2023-10-31

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Original Article

SAGE Open Medicine

SAGE Open Medicine
Volume 11: 1–7

DOI: 10.1177/20503121231207207
journals.sagepub.com/home/smo
Sage

Body reprogramming for fibromyalgia and central sensitivity syndrome: A preliminary evaluation

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Abstract

Objectives: Central sensitivity syndrome disorders such as fibromyalgia, provoke continued debate, highlighting diagnostic and therapeutic uncertainty. The Hyland model provides a way of understanding and treating the medically unexplained symptoms of central sensitivity syndromes using complexity theory and principles of adaption in network systems. The body reprogramming is a multi-modal intervention based on the Hyland model designed for patients living with medically unexplained symptoms. This preliminary, naturalistic and single-arm service evaluation set out to evaluate outcome after attending a body reprogramming course in patients living with fibromyalgia or central sensitivity syndrome.

Methods: Patients diagnosed with fibromyalgia or central sensitivity syndrome were recruited. The body reprogramming courses consisting of eight sessions, each 2.5 h in length, were run at two study sites in England. Data were collected at baseline, post course and 3-months post course using questionnaires assessing symptomatology (FIQR/SIQR), Depression (PHQ9), Anxiety (GAD7) and quality of life (GQoL). Repeated measures *t*-tests were used, and all comparisons were conducted on an intention to treat basis.

Results: In total, 198 patients with a mean age of 47.73 years were enrolled on the body reprogramming courses. Statistically and clinically significant improvement were observed in the FIQR from baseline to post course (mean change: 11.28) and baseline to follow-up (mean change: 15.09). PHQ9 scores also improved significantly from baseline to post course (mean reduction 3.72) and baseline to follow-up (mean reduction 5.59).

Conclusions: Our study provides first evidence that the body reprogramming intervention is an effective approach for patients living with fibromyalgia or central sensitivity syndromes on a variety of clinical measures. Besides these promising results, important limitations of the study are discussed, and larger randomized controlled trials are clearly warranted.

Keywords

Fibromyalgia, central sensitivity syndromes, adaptive network theory, Hyland model, body reprogramming

Date received: 21 February 2023; accepted: 26 September 2023

Introduction

Conditions presenting with pain as a dominant symptom continue to pose a diagnostic and therapeutic challenge. Diagnostic nomenclature appears confusing, with overlapping clinical usage of terms including central sensitivity syndromes (CSSs),[1] functional disorders, chronic primary pain, body distress syndrome, unexplained symptoms, somatic symptom disorder[2] in addition to the more established diagnosis of fibromyalgia.

The recently introduced International Classification of Disease-11 (ICD-11) has placed 'persistent pain' within a separate diagnostic within the sub-category, chronic primary pain used for pain disorders without an established cause.[3] However, the focus on pain-related nomenclature appears to

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minimise the significance and relevance of other widespread symptoms associated with these central sensitivity sydromes.[4]

Fibromyalgia is a potentially debilitating condition with a prevalence of 2%–5% depending on the criteria used,[5] resulting in a significant economic, emotional and clinical burden.[6,7] Both patient and healthcare provider often find it difficult to describe, understand and subsequently manage fibromyalgia.[8,9] Widespread pain is accompanied by multiple other symptoms including but not limited to post-exertional malaise, non-refreshing sleep pattern, perceptual sensitivities, temperature dysregulation and cognitive issues.[10]

There has been growing recognition that medical conditions including fibromyalgia, chronic fatigue syndrome/myalgic encephalomyelitis, irritable bowel syndrome, interstitial cystitis and others, share comorbid symptoms despite having a different primary body location focus.[11] The term 'central sensitivity syndrome' has evolved as an umbrella label for these conditions, where 'pain' is a dominant symptom, and 'central sensitisation' considered a core mechanism.[12]

In addition, there is often societal and medical scepticism that this patient group have a 'real' illness.[13,14] These patients lack a conceptual and evidence-based model that allows them to both understand and share their experience. The provision of a plausible and evidence-based rationale is one of the key mechanisms for interventions to be beneficial. Importantly, any treatment takes place in a context of care[15] where – besides the specific method of treatment – the plausible rationale,[16] the interpersonal physician–patient relationship,[17] patients' expectations,[18] as well as the motivational concordance,[19] are significant mechanisms. This set of components have also been named as common factors.[20]

Common factors are often a neglected part in treatments and there is also limited healthcare support with the chronicity of the condition.[21] Amplification of debility can result from these increased physical and emotional stressors, impacting further on adverse quality of life.[19] Disturbed sleep, negative cognition, social isolation, depression and anxiety are also common in fibromyalgia, further complicating the management of the syndrome.[22]

