequent visitors and those with a long GP relationship were less likely to report primary care quality as poor.

Conclusions: A large proportion of women with CFS/ME rated quality of care poor/very poor in primary care, specialist care, and in coordination of care. The dissatisfaction was higher for primary care than for specialist care. Overall, poorer self-rated health and a more severe CFS/ME were associated with lower quality scores in primary and specialist care, but not in coordination of care. Health care services, as assessed by women with CFS/ME, do have a large potential for improvement.

STRENGHTS AND LIMITATIONS OF THIS STUDY

- Recruitment of a homogenous group of study participants from a patient organization is a study strength
- Use of a systematically tested questionnaire and an acceptable response rate is a strength
- There might be a selection bias due to recruitment from a patient organization, as well as a possible recall bias as for most questionnaire data, which are limitations of the study
- Validity of self-reported measures might be discussed, and in particular measures of disease severity and health
- The cross-sectional design precludes any causal interpretation

How do women with chronic fatigue syndrome/myalgic encephalomyelitis rate quality and coordination of health care services?

A cross-sectional study

INTRODUCTION

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME), is a condition with an estimated prevalence around 1-2 per thousand [1]. It affects women more than men (70-85%), and etiology is unknown [2-4]. CFS/ME is characterized by its fluctuating nature, physical and mental fatigue, persistent post-exertional malaise, sleep disturbances, problems with concentration and memory, pain in muscles and joints, headache, and other symptoms related to cognitive, immune, and autonomous dysfunctions [5-7]. The terms CFS and ME are used interchangeably but mostly as a composite term, and the distinction between them is disputed [5]. In this paper we use the combined term CFS/ME as recommended by Norwegian health authorities and others [7, 8].

CFS/ME cannot be confirmed by specific tests, it is not localized to a specific part of the body, and it challenges the traditional distinction between psyche and soma [9]. In line with this, CFS/ME is a controversial condition and poses particular challenges in terms of diagnosis, therapy, and communication, to patients as well as to physicians [10].

CFS/ME patients often need coordinated health services from multiple providers over time. General practitioners (GPs) are their main contacts. All Norwegian citizens are provided a regular GP, and only 0,4 % of the population has chosen to remain outside GPs' lists [11]. Residents can change GP twice a year without justification. Together with universal tax-funding and gate-keeping, the list system provides strong incentives for personal continuity of care [12]. First line medical services including emergency clinics are run by the municipalities. Specialist services, consisting of hospitals and outpatient clinics, are run by regional health enterprises and are mainly owned by the state. Access is usually achieved by referrals from the GP (the gate-keeper role). GP and specialist outpatient visits are co-payed by adult patients.

Health care services, and GPs in particular, are well regarded in the population [13]. However, Norway's scores in the annual patient/population assessed international comparisons of health systems by the Commonwealth Fund (CWF) have been less than average in areas like general quality of care, waiting time for appointments, information sharing, communication, and coordination of services [14, 15]. Patient satisfaction, defined as "an individual's cognitive evaluation of, and emotional reaction to, his or her health-care experience" [16], is considered an important indication of overall quality in health care [17]. The Norwegian scores, thereby, indicate particular challenge areas for health care providers and policy makers.

Across health care settings, patient satisfaction or patients' assessment of health care quality is positively associated with better self-rated health and functional status, and inversely associated with the complexity of health problems [18-21]. In striving towards

evidence based improvement of health care delivery, research on patient-based assessments of quality and coordination is informative, especially for groups with chronic diseases and long-term needs of care from multiple providers, like CFS/ME. Moreover, this group of patients is interesting due to previously reported serious challenges in collaboration with health care services and providers, in line with the contested status of the condition [9].

The familiar measure self-rated health captures a comprehensive range of aspects and provides summative information about the individual's history of health and disease as well as the current level of health [22, 23]. However, a measure related to the disease of interest might provide a more specific indication of how disease severity and patients' ratings might be associated. Self-rated degree of CFS/ME might be such a measure. Solid evidence whether differences in health status might be associated with CFS/ME patients' assessment of quality has been lacking.

