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Developing a service user informed intervention to improve participation and ability to perform daily activities in primary Sjögren's syndrome: a mixed-methods study protocol

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ABSTRACT

Introduction: A significant proportion of patients with primary Sjögren's syndrome (PSS) is functionally impaired and experience difficulties participating in various aspects of everyday life. There is currently no evidence of efficacy for non-pharmacological interventions aimed specifically at supporting the patients with PSS to improve their participation and ability to perform daily activities. This paper describes a research protocol for a mixed-methods study to develop an intervention to improve these outcomes. The protocol follows the Medical Research Council framework for complex interventions.

Methods and analysis: We will use group concept mapping with the patients, adults who live with them and healthcare professionals to identify factors which prevent people with PSS from participating in daily life and performing daily activities. The factors will be prioritised by participants for importance and feasibility and will inform an intervention to be delivered within a National Health Service (NHS) setting. Evidence-based intervention techniques will be identified for the prioritised factors and combined into a deliverable intervention package. Key stakeholders will comment on the intervention content and mode of delivery through focus groups, and the data will be used to refine the intervention. The acceptability and feasibility of the refined intervention will be evaluated in a future study.

Ethics and dissemination: The study has been approved by an NHS Research Ethics Committee, REC Reference: 13/NI/0190. The findings of this study will be disseminated in peer-reviewed journals and through presentation at national and international conferences.

Trial registration number: UKCRN Study ID: 15939.

Strengths and limitations of this study

- This study will allow people with primary Sjögren's syndrome, their families and healthcare professionals to identify and prioritise key factors or barriers to participation and daily activities.
- This information will allow us to develop an evidence-based intervention to improve participation and the ability to perform daily activities for people with primary Sjögren's syndrome.
- This study will result in an intervention package and a feasibility study protocol in readiness for testing the intervention for acceptability and feasibility within a National Health Service setting.

BACKGROUND

Primary Sjögren's syndrome (PSS) is an autoimmune rheumatic condition which a recent meta-analysis has identified as having a prevalence rate of 74/100 000 inhabitants[1] using the American European Consensus Criteria (AECC).[2] In addition to the classic symptoms of oral and ocular dryness, arthralgia and myalgia[3] the patients with PSS experience significant fatigue,[4] sleep disturbance,[5] autonomic dysfunction[6] and a markedly reduced quality of life (QOL).[7] Furthermore, PSS is associated with significant direct and indirect healthcare costs equating to around £12–£15 000 per patient per year.[8 9]

We recently conducted a literature review on the impact of PSS on participation and the ability to perform daily activities and concluded that many patients with PSS are restricted in their ability to participate in various aspects of everyday life and are functionally limited, which is affecting their ability to carry out daily activities.[10] Consistently, using the Improved HAQ, a validated instrument for assessing ability to perform daily activities, we demonstrated that people with PSS have significant

impairment that is comparable with those with chronic fatigue syndrome.[11]

We also conducted a systematic review of non-pharmacological interventions for PSS which identified very few studies in the literature and concluded that there was no current evidence for the effectiveness of non-pharmacological interventions for PSS.[12] Current medical care for the patients with PSS has mainly focused on pharmacological interventions for their classic symptoms, which at best are only partially effective.[13] Currently there is no evidence of efficacy for non-pharmacological interventions aimed at improving participation and the ability to perform daily activities for the patients with PSS. This is in contrast to patients with other long-term conditions such as cancer, chronic fatigue syndrome and chronic pain who have access to psychosocial therapies which have been shown to improve symptoms and functional ability.[14–17] Therefore, there is a need to develop effective interventions to improve function and participation in everyday activities for people with PSS.