Treatment strategies should be ideally modelled on a conceptual and evidence-based understanding of the condition. Unfortunately, in fibromyalgia there is no universally agreed conceptual model to guide therapeutic direction.[23] International management guidelines for fibromyalgia typically advocate a multidisciplinary approach, recommending individualised considerations of pharmacological and non-pharmacological modalities.[24,25] In practice, symptom reducing pharmacological options, directed primarily at possible neuropathic pain origin,[26] also typically have a weak evidence base when used in CSSs with trials providing only short-term outcome data.[27] For non-pharmacological interventions, advice is often limited to a generic recommendation to exercise and maintain activity but little clarity as to how this is best addressed.[28] Psychological interventions such as cognitive behavioural therapy (CBT) are sometimes utilised, but effect size is modest and there can be recipient frustration with their recommendation.[29,30]

Conceptualisation models based on complexity theory have more recently been suggested for understanding CSSs such as fibromyalgia.[31] One such model is the Hyland model, which is based on the principle of a comprehensive complex adaptive framework within a network system. The model argues that symptom-causing mechanisms are causally connected, forming a network that has emergent properties.[32]

The Hyland model introduces the theoretical constructs of 'stop signals' and 'stop programs' within the framework of this integrated mechanism.[31] Stop signals are considered as adaptive symptom clusters, triggered by either biological or psychological events, designed to change and typically inhibit behaviour. Sometimes people fail to modify behaviour and respond to the stop signals due to ongoing physical, cognitive or emotional stressors. Over time, failure to respond to stop signals causes adaption within the network and a gradual potentiation of the stop signals so the symptoms 'shout louder' becoming eventually fixed in a heightened state of a 'stop program' which drive the symptoms described by patients with CSS. A layered, metaphorical narrative has been constructed from this adaptive network theory incorporating the analogy of a sophisticated computer.[33]

The body reprogramming (BR) therapeutic approach for fibromyalgia is designed primarily from the Hyland model.[33] It is multi-faceted, developed in partnership with patients and consistent with non-pharmacological evidence-based guidelines.[34] There are parallels to the recovery model with the focus on greater patient understanding of the condition and facilitating patients to take control of their own recovery rather than awaiting 'hardware medical fixes'.

The aim of this preliminary, naturalistic, and single-arm service evaluation was to assess the impact of the BR approach for patients with fibromyalgia and CSS. To do this we conducted within-group comparisons on patient-reported outcomes (PROs) collected at pre and post course. We also investigated if any benefits from the course persisted by collecting the same PROs at 3-month post course.

Methods

Design

Data were collected as part of a clinic service evaluation into the impact of introducing a BR group course for patients with fibromyalgia and CSSs.

Participants

Patients aged ⩾18 years with fibromyalgia or CSS diagnosed by their primary care medical practitioner or hospital

specialist who were enrolled on group community-based BR courses run by the Plymouth Pain Management Service or Cornwall Partnership NHS Foundation Trust.

Patients were ineligible to participate in a BR group if they had other medical conditions requiring active specialist input, such as autoimmune disorders, major psychiatric illness or substance abuse disorder. Further exclusion criteria included having insufficient English to benefit from a group intervention, non-acceptance of their CSS diagnosis or unwilling to take part in a group-based therapeutic intervention.

Questionnaires

The primary outcome measure to assess severity of symptomology was the fibromyalgia impact questionnaire (revised) (FIQR),[35] or its identical content equivalent the symptomology impact questionnaire (SIQR). The latter title to the questionnaire is recommended by the original authors when there is no specific reference to the diagnosis of fibromyalgia, thus minimising confusion in those who have been provided an alternative diagnosis such as a CSS.[36] Scores range from 0 to 100 with higher scores signifying greater impairment a $\geqslant$60 indicating severe symptomatology.[37]

The FIQR/SIQR consist of three domains (function, overall impact and symptoms) providing a validated outcome measure on the clinical severity of the condition. The minimum clinically important difference (MCID) of 8.1 was used as the threshold for a clinical meaningful change in symptomology.[38]

Secondary outcome measures assessed:

Depression with the patient health questionnaire-9 (PHQ-9) which is scored from 0 to 3 (not at all – everyday). A total of the scores is calculated to provide an aggregate score. Higher score indicates greater severity: 5–9 mild, 10–14 moderate, 15–19 moderately severe, 20–27 severe.[39,40]

Anxiety with the generalised anxiety disorder-7 questionnaire (GAD-7),[41] which is scored from 0 to 3 (not at all – nearly half the day). The scores from the seven questions are totalled to produce an overall score. Higher scores indicate worse anxiety: 5–9 mild, 10–14 moderate, 15+ severe.

