Accepted Manuscript

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PII: S0022-3999(16)30349-X
DOI: doi: 10.1016/j.jpsychores.2016.07.008
Reference: PSR 9185

To appear in: *Journal of Psychosomatic Research*

Received date: 17 November 2015
Revised date: 12 July 2016
Accepted date: 15 July 2016

Please cite this article as: Mallet Miriam, King Eleanor, White Peter D., A UK based review of recommendations regarding the management of chronic fatigue syndrome, *Journal of Psychosomatic Research* (2016), doi: 10.1016/j.jpsychores.2016.07.008

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A UK based review of recommendations regarding the management of chronic fatigue syndrome

Short title: Recommendations for management of CFS

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Keywords: Chronic fatigue syndrome, myalgic encephalomyelitis, management, treatment, recommendations

Word count 1490

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Highlights:

- There were marked discrepancies between sources
- Patient organisations preferentially recommended medicines, pacing and complementary treatments
- Medical organisations recommended rehabilitation therapies

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ABSTRACT

Objectives : Chronic fatigue syndrome (CFS) is a controversial illness, with apparent disagreements between medical authorities and patient support organisations regarding safe and effective treatments. The aim of this study was to measure the extent of different views regarding treatments, comparing patient support organisations and medical authorities in the UK.

Methods: Two independent raters analysed two groups of resources: UK patient support websites and both medical websites and textbooks. A 5-point Likert scale was developed with the question 'With what strength does the source recommend these treatments?' The various treatments were divided into the following four groups: complementary and alternative medicine (CAM), pharmacological, rehabilitative, and pacing therapies.

Results: There were significant differences between the scores for patient support organisations and medical authorities for all 4 treatment groups. The results for supporting CAM were 74% (patient group) vs 16% (medical authority) ($p<0.001$), 71% vs 42% for pharmacological ($p=0.01$), 28% vs 94% for rehabilitative ($p<0.001$) and 91% vs 50% for pacing treatments ($p=0.001$).

Conclusions: There were substantially different treatment recommendations between patient support organisations and medical authorities. Since expectations can determine response to treatment, these different views may reduce the engagement in and effectiveness of rehabilitative therapies recommended by national guidelines and supported by systematic reviews.

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Introduction

Chronic fatigue syndrome (CFS), also named myalgic encephalomyelitis (ME), is a condition that still creates debates regarding definition, aetiology, diagnosis and treatment [1]. The condition is found world-wide, with a meta-analysis suggesting a prevalence of 0.76% [2]. Treatment trials have focused on rehabilitation therapies [3]. People with CFS/ME can receive conflicting treatment options from medical sources and patient support organisations.

The National Institute of Health and Care Excellence (NICE) guidelines on CFS/ME [4] have been subject to criticisms from some patient organisations on the basis that they consider the main treatments recommended by NICE, namely cognitive behaviour therapy (CBT) and graded exercise therapy (GET), as either unsafe or unhelpful. [5,6]. Some CFS/ME patient support organisations have claimed that NICE has overlooked the physical component of the illness [6]. Of relevance to this, Hossenbaccus and White found that patient support organisations considered CFS/ME to be a physical disease, whereas medical authorities were more likely to consider CFS/ME as being a combination of physical and psychological factors [7].

The UK based PACE trial agreed with the NICE guidelines [4], finding that CBT and GET, when combined with specialist care, were safe and effective treatments for CFS/ME, whereas pacing was ineffective [3]. Some patient support organisations have continued to promote a pacing approach [5, 7, 8, 9].

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Stories about CFS/ME are often reported in the media and online forums are expanding [1, 7, 10]. For illnesses such as CFS/ME, where recommended treatments are not universally supported, patients can search for alternative treatments through forums and patient organisations.

Knudsen et al [1], found that newspaper articles in Norway were generally positive towards alternative treatment and negative towards evidence-based treatments (mainly CBT and GET); patients preferred alternative treatments; physicians favoured evidence based treatments; and the Norwegian ME association was critical of evidence-based treatments [1].

We are not aware of any published studies of views of treatments comparing patient support groups and medical authorities. This study aimed to determine whether there were significant differences between which treatments were recommended, or discouraged, between these two groups. We predicted that patient groups would favour pharmacological, complementary and alternative (CAM) treatments, and pacing, whereas medical sources would prefer rehabilitative based therapies.

Methods

A database was created using both internet searches and a catalogue of CFS/ME patient organisations previously used in the study by Hossenbaccus and White (see supplementary material) [7]. The medical sources group was comprised of websites from medical Royal Colleges, the UK National Health Service, NICE, the Department of Health and British United Patients Association, together with textbooks found in

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the medical school library of Barts and the London. The appendix contains all sources reviewed.