In order to develop an effective intervention that is deliverable in the UK's National Health Service (UK NHS), it is important to understand what the key issues are for service users, their families, healthcare professionals and those who commission and manage NHS services for the patients with PSS. This will maximise the likelihood that the intervention addresses important issues and is feasible and acceptable. The Medical Research Council (MRC) framework for complex interventions[18] advises an iterative stepped, structured and mixed-methods approach. At the early stages, when little is known regarding the determinants of illness and illness-associated impairments, a combination of qualitative techniques to explore patient experience and a review of the existing evidence is recommended. Such findings will then form the theoretical and empirical basis for developing an intervention specific for PSS. The key tasks are to identify intervention targets, the mechanisms whereby the proposed intervention will lead to change in participation and the ability to perform daily activities. Furthermore, recommendations will be made for specific measures to be used.

While the MRC guidance provides a useful framework for intervention development, it is less clear on the exact methods to achieve this. A variety of methods from economic modelling to computer simulation have been suggested, many of which may not be applicable to complex psychosocial interventions. In this regard, group concept mapping (GCM) methodology has been used to good effect for strategic complex planning in other diseases including dementia[19] in addition to intervention planning and treatment decisions in cancer.[20 21]

GCM, developed by Trochim,[22] is a mixed-methods participatory approach that uses a combination of group processes (brainstorming, sorting, rating and group interpretation) and a sequence of multivariate statistical analysis (multidimensional scaling and hierarchical cluster analysis) that result in visual representations of all stakeholders opinions in the form of concept maps. Priority values are added by participants to qualitative statements gathered during the brainstorming phase and these can be interpreted in pattern matches and value plots and used in planning or evaluation studies.[23] An advantage of GCM over some other methods is that it is an equitable process, giving an equal voice to all stakeholder groups and does not direct the participants to form a consensus. In the rheumatology field GCM has been used to design interventions to prevent work disability in patients with rheumatoid arthritis[24] and to understand their work requirements.[25]

In this study, we will identify priorities and intervention strategies that could improve participation and the ability to perform daily activities for people with PSS. The findings of the GCM exercise will be used to identify priority factors or targets, which are perceived to prevent the patients with PSS from participating in daily activities. Furthermore, the chosen factors will be measurable and tools selected to measure change for each. Existing evidence for each identified factor will be identified from the literature as the basis for the development of an intervention package aimed at improving daily function for people with PSS. The results of the concept maps and the planned development package will be discussed in focus groups of the patients with PSS, their family members or supporters and generic occupational therapists working in the UK NHS. The planned intervention will be further refined, drawing on feedback from those who will be at both the receiving and delivery ends of the proposed intervention.

At the conclusion of this study a detailed feasibility protocol will be drawn up following the proposed CONSORT reporting guidelines for feasibility and pilot studies[26] and ethical approvals will be sought. The intervention will subsequently be tested for feasibility on a small number of the patients with PSS in an NHS setting in a future study.

Although the project is primarily aimed to develop an intervention for use in UK NHS settings, its findings may be useful for other publicly funded health services or private healthcare providers.

AIMS AND OBJECTIVES

Aim

To build a model for intervention in PSS.

Objectives

1. To collect data from different stakeholder groups for priority intervention target areas in PSS.
2. To use existing clinical evidence to establish the optimum intervention and mechanism of effect of intervention for selected intervention targets.
3. To use existing clinical evidence to identify outcome measures best suited to capturing intervention effect.
4. To establish which of these priority areas could be realistically delivered within a UK NHS setting.
5. To design protocol for a future feasibility study of the refined intervention.

METHODS AND ANALYSIS

We will use GCM to explore the perspectives of three stakeholder groups on what will improve functional capacity of the patients with PSS. The stakeholder groups will be patients with PSS, adult household members (here onwards referred to as 'family') who live with someone who has PSS and healthcare professionals, managers and commissioners (here onwards referred to as 'providers') involved in the care delivery or service provision for patients with PSS.

Sample size

The recommended minimum number of participants for a GCM exercise is n=40.[27] We will seek to recruit approximately n=280 participants in order to enable subgroup analyses and to allow for modest attrition rates at each step of the GCM exercise (see figure 1). We aim to recruit 50 providers, 50 family members and 180 patients with PSS. This will allow us to detect a difference of the same order of magnitude as the background variability with 80% power. Subgroups within the patient group will allow us to compare opinions of the patients with PSS with varying levels of fatigue, QOL, perceived dryness, pain, cognitive symptoms and mood disturbances.