Overall quality of life (QoL) was assessed with the global quality of life questionnaire (GQoL),[42] a 0–100 (no QoL – perfect QoL) category rating scale based on a Borg scale. It should be noted that this scale was introduced into the pain management service as a routine service evaluation measure part way through the courses.

Details on how to obtain the questionnaires used in this study can be found in the Supplemental Material.

Procedure

This clinical service evaluation was undertaken at the two service sites between November 2017 and January 2020. The BR course consisted of eight sessions, each lasting 2.5 h per session. One session was run per week and were held in a community setting for eight consecutive weeks. The therapeutic approach used is derived from the conceptual framework called the Hyland model which is based on adaptive network theory. Further details of the therapeutic principles incorporated, can be found in the Appendix. The courses were led and delivered by two senior allied health care professionals; either Clinical Psychologists or Clinical Specialist Physiotherapists with experience in managing chronic pain conditions. The format of the course can be found in the Supplemental Material.

Patients were asked to complete all questionnaires at the start of the course (baseline), at the end of the course (post course) and at a 3-month follow-up.

Approvals

Clinical service evaluation approval as granted by University Hospitals Plymouth NHS Trust (Reference: CA_2016-17-151). The institutional review board confirmed that this study was a clinical service evaluation using the service-based routine outcome questionnaires and all data anonymised. The requirement for written informed consent was waived by the institutional review board.

Statistics

Questionnaire scores are presented as descriptive statistics on an intention to treat basis. To evaluate the effect of the course on our four outcome measures, questionnaire scores were compared at two time points: 1. Baseline to post course, 2. Baseline to three-month follow-up. Repeated measures *t*-tests were used, and all comparisons were conducted on an intention to treat basis (i.e. all those that provided PRO data at relevant time points were included in the analysis).

To investigate change in questionnaire scores after course completion, we also compared post course questionnaire scores to 3 months follow-up.

Results

One hundred and ninety eight patients were enrolled on the BR course, 144 from Plymouth and 54 from Cornwall. Nine patients did not provide baseline data. Demographic and disease duration data can be found in Table 1.

Course attendance

Twelve patients only attended one to two sessions (6%), 18 attended three to four sessions (9%), and 35 attended five to six sessions (18%). One hundred twenty-eight patients (65%) enrolled on the course attended seven sessions or more. Despite being enrolled on the course, five patients (3%) did not attend any sessions or contribute any PROM data.

Table 1. Demographic information for all patients (*n*), *standard deviation*.

Mean	
Age	46.73 (198) *12.93*
Sex (%)	
Females	88% (175)
Males	12% (23)
Duration of symptoms (months)	102 (177) *105.45*
Time since diagnosis (months)	46 (174) *77.41*

Italicised values are standard deviation.

Table 2. Questionnaire scores (*n*) and *SD* at three time points.

Questionnaires	Baseline	Post course	Follow-up
FIQR/SIQR#	74.35 (189) *15.62*	62.59 (135) *17.9*	58.68 (81) *20.43*
PHQ9#	18.59 (184) *5.54*	14.42 (134) *5.68*	12.88 (81) *6.49*
GAD7#	14.13 (187) *5.5*	10.25 (137) *5.59*	10.03 (79) *5.23*
GQoL	37.41 (141) *20.55*	48.29 (99) *17.19*	54.81 (57) *17.43*

Italicised values are standard deviation.
#Lower score indicates improvement.

See Table 2 for the questionnaire scores provided by all patients at the three time points. In total, 158 of 189 patients that contributed FIQR/SIQR data (84%) scored ⩾60, indicating severe symptomology, 89 of 184 patients (48%) scored 20–27 on the PHQ9 indicating severe symptoms of depression, 100 of 187 patients (53%) scored ⩾15 on the GAD7 indicating severe symptoms of anxiety.

Baseline versus post course

Paired samples *t*-tests were used to compare baseline and post course questionnaire scores.

There was a significant improvement in FIQR/SIQR scores from baseline ($M=74.07$, SD = 15.10) to post course ($M=62.79$, SD = 17.81); $t(129)=8.42$, $p<0.001$. A reduction of 11.28 in the FIQR/SIQR surpasses the questionnaire's MCID.