We embarked on this study for three main reasons. First, there is a need to explore how a challenging condition like CFS/ME relates to patients' evaluations. Second, we wanted to explore possible associations between quality assessment and health status as measured both by self-rated health and by a more specific measure related to CFS/ME. Third, we wanted to investigate CFS/ME patients' evaluation of primary and specialist care as separate entities, as well as their evaluation of coordination between care givers.

In the present study we had the opportunity to explore all of these aspects, which is relevant because it may influence GPs', specialists', patients', and policy makers' awareness of assessed quality, with possible consequences for clinical practice, communication, cooperation, health outcomes, and planning and organising of health services for CFS/ME patients.

Our aim was to study how CFS/ME patients rated health care quality and coordination of services, and to investigate whether self-rated health and self-rated degree of CFS/ME were associated with CFS/ME patients' assessment of quality in primary and specialist care, and in the coordination of care.

METHODS

Data

This cross-sectional study used postal survey data obtained in April and May 2013 from members of The Norwegian ME Association. Invitations were distributed by Norwegian Social Science Data Service (NSD) Web Survey to a total of 811 members with known email addresses (about 40 % of all members). Non-respondents were given one reminder.

Initially, we had no information about age or reasons for membership, thus, members were asked to refrain from participating if they were below 16 years or if they did not suffer from CFS/ME themselves (health professionals, parents, others). We do not know how many of the non-respondents were not eligible to participate, therefore, an exact response rate cannot be calculated. Other parts of this study have previously been published [24].

The questionnaire included information about demographic and socio-economic characteristics, health status including specific questions about symptoms, duration, severity, and treatment of their CFS/ME, and use of and experiences with health care services. The

questions used in the current study was based on a questionnaire previously validated and used in an evaluation of GP services in Norway [25], and the revised version was piloted among 143 people belonging to the targeted groups before the final design was settled.

Participants

Women comprised 89.1 % of the 488 respondents. Due to low numbers, as well as to avoid overfitting [26] and a possible confounding effect of gender in the regression models, we excluded all men (53 respondents). We also excluded those who did not give information about gender (2 respondents) or age (2 respondents). This gave a net sample of 431 respondents (Figure 1).

Variables

Participants were asked about their overall personal experiences with quality and coordination of follow-up and treatment in health care services, from their first contact because of CFS/ME, and until the time of the survey. The dependent variables were based on the three questions presented in Table 1. For an easier interpretation of logistic regressions all three variables were dichotomised by merging the original answering options into "good/very good" and "poor/very poor". Those who answered "not relevant" were excluded from the analyses.

Table 1. Female CFS/ME patients' assessment of quality in primary care, specialist care and in coordination of care (%)

	Very good	Good	Poor	Very poor	Not relevant
	n (%)	n (%)	n (%)	n (%)	n (%)
How would you describe the quality of the medical care you have received from primary care? (n=396)	30 (7.6)	114 (28.8)	132 (33.3)	108 (27.3)	12 (3.0)
How would you describe the quality of the medical care you have received from specialist care? (n= 392)	33 (8.4)	128 (32.7)	106 (27.0)	81 (20.7)	44 (11.2)
How do you think the services in the various parts of the support system are coordinated? (n=385)	2 (0.5)	61 (15.8)	152 (39.5)	122 (31.7)	48 (12.5)

The key independent variables were self-rated health and self-rated degree of CFS/ME. Self-rated health was obtained from the question "How would you assess your own health in general?" Response options were reduced from five original categories (very poor – poor - fair - good - excellent) to four by merging the good and excellent categories due to low

numbers (five individuals reported excellent health). Self-rated degree of CFS/ME was obtained from the question "What degree of ME do you have as of today?" Four answering options were given; mild (about 50 % reduction in activity), moderate (housebound most of the day), severe (bedridden most of the day), and very severe (completely bedridden). This classification, defined by an international consensus panel [27], has been widely discussed in the ME Association's membership magazine, and is well known to the members of the ME Association. We merged the severe and very severe categories due to low numbers (five individuals reported very severe disease).