Recruitment

Patients who are participants in the UK Primary Sjögren's Syndrome Registry (UKPSSR)[28] and have consented to be approached about further studies will be invited to take part in this study via a postal invitation. Up to 12 patient identification sites across England will be used to identify potential participants. Family members will be invited to participate in an invitation addressed to an adult household member in the patient invitation pack. Providers will be invited to participate at professional meetings and through email invitation via email distribution lists.

Consent

The participants will be sent an invitation pack in the post. Within the pack will be a participant information sheet, a consent form and a reply form. They will be asked to reply to the invitation, indicating on the reply form whether they would like to participate. They will be provided with a telephone number and an email address to contact the research team if they have any questions. Once any queries have been satisfied they will be asked to sign, complete and return the consent and reply forms.

Data collection

Baseline demographics will be collected from all the participants (see figure 2). The PSS group will be asked to complete self-assessment on mood, QOL, function, fatigue, cognitive symptoms, dryness, discomfort and pain using validated instruments (see figure 3). This will allow us to perform subgroup analyses and compare whether opinions differ or not within the patient group. For example comparisons of the patients who are functionally impaired with those who are not; or those who are fatigued and those who are not. Family members will be asked to complete short validated questionnaires to

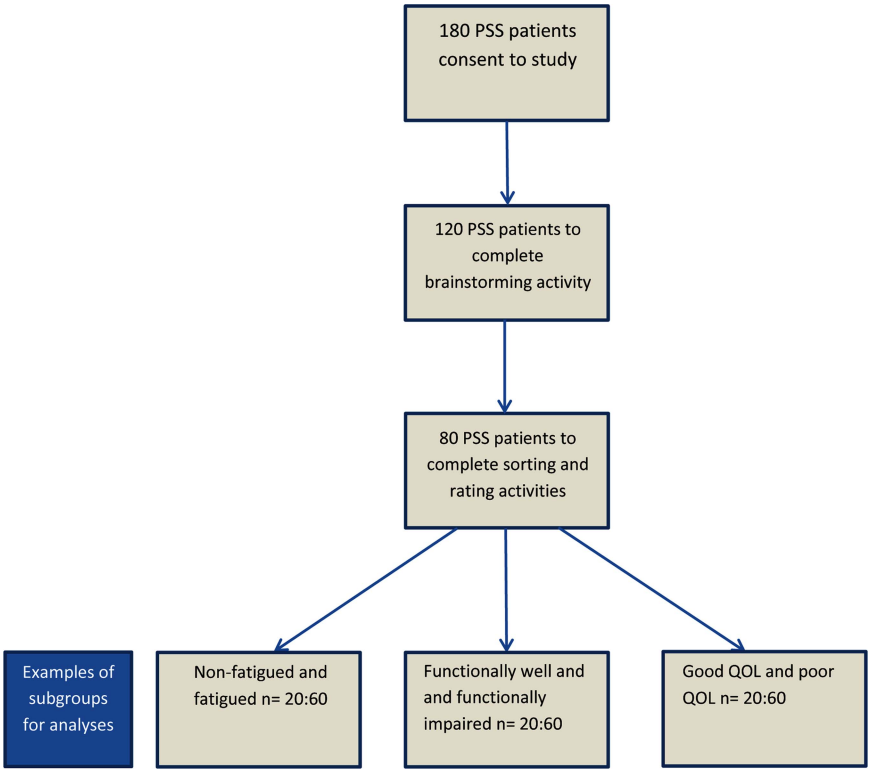

Figure 1 Flow chart of the participants to be recruited to the PSS participant group (PSS, primary Sjögren's syndrome; QOL, quality of life).

measure the impact of their spouse/relative's disease on them as well as their own QOL, as this may influence their opinion in the concept mapping exercise, regarding what they feel as priority areas for a healthcare intervention. Health care professionals (HCPs) will be asked to indicate which professional group they belong to. These data will allow us to perform additional subgroup analyses within the family and provider groups.