Significant improvements were also observed in the:

PHQ9: Baseline ($M=18.19$, SD = 5.53) to post course ($M=14.47$, SD = 5.72); $t(126)=7.76$, $p<0.001$. PHQ9 scores indicate patients moved from moderately severe depression to moderate depression.

GAD7: Baseline ($M=14.05$, SD = 5.43) to post course ($M=10.30$, SD = 5.58); $t(131)=8.33$, $p<0.001$. At baseline, GAD7 scores indicated that patients experienced moderate anxiety. No meaningful cut points for GAD7 scores were crossed by post course.

GQoL: Baseline ($M=39.84$, SD = 20.85) to post course ($M=48.54$, SD = 16.95); $t(94)=-5.03$, $p<0.001$.

Baseline versus follow-up

Paired samples *t*-tests were used to compare baseline and questionnaire scores collected at follow-up.

There was a significant improvement in FIQR/SIQR scores from baseline ($M=73.46$, SD = 15.05) to follow-up ($M=58.42$, SD = 20.41); $t(79)=7.24$, $p<0.001$. A reduction of 15.09 surpassed the MCID of the FIQR.

Significant improvements were also observed in the:

PHQ9: Baseline ($M=18.58$, SD = 5.44) to follow-up ($M=12.99$, SD = 5.72); $t(77)=7.45$, $p<0.001$. The PHQ9 scores indicate that patients moved from moderately severe depression to moderate depression.

GAD7: Baseline ($M=14.21$, SD = 5.19) to follow-up ($M=9.92$, SD = 5.16); $t(76)=7.61$, $p<0.001$. The GAD7 scores indicate that patients moved from moderate anxiety to mild anxiety.

GQoL: Baseline ($M=38.75$, SD = 20.05) to follow-up ($M=55.25$, SD = 16.77); $t(54)=-6.62$, $p<0.001$.

Post course change

None of the questionnaire scores showed significant worsening from post course to follow-up 3 months later ($p>0.05$).

Discussion

The BR therapeutic group-based course is based on a novel conceptual model for CSSs. This evaluation of its therapeutic introduction within two clinical settings offers preliminary evidence of clinical benefit in symptom interference, anxiety, depression, and overall QoL for patients with CSSs including fibromyalgia. We observed clinically meaningful reductions in anxiety and symptom burden as well as increases in QoL. All improvements were sustained 3 months after course completion.

This is a proof-of-concept study to explore initial validation for the approach and to inform its continued development. It is a naturalistic study representative of patients within two health care settings in the Southwest of England receiving this novel therapeutic intervention in clinical practice. The intervention has been conducted by healthcare professionals in a community setting using relevant outcome measures to inform healthcare treatment decisions.

The study included a broadly inclusive population, representative of patients referred within day-to-day clinical practice at two healthcare centres rather than being conducted on a highly selective population within the remit of a randomised controlled trial (RCT). Real world evidence and RCT's should be mutually complementary with the former

helping to inform the latter. As well as determining early feasibility of the therapeutic approach it can support adequate statistical powering within a subsequent RCT study as well as assisting in the patient selection process.

Key patient selection issues include the potential stratification of severity of the illness, as well as consideration of patients with a concomitant mental health issue such as a moderately severe depressive illness. The patients assessed within this study displayed a high mean FIQR/SIQR (74.35) at baseline with 84% of those providing FIQR/SIQR data scoring 60 or greater (severe range) at the start of the study.[37] The data suggest that the BR approach is appropriate for patients suffering from severe symptomology where therapeutic options available to health care professionals, particularly relating to exercise will be more limited.[43]

In addition, patients had high levels of depression and anxiety, with 48% and 53% scoring in the upper ranges on the PHQ9 and GAD7 respectively. Research has demonstrated that patients with fibromyalgia displaying comorbid mood disorders and anxiety tend to have poorer overall outcomes.[44] Further to this some recent research has suggested that fibromyalgia patients with moderately severe or worse depression should be considered for exclusion from multicomponent therapy until mood is stabilised.[45] The current data would dispute this and indicates that the BR approach can have a positive impact on mood issues and thus would not be a contraindication to inclusion.