Adjustment independent variables were age, education, duration of the current GP relation (GP duration), and number of GP visits the previous year (GP frequency). Six original education categories were merged into four due to low numbers in the outermost groups (one individual with no education and three individuals with a PhD). GP duration was obtained from the question "Approximately for how long have you had your current GP?" Responses were dichotomised into 0-2 years and 3 years or more. GP frequency was obtained from the question "Approximately how many times have you seen your GP, another GP or visited an emergency clinic during the previous 12 months for issues related to your ME?" The answers were categorized into four levels; 0 visits, 1-4 visits, 5-9 visits, and 10 visits or more.

Analyses

Data was analysed by means of descriptive statistics and logistic regressions. Correlations were tested with Spearman's correlation coefficients.

We constructed two sets of multivariable regression models for each of the three dependent variables. The first set included the independent variables self-rated health, age, education, GP duration, and GP frequency, which were introduced collectively into the model. In the second set of models self-rated health was replaced by self-rated degree of CFS/ME. We performed dummy-analyses as well as trend analyses. Since some of the groups were small, and there was no significant lack of linearity, we chose to report the trend analyses exclusively. We tested first order interactions by introducing interaction terms in the regression models. Where interactions were present, we performed stratified analyses accordingly.

We used 95% confidence intervals (CI) throughout the study. All analyses were accomplished using Stata, version 13.1.

Ethics

The study has been approved by the Norwegian Data Protection Official (id. 31784).

RESULTS

In total 488 members of the ME-association aged 16-73 years (mean age 46.2 years) participated, constituting an overall estimated response rate of 60% (Figure 1). Due to non-response from non-eligible receivers and return of emails from email addresses that were not in use, the actual response rate is assumed to be higher. The 431 women constituting the final

sample for analyses reported having the diagnoses ME (n=354), CFS (n=31) and/or post viral fatigue syndrome (n=70) (more than one diagnosis possible).

Most participants (61.8 %) had suffered from CFS/ME for 10 years or more. The highest percentage of people were aged 40-59 years, had university education, poor self-rated health, moderate degree of CFS/ME, a GP relation of 3 years or more, and 1-4 GP visits the previous year (Table 2). In the previous year, 92% of the patients had visited primary health care services at least once for issues related to their CFS/ME (Table 2).

Table 2. Sample characteristics

	Total sample	
	n	%
Age	431	100.0
16-19	5	1.2
20-39	116	26.9
40-59	244	56.6
60+	66	15.3
Education	398	100.0
Primary	29	7.3
High school	128	32.1
University 1-4 years	156	39.2
University 5 years +	85	21.4
Self-rated health	399	100.0
Very good/excellent	44	11.0
Fair	83	20.8
Poor	205	51.4
Very poor	67	16.8
Degree of CFS/ME	396	100.0
Mild	88	22.2
Moderate	268	67.7
Severe/very severe	40	10.1
GP duration	398	100.0
0-2 years	144	36.2
3 years +	254	63.8
GP frequency*	368	100.0
0 visits	29	7.9
1-4 visits	159	43.2
5-9 visits	107	29.1
10+ visits	73	19.8

* Number of GP visits related to CFS/ME in the previous year

The quality of medical care was assessed poor/very poor by 60.6% in primary care, and by 47.7% in specialist care, whereas 71.2% of the participants regarded coordination between services as poor/very poor (Table 1).

In multivariable analyses we found that the associations between primary care quality assessments and self-rated health were modified by age (interaction term between self-rated health and age, p-value (p)=0.010), education (interaction term between self-rated health and education, p=0.016), and GP frequency (interaction term between GP frequency and self-rated health, p=0.025), indicating a stronger association between poorer quality assessment and poorer self-rated health among women in higher age, with higher education, and less frequent GP visits. In analyses stratified by age, education, and GP frequency, these associations were statistically significant only in women 40 years and over, in women with university education, and in women who visited their GP four times or less during the previous year (Table 3).