BUILDING A MODEL FOR INTERVENTION IN PSS

GCM phase

Identifying factors: idea generation/brainstorming

We will seek open contribution of ideas from each stakeholder group in response to a focus prompt. A focus prompt is an incomplete sentence designed to elicit ideas from the participants during the brainstorming phase of a GCM exercise. To generate a range of factors, the participants will be asked to complete the following focus prompt:

> People with primary Sjögren's syndrome would be able to do more of the things they want to do and the things they have to do if…

The participants will be asked to think and record as many responses as they can during this process.

Ideas analysis

All statements will be analysed and synchronised by a study advisory group consisting of representatives of all stakeholder groups and the research team. We will use a structured process recommended by Kane and Trochim[23] to remove duplicate statements and ensure

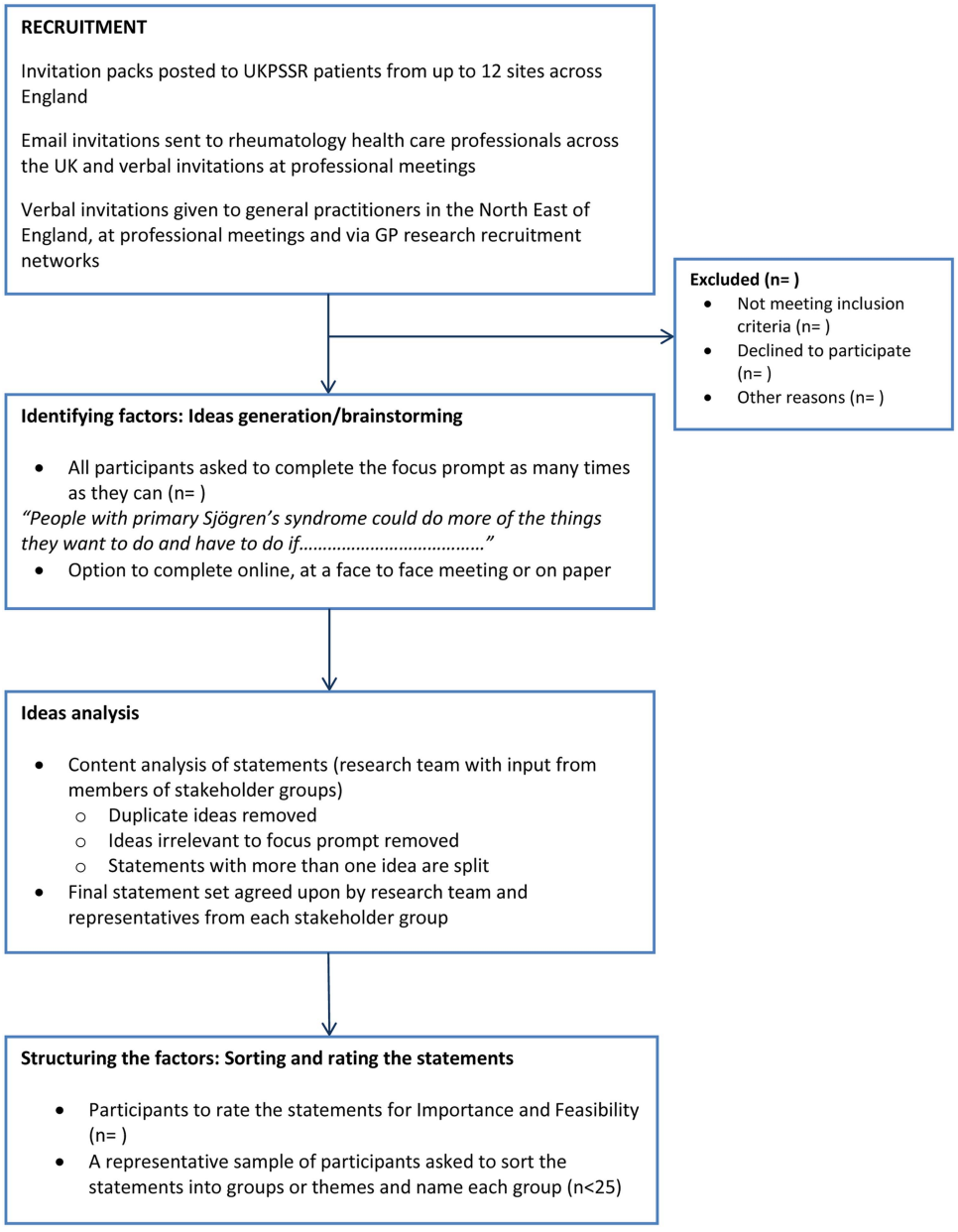

Figure 2 Flow chart of concept mapping study (GP, general practitioner; UKPSSR, UK Primary Sjögren's Syndrome Registry).

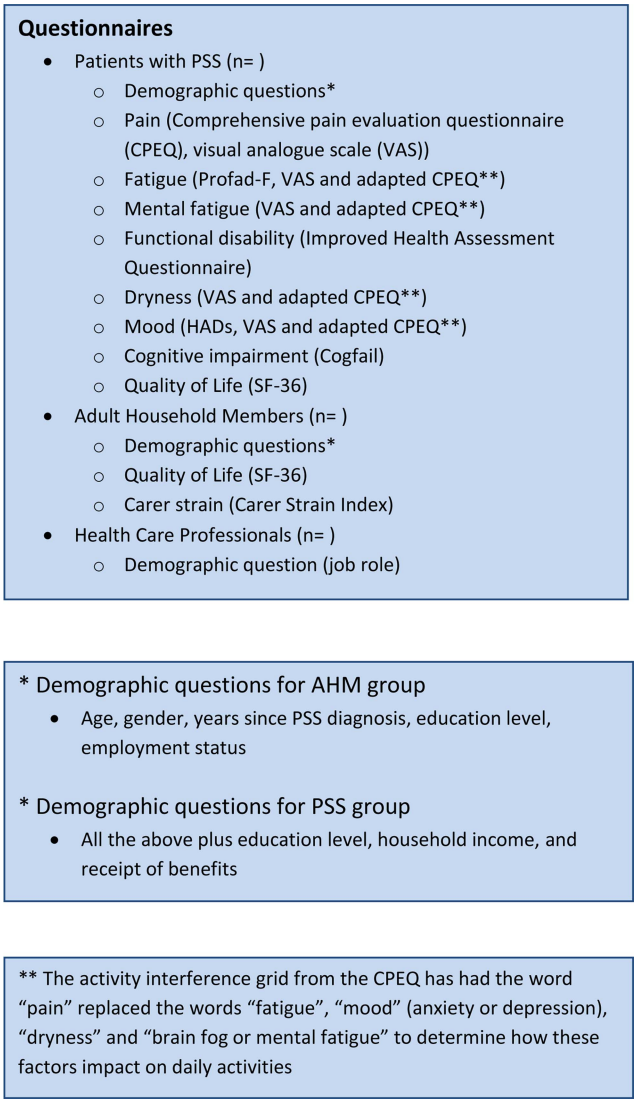

Figure 3 Measures to be used during the concept mapping phase (AHM, adult household member; HADS, Hospital Anxiety and Depression Scale; PSS, primary Sjögren's syndrome). **The activity interference grid from the CPEQ.

wording is clear. This will condense the statement set to one which is of manageable size (≤96 statements)[27] for the subsequent sorting and rating exercise but large enough to ensure saturation of the topic.[22 27]

Structuring the factors: sorting and rating the statements

The participants who previously took part in the brainstorming exercise will be invited to take part in the sorting and rating phase. Some participants may be recruited solely to the sorting and rating phase if saturation of the brainstorming topic is achieved prior to recruiting the planned number of participants, or if there is significant participant attrition between the two phases of the study. The participants will be asked to rate each statement in the distilled set on a five-point rating scale for importance and feasibility. When asked about feasibility they will be asked to consider how realistic it is to address the particular issue presented by the statement in an NHS setting. Next they will be asked to sort the statements into themes or groups of similar statements or ideas.