The different types of treatment were grouped under the following general headings: Complementary and alternative medicine (CAMs), pharmacological treatment, rehabilitative based therapies (CBT and GET) and pacing.

The primary question used for scoring of each source was as follows, 'With what strength does the source recommend these treatments?' After a pilot study, a five point Likert scale guided the scoring. A score of 5 was given if the source was strongly supportive, stated a benefit or that they recommended the treatment. A score of 4 indicated being moderately supportive, or specifically recommended for symptomatic relief, which particularly applied to pharmacological treatment. A score of 3 was given if the treatment was mentioned in the source, but there was no indication whether the authors supported or rejected the treatment. A score of 2 indicated that the author was moderately unsupportive of a treatment. A score of 1 indicated that the source was strongly against or clearly not recommending the treatment. Each individual treatment mentioned was scored individually, and a composite mean calculated as a final score for each treatment. All sources were reviewed in 2015. Ratings were made independently by two raters, and any discrepancies in scoring were discussed and re-evaluated consensually.

The analysis of this study was done using the SPSS software version 22, using the agreed mean scores. An inter-rater reliability analysis was undertaken, using intra-class correlations for interval data. Frequencies were assessed and quartiles and

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median values derived for each treatment group for the two sources, since the data were not normally distributed. Non-parametric Mann-Whitney U tests were then carried out to compare scores from the two sources.

Results

The total number of sources used for analysis was 95, with 60 medical authorities and 35 CFS/ME patient support organisations surveyed. Not all sources mentioned every treatment group; so numbers of sources for each treatment group were 48, 65, 81 and 38 for CAM, pharmacological, rehabilitative and pacing respectively. The two raters sought and achieved consensus for 37 mean scores out of 380 (10%). The intra-class correlation between independent raters' scores was 0.96 ($P < 0.001$) with a median (quartiles) difference in scores, when one was present, of 0.3 (0.025, 1.0) points.

Frequencies for the different treatment groups for CFS/ME patient support organisations and medical authorities are shown in table 1 and further demonstrated in figure 1. Here, a positive score is an agreed mean score greater than 3, a negative score is less than 3 and neutral is a score of 3.

Descriptive statistics of the data for CFS/ME patient support organisations demonstrated the median (quartiles) for CAM, pharmacological, rehabilitative and pacing treatment groups to be 4.0 (3.0, 4.2), 4.0 (3.0, 4.3), 3.0 (1.5, 3.5) and 5.0 (4.0, 5.0) respectively, and 1.5 (1.0, 3.0), 2.9 (2.0, 4.0), 4.5 (4.0, 5.0) and 3.3 (2.3, 4.0) for the medical authorities respectively. There were significant differences in the scores between patient support organisations and medical authorities for every treatment

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group (CAM $p < 0.001$, pharmacological $p = 0.011$, rehabilitative $p < 0.001$ and pacing $p = 0.001$).

Discussion

There were significantly different recommendations between CFS/ME patient support organisations and medical sources across all four treatment groups. Whilst patient support organisations favoured CAMs, pharmacological and pacing therapies, about half were against rehabilitative therapies. The medical authorities viewed CAMs negatively, and recommended rehabilitative treatment.

These findings support our hypotheses and support the finding from Knudsen et al. [1]. Together with the study by Hossenbaccus and White [7], this study provides an insight into the reasons for these discrepancies; patient support organisations seem to prefer treatments that reinforce the physiological or physical view of CFS/ME, and reject the more behavioural therapies contained within the rehabilitative group. The reticence of patient organisations to recommend rehabilitative therapies may also be related to instances of poor delivery of these therapies [8]. It should be remembered that the moderate success of behavioural approaches does not imply that CFS/ME is a psychological or psychiatric disorder. Such dualistic beliefs should have no place in modern medical understanding of conditions such as CFS/ME [7, 10, 11].

In this study it was possible to score a large number of sources, including all available UK based CFS/ME patient support organisations that had a website, and a large number of medical textbooks and websites. The high intra-class correlation between

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independent rating scores suggests a reliable scoring method, although raters with different backgrounds may have rated sources differently.

Limitations included the subjective nature of analysing language, as well as minor variations between the independent rater scores. Furthermore, the raters were unable to be blinded to the source. Both raters were medical students, and it may be that other raters, such as patient group members, might have obtained different scores. There were also fewer patient support organisations sources (N=35), compared to medical authorities (N=60).