Table 3. Female CFS/ME patients' probability of assessing quality poor/very poor in primary care, according to self-rated health (multivariable regression analyses stratified by age, education, and GP frequency).

	Age		Education		GP frequency	
	16-39 years (n=103)	40 years and over (n=245)	Primary/high school (n=137)	University (n=211)	0-4 visits (n=177)	5+ visits (n=171)
	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)	OR for trend (95% CI)
Self-rated health*	1.35 (0.85-2.15)	**2.38 (1.63-3.49)**	1.28 (0.85-1.94)	**2.57 (1.68-3.94)**	**2.46 (1.60-3.78)**	1.34 (0.90-1.99)
Age in years	-	-	1.00 (0.97-1.03)	0.98 (0.95-1.01)	1.02 (0.98-1.05)	0.99 (0.96-1.02)
Education**	1.26 (0.80-1.98)	0.73 (0.52-1.04)	-	-	0.67 (0.45-1.02)	1.33 (0.92-1.95)
GP duration***	0.52 (0.22-1.19)	0.70 (0.38-1.30)	0.88 (0.42-1.85)	**0.49 (0.25-0.97)**	**0.43 (0.19-0.95)**	0.93 (0.50-1.75)
GP frequency ****	0.83 (0.52-1.32)	**0.62 (0.44-0.88)**	**0.55 (0.36-0.84)**	0.75 (0.51-1.10)	-	-

OR odds ratio; CI confidence interval.

* Self-rated health in four groups: 1=very good/excellent, 2=fair, 3=poor, 4=very poor.

** Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.

*** GP duration: 0=0-2 years, 1=>3years.

**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.

Statistically significant findings are marked in bold.

Poorer self-rated health increased the probability of reporting the quality as poor/very poor in specialist care, whereas we made no significant findings regarding coordination of care (Table 4).

Table 4. Female CFS/ME patients' probability of assessing quality poor/very poor in specialist care and coordination of care, according to self-rated health (multivariable logistic regressions)

	Specialist care (n=319)	Coordination of care (n=309)
	OR for trend (95% CI)	OR for trend (95% CI)
Self -rated health*	**1.38 (1.05-1.82)**	1.24 (0.87-1.78)
Age in years	0.99 (0.97-1.01)	1.00 (0.98-1.03)
Education**	1.13 (0.86-1.47)	0.87 (0.61-1.22)
GP duration***	1.06 (0.66-1.71)	0.62 (0.32-1.19)
GP frequency ****	0.95 (0.73-1.25)	1.16 (0.82-1.65)

OR odds ratio; CI confidence interval.

* Self-rated health in four groups: 1=very good/excellent, 2=fair, 3=poor, 4=very poor.

** Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.

*** GP duration: 0=0-2 years, 1=>3years.

**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.

Statistically significant findings are marked in bold.

In models where self-rated health was replaced by self-rated degree of CFS/ME, we found that a more severe CFS/ME increased the probability of rating quality in primary care poor/very poor (Table 5). In this model, those who had the same GP for three years or more, and those who visited more frequently, were less likely to report the quality of primary care as poor/very poor. The effect modifications in the self-rated health model were not replicated in this model. No significant associations in quality reports were observed for specialist care or coordination of care (Table 5).

Table 5. Female CFS/ME patients' probability of assessing quality poor/very poor in primary care, specialist care, and coordination of care, according to self-rated degree of CFS/ME (multivariable logistic regressions)

	Primary care (n=346)	Specialist care (n=317)	Coordination of care (n=308)
	OR for trend (95% CI)	OR for trend 95% CI	OR for trend 95% CI

Degree of CFS/ME*	**1.68 (1.10-2.57)**	1.46 (0.95-2.24)	1.52 (0.87-2.67)
Age in years	1.00 (0.98-1.02)	0.98 (0.96-1.01)	1.01 (0.98-1.03)
Education**	1.05 (0.81-1.37)	1.18 (0.90-1.54)	0.86 (0.61-1.22)
GP duration***	**0.61 (0.38-0.99)**	1.00 (0.62-1.60)	0.61 (0.32-1.17)
GP frequency****	**0.70 (0.54-0.93)**	0.94 (0.72-1.24)	1.14 (0.80-1.64)