Procedures for the concept mapping phase

The patients with PSS and AHMs will be given the option of participating in the concept mapping exercises via one of the following means: (1) face-to-face focus groups of 5–10 people (if they live within 10 miles of the research centre), (2) a web-based interface or (3) paper-based questionnaire. HCPs will participate via the online web-based interface or on paper-based questionnaires. The participants completing the web-based concept mapping exercise will be given a unique username (not their name) and password.

In the group brainstorming session, a trained facilitator may use neutral prompts if ideas start to dry up. The participants also have the option of completing their responses on a piece of paper and handing it to a facilitator if they do not feel comfortable verbalising their responses. In the online format, the participants will be able to see the anonymous responses of the participants who have already completed the brainstorming process, which may help to prompt their own ideas. The participants choosing to complete the exercises on paper via the post will have a blank piece of paper to record their ideas on and will not have the benefit of seeing other peoples' responses.

When conducting the sorting and rating exercises, the participants may choose a different method to their chosen one for brainstorming if they wish. For example, they may complete the sorting and rating exercise online if they attended a face-to-face meeting for brainstorming. Furthermore, the participants may take part in sorting or rating even if they did not originally undertake the brainstorming exercise.

Concept mapping analysis and interpretation

The data generated from the above exercises will be analysed and represented in objective form as visual maps using the Concept Systems Global software package.

The maps will highlight the priority areas for each stakeholder group in improving function of the patients with PSS. Similarities and disparities between the stakeholder groups can be identified and if appropriate subgroup analyses will be carried out using the baseline demographic data collected at the start of the study and the self-assessment data. The "go zones" represent areas that are of most importance for more than one stakeholder group and so are of particular importance for planning interventions.

Intervention development phase

The key barriers or factors identified in the GCM exercises will be used as the basis for developing interventions and selecting appropriate measures which can be used later for a formal evaluation of effectiveness. We

will use an existing, reportable method described by Kolehmainen and Francis[29] to identify the techniques to be used in the intervention and will review existing literature to identify the measures. This involves three steps.

First, potential intervention techniques (active ingredients) will be identified from existing literature and relevant evidence-based theories. These will be identified in relation to the prioritised factors. Where the literature to indicate a technique for a factor is scarce, evidence-based theories from related fields, such as behaviour change will be used. Existing tools such as a matrix of behaviour change techniques[30] enable systematic identification of techniques from these fields. Second, the factor is specified as an observable and measurable 'construct' and a measure for identifying change in it will be selected. Third, each technique and factor pair is presented as a testable hypothesis in relation to the primary outcome.

An illustrative example of the application of this three-step method is shown below.

Illustrative example: potential factor—chronic fatigue

Evidence-based intervention techniques to target chronic fatigue include, grading physical activity/exercise[15] and goal setting, biofeedback and body changes.[30] Chronic fatigue is commonly measured as the self-reported mental and physical tiredness, by using tools such as the Chalder Fatigue Scale[31] or the Profile of Fatigue.[32] The testable hypothesis may be articulated as "grading, goal setting, biofeedback and body changes can be used to reduce chronic fatigue in order to increase participation in meaningful occupations."

The intervention will be reported using the Template for Intervention Description and Replication (TIDieR).[33]

Intervention refinement phase: focus groups

Once the potential intervention techniques have been identified from the literature, these will be presented to people with PSS, their families and occupational therapists in focus groups. The focus groups will provide opportunities to gather feedback regarding the specific intervention techniques, how the techniques will be best delivered and measures of effectiveness. The participants will be asked to generate ideas regarding how to deliver the techniques in an effective and acceptable way. Patient and carer participants will be identified from a regional specialist medical service for PSS and will be those who have previously indicated an interest at being involved with further research. Potential therapist participants will be recruited from local hospitals and community-based occupational therapists.