Comparing this study to others is difficult as not all use the FIQR/SIQR. A systematic review and meta-analysis of mindfulness-based stress reduction for fibromyalgia reports data from six studies meeting their eligibility criteria, three of which include FIQR data.[46] An 8-week course of mindfulness-based mediation plus Qigong reported a significant reduction of 11.4 in the FIQR at 24 weeks follow-up.[47] One study of stress reduction – cognitive behavioural treatment (SR-CBT) delivered over 10 weeks did not find a significant improvement in FIQR scores, and the third used mindfulness-based stress reduction but did not report change in FIQR scores post intervention. A more recent study of tai chi for fibromyalgia reported a significant reduction of 14.7 in the FIQR after 12-weeks of once weekly tai chi and 25.4 points after 24 weeks of 2-times weekly tai chi.[48] These improvements are comparable to the reduction in the FIQRE/SIQR of 15.09 observed in this present study at 3-month follow-up.

Two linked national surveys profiling UK healthcare services for those with fibromyalgia (PACFiND study) recently highlighted the need for innovative therapeutic models within primary and community care.[49] A systematic review in 2020 failed to identify any evidence-based care models that traversed the entire healthcare system.[50] The development of a robust conceptual model successfully operationalized within the environment of a 'troublesome label', heterogenous symptom profile and difficult patient–provider relationship, is perhaps the 'holy grail' of this difficult clinical arena.[51,52] The translational methodology incorporating the initial 'Hyland' conceptual model and subsequent BR intervention, offers a potential vehicle, both for framing the explanatory narrative and providing a coherent, integrated, therapeutic approach for CSSs such as fibromyalgia.

The BR approach has the potential as a unified framework for the delivery of care aligned to CSSs. This should impact on the consistency of usage of effective healthcare resources. Also, it will nurture healthcare professional confidence in their programme delivery as well as optimising engagement with the most important stakeholders, namely the individuals suffering from this clinically challenging condition.

Limitations of study

Data for this study was collected as part of a clinical service evaluation and normal clinical practice. As such, no formal sample size calculation was conducted. Future investigations of this intervention will be conducted as formal research studies, with a sample size calculation and a suitable control group such as current clinical practice, to determine the relative benefit of BR.

CSSs are not exclusion diagnoses but often co-exist and can potentially be triggered by other clinical conditions.[53] This raises the possibility of a potential difference in therapeutic impact of the intervention for those with a primary or idiopathic fibromyalgia versus those with a secondary fibromyalgia occurring in association with underlying disorders such as rheumatoid arthritis.[54] We did not collect data on whether patients had primary or secondary fibromyalgia. Thus, analysis to determine if BR impacted these two patient groups differently was not conducted.

We report sustained benefit on the FIQR/SIQR, PHQ9 and GAD7 as well as increased gain on the GQoL at 3 months. It is not possible to determine if the observed benefits persisted past this time point as no further data were collected. Maintaining improvement is a known challenge in the management of fibromyalgia and CSS.[34] Therefore, future evaluations of BR need to collect data over a longer time period to determine if these improvements are sustained.

Despite course attendance remaining high with 65% of enrolled patients completing seven sessions or more, questionnaire completion was poor. Posting questionnaires to patients and requesting they are completed and posted back is not a reliable method of collecting PRO data. Future studies of this intervention with longer follow-up periods will need to consider use of a more robust method to collect follow-up data, such as a secure electronic method (e.g. Microsoft forms).

These BR course and these data were collected during 2018–2019, before the COVID-19 pandemic. During and after the pandemic, it has been necessary to adapt BR so that it can be delivered remotely using an online video call

method such as Microsoft Teams. The feasibility and acceptability of delivering BR remotely will be evaluated in future research project.

Conclusion

BR significantly reduced the symptom burden, anxiety, and depression and increased the QoL of patients with fibromyalgia and CSS. These improvements were captured at 3 months post course indicating that the benefits of participating in BR persist after the course has finished.

Acknowledgements

We would like to thank Prof Michael Hyland for all his work in developing the Hyland model and advice on the preparation of this manuscript. JL is supported by the National Institute for Health Research (NIHR) Applied Research Collaboration (ARC) South West Peninsula. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR or the Department of Health and Social Care.

Author contributions

JWL, EH, CL, AD, KE and AFD researched literature and conceived the study. EH, AD and AFD we involved in protocol development, gaining ethical approval, patient recruitment and running the BR courses. JWL conducted the data analysis and wrote the first draft of the manuscript. All authors reviewed and edited the manuscript and approved the final version of the manuscript.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Guarantor

AFD: The guarantor is the person willing to take full responsibility for the article, including for the accuracy and appropriateness of the reference list. This will often be the most senior member of the research group and is commonly also the author for correspondence.

Trial registration

This randomized clinical trial was not registered because this was a clinical service evaluation.

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Supplemental material

Supplemental material for this article is available online.

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