OR odds ratio; CI confidence interval.
* Degree of ME in 3 groups: 1=mild, 2=moderate, 3=severe/very severe.
** Education in four groups: 1=Primary, 2=High school, 3=University 1-4 years, 4=University 5 years +.
*** GP duration: 0=0-2 years, 1=>3years.
**** GP visits last year: 0=0 visits, 1=1-4 visits, 2=5-9 visits, 3=10+ visits.
Statistically significant findings are marked in bold.

There were no strong correlations (defined as rho >0.5) between any of the independent variables in any of the models. We found a modest correlation between self-rated health and self-rated degree of CFS/ME (rho=0.5067) but these variables were not both included in any model.

DISCUSSION

We found that primary and specialist care quality was rated as poor/very poor by 60.6 and 47.7 % of study participants, respectively. Poorer self-rated health increased the probability of poor/very poor quality scores both in primary and specialist care. In primary care, these findings were statistically significant among women 40 years and over, among women with higher education, and among women who visited their GP four times or less during the previous year. A more severe CFS/ME was associated with higher probability of rating primary care, but not specialist care, poor. Coordination of care was assessed poor/very poor by most of the study participants, regardless of self-rated health and self-rated degree of CFS/ME. Overall, frequent visitors and those with a long GP relationship were less likely to report poor primary care quality.

CFS/ME patients' assessment of health care services in relation to self-rated degree of CFS/ME is largely unknown, as this measure has hardly been used in previous studies. Self-rated health is a more commonly used but less specific measure as it refers to general health and not specifically CFS/ME-related health. Our finding that poorer self-rated health was associated with lower quality scores confirms with most previous studies across diagnoses and health care settings [18-21, 28]. It is worth noting that similar findings were slightly

weaker for self-rated degree of CFS/ME compared to self-rated health, indicating that self-rated health encompasses a wider range of issues and complexity of health problems [19, 22, 23], even for patients with a complex and challenging condition like CFS/ME. This is also confirmed by a no more than modest correlation between these two variables.

Overall, quality in primary care was more likely reported low by patients with a shorter GP relation and less frequent GP visits. These variables might be intertwined. Previous studies have suggested that continuity of GP care is associated with higher patient satisfaction [24], and that people in poorer health are more likely to have shorter GP relationships [12]. Some of these patients might suffer from ailments that do not fit into specific diagnoses, thus generating dissatisfaction and a search for another GP [28]. In the present study, 36.2 % reported a short duration of their GP relation, indicating that CFS/ME patients might replace their GPs to a higher extent than the general population [12]. In line with our findings, others have reported a positive GP assessment to be associated with increased frequency of attendance [28]. This might indicate that once patients have found an understanding GP, they consider GP visits beneficial and therefore visit more frequently.

In an international comparison of patient-evaluated GP care in 10 European countries, 76 % of Norwegian patients viewed care as good/excellent, a score below the study average [29]. Only 36.4 % of our study participants viewed GP care as good/very good. Despite differences between the studies, both strongly indicate that CFS/ME patients are less satisfied than the general population. This is reinforced by the fact that there are only women in our study, as women in general are more satisfied with health care than men [30]. Regarding the notion that communication and the GP-patient relationship are important tools in the treatment of medically contested conditions like CFS/ME [31], this is a cause for concern.

Slightly more than half of the participants reported the quality of specialist care as good/very good. A Swedish study of outpatient care in all hospital specialties found that on average more than 80 % were satisfied [30], which largely contradicts our findings. Hence, similarly to primary care, CFS/ME patients seem to be less satisfied than patients in general. This is not surprising, since quality of health care is often regarded lower by people in poorer health [18, 19, 21].