In addition the focus group participants will be invited to comment on the proposed outcome measures, including the choice of instrument(s) and the processes of administering and completing these instrument(s). The results will be used to refine the intervention package.

Designing a feasibility study

Once the intervention has been designed and the measurement tools identified, a detailed protocol following CONSORT guidance[26] will be drawn up and ethical approvals sought. This will form the next stage of the intervention development process.

ETHICS AND DISSEMINATION PLAN

This study has received approvals from the Office for Research Ethics Committees Northern Ireland (Ref: 13/NI/0190) and has been adopted onto the UK Clinical Research Network Study Portfolio (UKCRN Study ID: 15939). The results from both the concept mapping phase and an outline of the refined intervention will be disseminated nationally and internationally and submitted to scientific journals for publication.

SUMMARY

This study seeks to identify important factors from the patients with PSS, their families and providers regarding what could improve participation and the ability to perform daily activities for people with PSS. The identified factors will be targets for specific evidence-based intervention techniques and appropriate measures will be identified for each. This will result in an intervention package which will be refined in focus group discussions with key stakeholders. The next stage of the intervention development process will be a feasibility study of the refined intervention and this will be the subject of a future protocol.

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REFERENCES

1. Qin B, Wang J, Yang Z, *et al*. Epidemiology of primary Sjögren's syndrome: a systematic review and meta-analysis. *Ann Rheum Dis* 2014; pii: annrheumdis-2014-205375. Published online first. doi: 10.1136/annrheumdis-2014-205375
2. Vitali C, Bombardieri S, Jonsson R, *et al*. Classification criteria for Sjogren's syndrome: a revised version of the European criteria proposed by the American-European Consensus Group. *Ann Rheum Dis* 2002;61:554–8.
3. Fox RI. Sjogren's syndrome. *Lancet* 2005;366:321–31.
4. Norheim K, Le Hellard S, Nordmark G, *et al*. A possible genetic association with chronic fatigue in primary Sjögren's syndrome: a candidate gene study. *Rheumatol Int* 2014;34:191–7.
5. Theander L, Strombeck B, Mandl T, *et al*. Sleepiness or fatigue? Can we detect treatable causes of tiredness in primary Sjogren's syndrome? *Rheumatology (Oxford)* 2010;49:1177–83.
6. Newton JL, Frith J, Powell D, *et al*. Autonomic symptoms are common and are associated with overall symptom burden and disease activity in primary Sjogren's syndrome. *Ann Rheum Dis* 2012;71:1973–9.
7. Lendrem D, Mitchell S, McMeekin P, *et al*. Health-related utility values of patients with primary Sjogren's syndrome and its predictors. *Ann Rheum Dis* 2014;73:1362–8.
8. Callaghan R, Prabu A, Allan RB, *et al*. Direct healthcare costs and predictors of costs in patients with primary Sjogren's syndrome. *Rheumatology (Oxford)* 2007;46:105–11.
9. Bowman SJ, St Pierre Y, Sutcliffe N, *et al*. Estimating indirect costs in primary Sjogren's syndrome. *J Rheumatol* 2010;37:1010–15.
10. Hackett K, Newton J, Ng W-F. Occupational therapy: a potentially valuable intervention for people with primary Sjogren's syndrome. *Br J Occup Ther* 2012;75:247–9.
11. Hackett K, Newton J, Frith J, *et al*. Impaired functional status in primary Sjogren's syndrome. *Arthritis Care Res* 2012;64:1760–4.