The findings from this study inform healthcare professionals about what types of treatments are commonly recommended by patient support groups on the internet, and that this may conflict with established treatment guidelines. This may additionally have an effect on the response and expectations of patients; Cho et al [12] demonstrated that CFS/ME patients respond to the treatment that best reflects their views on the illness. CFS/ME is almost unique within medicine in having such a marked discrepancy of views between patient organisations and medical authorities, although another example is chronic Lyme disease [9, 13]. These discrepancies are often based on different understanding of the concepts of illness and disease [14], as well as using different language to describe similar approaches, such as baseline setting and pacing [8].

Further investigations into the reasons for the differences in opinion might help reduce the discrepancies regarding treatment recommendation from different sources

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[8]. Research in countries with different healthcare systems would further provide insight into whether these discrepancies are world-wide.

In conclusion, although much is still in dispute in regards to CFS/ME, it is undoubtedly a debilitating illness, and clinicians need to be able to provide treatment as well as advice regarding whether to join a patient organisation [4]. Work needs to be done to establish a common understanding regarding treatment recommendations between medical sources and patient support organisations in order to provide consistent advice to patients about the most effective and safest treatments available.

Conflicts of Interest

PDW is an appointed member of the Independent Medical Experts Group, which advises the UK Ministry of Defence regarding the Armed Forces Compensation Scheme. PDW provides advice to the Department for Work and Pensions and does consultancy work for a re-insurance company. The other co-authors declare no conflicts of interest.

Acknowledgements

We are grateful for feedback on a previous version of this paper.

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Figure 1

Frequencies of views for each treatment group

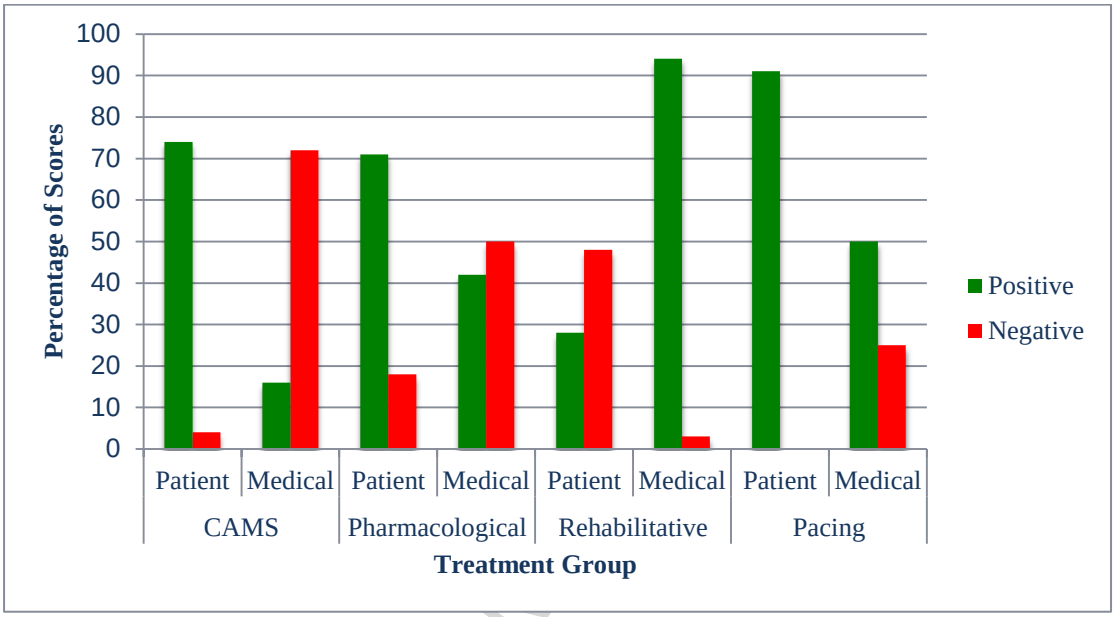

Fig 1: Frequencies of positive and negative opinions for each treatment group. Neutral results have been omitted.

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Table 1
Frequencies of views for each treatment group

Treatment Group	Organisation	Positive	Neutral	Negative
		Frequency (%)	Frequency (%)	Frequency (%)
CAM	Patient	17/23 (74)	5/23 (22)	1/23 (4)
	Medical	4/25 (16)	3/25 (12)	18/25 (72)
Pharmacological	Patient	12/17 (71)	2/17 (11)	3/17 (18)
	Medical	20/48 (42)	4/48 (8)	24/48 (50)
Rehabilitative	Patient	6/21 (28)	5/21 (24)	10/21 (48)
	Medical	56/60 (94)	2/60 (3)	2/60 (3)
Pacing	Patient	20/22 (91)	2/22 (9)	0/22 (0)
	Medical	8/16 (50)	4/16 (25)	4/16 (25)

CAM = Complementary and Alternative Medicines. A positive score is a mean score greater than 3, negative is less than 3 and neutral is a mean score of 3.

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