We found that quality scores for specialist care were better than for primary care. Previous research report that many GPs are constrained by the scientific uncertainty of CFS/ME [32], unconfident with diagnosing and treating the condition [33], and worried that the label of CFS/ME might be potentially harmful to the patient [34]. Patients with CFS/ME or other medically unexplained conditions on the other hand, have reported feeling belittled, stigmatized, distrusted, rejected and ignored by their doctors, and that their moral character and the reality of their symptoms are questioned [35, 36]. These aspects might partly explain the low quality scores, especially in primary care where doctors are likely to be more skeptical towards the CFS/ME diagnosis than those who have specialized in dealing with them [37]. We reported in a previous paper that patients do value referrals [24], which is in

accordance with patients' reports that specialist services provide acknowledgment of their ailments, treatment, better handling of daily life issues, and improved dialogue between professionals [38]. However, CFS/ME patients often have to struggle for a referral [39, 40], which might affect the relationship with their GP. Patients in Europe have evaluated GP care more positively in countries without gate-keeping [29], and the Norwegian gate-keeping system could explain some of the low primary health care scores.

Quality in coordination of care was rated poor/very poor by 71.2 % of the patients, with no significant differences according to the independent variables in the study. This corresponds to experiences of patients with complex health care needs internationally [41] and with the Norwegian scores in the CWF international comparisons [14, 15]. Despite the Norwegian Coordination Reform (2012), aiming to facilitate better coordination in health care [42], challenges seem large in this field.

Are self-rated health and self-rated degree of CFS/ME to be regarded as measures of individual characteristics or measures of health outcome? This is worth some reflection considering the controversies of this condition, where communication between patient and doctor is regarded as an important treatment tool [31]. Most participants in our study reported poor health and a moderate to severe degree of CFS/ME, thus indicating a complex health situation. The individual-characteristic perspective may indicate that a poor quality of care is influenced by the patients' receptiveness of the offered care, which in turn may lead to blaming the patient herself for a possible poor quality. On the other hand, the outcome perspective might indicate that a poor quality of care is influenced by the health care providers' inability to handle patients and their condition, which in turn may lead to blaming the doctor and health care services for a possible poor quality. In real life these perspectives might be intertwined. However, according to professional ethics and health care laws [43], the doctor is the one responsible for quality in the medical encounter, regardless of patient characteristics and issues raised. Considering health status as (at least partly) a measure of outcome, it is not surprising that poorer self-rated health and a more severe degree of CFS/ME was associated with lower quality scores. In line with this we assume that patients emphasize the outcome perspective. This notion might also be underpinned by the result that the low quality scores for primary and specialist services both were associated with poorer health status, whereas coordination of care was not.

A particular strength of the study was the recruitment of study participants from a patient organization since it is detached from sites of care, with the aim to enable patients to describe their experiences without fear of how this information might affect their relation to health care providers. We used a well-designed systematically tested questionnaire, and the response rate was acceptable. By studying a specific group of patients we have been able to interpret our findings according to a relatively homogenous group.

This study had some limitations. Our sample may not fully represent women with CFS/ME. First, there might be a selection regarding membership of the patient organization.

Survivor bias may be a part of this, indicating that the healthiest patients will not demand membership to the same extent as those in poorer health [44]. On the other hand, the most seriously affected members might have refrained from participating because of disease severity. The direction of a possible selection bias from these factors is not obvious. A general population satisfaction study reported that non-respondents were overrepresented in groups with lower satisfaction [30], indicating that a possible selection bias might have skewed our study in the direction of better satisfaction scores than would otherwise be found. However, female CFS/ME patients might differ from this population [36]. Second, the distribution of e-mail addresses might have been skewed, for instance towards younger members with higher education. However, since 93 % of Norwegian households have access to the internet [45] we find it unlikely that this have influenced our results to a significant degree. Third, our sample had lower age and higher education than the Norwegian average [46]. A possible skewness regarding these variables might be connected, since younger individuals will not have completed their education.