12. Hackett KL, Deane K, Deary V, *et al*. Non-pharmacological interventions for primary Sjogren's syndrome: a systematic review of the evidence. *Ann Rheum Dis* 2014;73(Suppl 2):1211–12.
13. Ng WF, Bowman SJ. Biological therapies in primary Sjogren's syndrome. *Expert Opin Biol Ther* 2011;11:921–36.
14. Steultjens EM, Dekker J, Bouter LM, *et al*. Occupational therapy for rheumatoid arthritis. *Cochrane Database Syst Rev* 2004;(1):CD003114.
15. White PD, Goldsmith KA, Johnson AL, *et al*. Comparison of adaptive pacing therapy, cognitive behaviour therapy, graded exercise therapy, and specialist medical care for chronic fatigue syndrome (PACE): a randomised trial. *Lancet* 2011;377:823–36.
16. Ruehlman LS, Karoly P, Enders C. A randomized controlled evaluation of an online chronic pain self management program. *Pain* 2012;153:319–30.
17. McMillan EM, Newhouse IJ. Exercise is an effective treatment modality for reducing cancer-related fatigue and improving physical capacity in cancer patients and survivors: a meta-analysis. *Appl Physiol Nutr Metab* 2011;36:892–903.
18. Medical Research Council G. *Developing and evaluating complex interventions: new guidance*. London: MRC, 2008.
19. Anderson LA, Day KL, Vandenberg AE. Using a concept map as a tool for strategic planning: the Healthy Brain Initiative. *Prev Chronic Dis* 2011;8:A117.
20. Graham AL, Kerner JF, Quinlan KM, *et al*. Translating cancer control research into primary care practice: a conceptual framework. *Am J Lifestyle Med* 2008;2:241–9.
21. McFall SL, Mullen PD, Byrd TL, *et al*. Treatment decisions for localized prostate cancer: a concept mapping approach. *Health Expect* 2014. Published online first: 9 Feb 2014 doi:10.1111/hex.12175
22. Trochim W. An introduction to concept mapping for planning and evaluation. *Eval Program Plann* 1989;12:1–16.
23. Kane M, Trochim WMK. *Concept mapping for planning and evaluation*. London: Sage, 2007.
24. Varekamp I, Haafkens JA, Detaille SI, *et al*. Preventing work disability among employees with rheumatoid arthritis: what medical professionals can learn from the patients' perspective. *Arthritis Rheum* 2005;53:965–72.
25. Detaille SI, Haafkens JA, van Dijk FJ. What employees with rheumatoid arthritis, diabetes mellitus and hearing loss need to cope at work. *Scand J Work Environ Health* 2003;29:134–42.
26. Dolgin E. Publication checklist proposed to boost rigor of pilot trials. *Nat Med* 2013;19:795–6.
27. Rosas SR, Kane M. Quality and rigor of the concept mapping methodology: a pooled study analysis. *Eval Program Plann* 2012;35:236–45.
28. Ng WF, Bowman SJ, Griffiths B. United Kingdom Primary Sjogren's Syndrome Registry—a united effort to tackle an orphan rheumatic disease. *Rheumatology (Oxford)* 2011;50:32–9.
29. Kolehmainen N, Francis JJ. Specifying content and mechanisms of change in interventions to change professionals' practice: an illustration from the Good Goals study in occupational therapy. *Implement Sci* 2012;7:100.
30. Michie S, Richardson M, Johnston M, *et al*. The behavior change technique taxonomy (v1) of 93 hierarchically clustered techniques: building an international consensus for the reporting of behavior change interventions. *Ann Behav Med* 2013;46:81–95.
31. Chalder T, Berelowitz G, Pawlikowska T, *et al*. Development of a fatigue scale. *J Psychosom Res* 1993;37:147–53.
32. Bowman SJ, Booth DA, Platts RG. Measurement of fatigue and discomfort in primary Sjogren's syndrome using a new questionnaire tool. *Rheumatology (Oxford)* 2004;43:758–64.
33. Hoffmann TC, Glasziou PP, Boutron I, *et al*. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. *BMJ* 2014;348:g1687.

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Notes