In questionnaire data there is always a potential for recall bias, particularly regarding minor events and distant past, usually leading to underreporting. Some studies indicate that doctors hold the opinion that CFS/ME patients often exaggerate the severity of their ailments [36], and it is difficult to judge whether overreporting or underreporting might be present in our data. The validity of self-reported data on disease severity may be disputable per se, but in the case of CFS/ME, where no objective tests are diagnostic or suitable to evaluate disease development and severity, self-rated degree of ME might actually be a strong contender as the golden standard for describing disease severity.

Self-rated health and self-rated degree of CFS/ME describes the health status at the time of the survey, whereas participants assess the quality of their care from the first onset of their symptoms. This might be a problem, in particular if self-rated health and self-rated degree of CFS/ME, with its fluctuating nature, are considered individual characteristics. However, we have argued that patients most likely consider these variables as outcome measures, and the difference in observation time might thus lack significant impact. Besides, self-rated assessments of health status might not be solely limited to the current status of health or CFS/ME [22, 23]. The questionnaire wording may draw in this direction for self-rated health, as it emphasizes health status more generally ("How would you assess your own health in general?"). However, for self-rated degree of CFS/ME the current situation is emphasized to a greater extent ("What degree of ME do you have as of today?"). All in all, it remains unclear whether this might have affected the assessments. Most likely it has not had any significant impact on our main results.

The cross sectional study design implicates that no causal relationships can be established. For future research we would recommend a longitudinal design investigating factors relevant to patients' quality assessment over time.

CONCLUSIONS

We concluded that a large proportion of women with CFS/ME rated the quality of their care poor or very poor for primary care, specialist care, and coordination of care. The dissatisfaction was higher for primary care than for specialist care, and even higher for coordination of care. Poorer self-rated health and a more severe degree of CFS/ME were associated with lower quality scores, particularly in primary care services, but were not associated with coordination between services. The findings indicate that quality in health care services, as assessed by patients with CFS/ME, do have a significant potential for improvement. In order to achieve this, health care services must recognise and acknowledge the voice of the users, which is a fundamental value in all medical practice as well as a precondition for congruence in doctor-patient relationships and shared decision-making. This is particularly important when the consultation is likely to provide neither an explanation nor a remedy.

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CONTRIBUTORSHIP STATEMENT

Both the authors contributed to the design and conduct of the study. OSL designed the questionnaire, provided funding and collected the data. AHH undertook the statistical analyses and drafted the manuscript. OSL contributed with major improvements and critical revisions. Both the authors approved the final version for publication.

COMPETING INTERESTS

The authors declare that they have no competing interests.

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DATA SHARING STATEMENT

No additional data available.

FIGURES

Figure 1

Flow chart of study participants.

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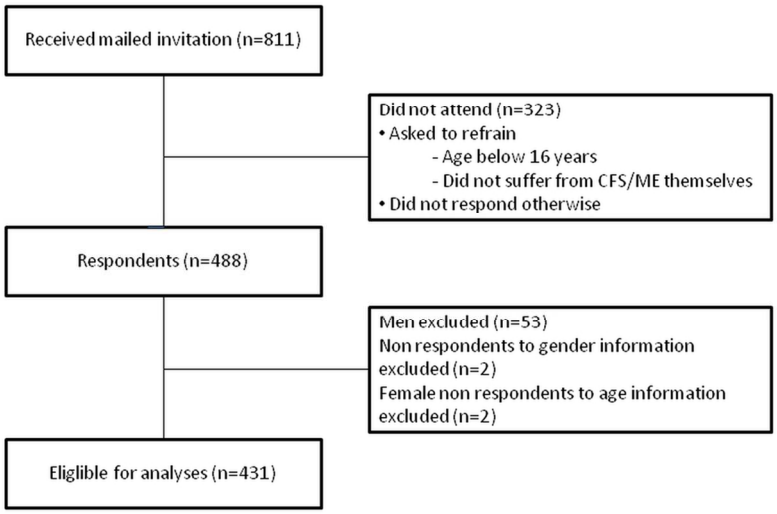

Figure 1. Flow chart of study population.
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