Northumbria Research Link

Title: A mixed-methods study identifying key intervention targets to improve participation in daily living activities in primary Sjögren's syndrome patients

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ABSTRACT

Objective: Functional ability and participation in life situations are compromised in many primary Sjögren's syndrome (PSS) patients. This study aims to identify the key barriers and priorities to participation in daily living activities, in order to develop potential future interventions.

Methods: Group concept mapping (GCM), a semi-quantitative, mixed-methods, approach was used to identify and structure ideas from UK PSS patients, adults living with a PSS patient (AHMs) and health care professionals (HCPs). Brainstorming generated ideas, which were summarised into a final set of statements. Participants individually arranged these statements into themes and rated each statement for importance. Multidimensional scaling and hierarchical cluster analysis were applied to

sorted and rated data to produce visual representations of the ideas (concept maps), enabling identification of agreed priority areas for interventions.

Results: 121 patients, 43 AHMs and 67 HCPs took part. 463 ideas were distilled down to 94 statements. These statements were grouped into seven clusters; 'Patient empowerment', 'Symptoms', 'Wellbeing', 'Access and coordination of healthcare', 'Knowledge and support', 'Public awareness and support' and 'Family and friends'. Patient empowerment and Symptoms were rated as priority conceptual themes. Important statements within priority clusters indicate patients should be taken seriously and supported to self-manage symptoms of oral and ocular dryness, fatigue, pain and poor sleep.

Conclusion: Our data highlighted that in addition to managing PSS symptoms; interventions aiming to improve patient empowerment, general wellbeing, access to healthcare, patient education and social support are important to facilitate improved participation in daily living activities.

Significance and innovations

- Widespread stakeholder engagement with patients, family and health-care professionals has identified key priority themes including patient empowerment, symptoms, wellbeing and access to healthcare which can all be addressed to improve functional ability in primary Sjögren's syndrome patients.
- The greatest priority is to take primary Sjögren's syndrome patients seriously and to provide individualised support to self-manage the symptoms of dryness, fatigue, pain and poor sleep.

Primary Sjögren's syndrome (PSS) is a systemic autoimmune disease characterised by sicca symptoms(1). Additionally, extra-glandular symptoms are commonly experienced(2), including pain(3), sleep disturbances(4), fatigue(5), low mood and anxiety(6). These symptoms impact significantly on quality of life(7, 8) and many patients experience difficulty with participation (involvement in a life situation) and undertaking functional activities (9, 10). Examples include problems with hygiene, grip, reach, eating, transfers, mobility, eating, vocational activities and sexual activity(9-12). There are currently no effective disease-modifying treatments available and management strategies typically focus on symptom management. Pharmacological treatments mainly comprise of topical treatments for dryness as well as systemic treatments(13, 14). However, such treatments have limited effect on quality of life(15-17).

Previous studies have demonstrated an association between functional impairment, disease activity, pain and fatigue(9, 12). However, the key barriers to the performance of daily function and participation among PSS patients have not been systematically studied. In addition to gathering information from PSS patients, close family members can often provide useful insight into factors that interfere with the daily activities of the patients. Furthermore, in planning future interventions that are effective and feasible in order to improve daily function and participation of PSS patients(18, 19), it is important to understand the perspective of both potential users (PSS patients), their supporters (people they live with) and the healthcare providers who are likely to deliver the interventions in the future. To our knowledge, there have been no published studies investigating perspectives of other stakeholder groups such as family members and health care professionals who provide care to PSS patients.

The aim of the study was to identify key barriers and priorities to participation in daily living activities for PSS patients, as targets for future interventions.

PARTICIPANTS AND METHODS

We used Group concept mapping (GCM) methodology(19) to determine important key barriers to participation and daily function in primary Sjögren's syndrome (PSS) patients (see Figure 1). Our specific objectives were to:

i. Identify barriers/facilitators to participation and performance of daily activities

ii. Structure the generated ideas into clusters or themes through a sorting exercise

iii. Identify priority targets from the identified barriers/facilitators and themed clusters

iv. Compare similarities and differences in priorities between different stakeholder groups

GCM has been used in the rheumatic diseases to investigate treatment for hip and knee osteoarthritis(20); to design and develop online self-management interventions(21) and a programme to prevent work disability in rheumatoid arthritis (RA) patients(22). We have previously used this approach to evaluate and plan improvements to a fatigue service(23). The advantage of using this mixed methods approach over qualitative interviews or focus groups is that is that it mixes both qualitative and quantitative methods, large numbers of stakeholders can be consulted and it provides a consensus vision containing the prioritised ideas of all participants.

GCM is a semi-quantitative, mixed-methods participatory approach, which uses a combination of individual and group processes (brainstorming, sorting, rating and interpretation) and multivariate statistical analyses (multidimensional scaling and hierarchical cluster analysis). These result in visual depictions of all stakeholders' opinions in the form of concept maps. Participants add quantitative values to qualitative statements which are gathered during the brainstorming phase and these can

be interpreted in pattern matches and go-zones and used in planning or evaluation studies(24). We published the methods of this study a priori(25).

Participant groups

We recruited participants from three stakeholder groups. Firstly, PSS patients (aged 18 and over) who were members of the United Kingdom Sjögren's Syndrome Registry (UKPSSR)(26) and as such fulfilled the American European Consensus Group classification criteria(27). The second group were adults living with a PSS patient (Adult Household Member (AHM)). The final group were health care professionals (HCPs) working with PSS patients. We were granted approval to recruit UKPSSR participants from 12 different sites in England. We invited all UKPSSR participants at these sites to take part via a mailed written invitation. An enclosed invitation was included in the pack addressed to an AHM.

Data collection and analysis

We collected baseline age and gender demographics from PSS and AHM participants. PSS participants also indicated whether they received disability benefits, number of dependants living with them and their employment status. We asked HCPs to specify which professional group they belonged. This GCM study took place in five discrete phases and we gave participants the option to complete the activities either online or on paper. Participants could also choose to complete the brainstorming at a face-to-face meeting at Newcastle.

Stage 1. Ideas generation/brainstorming

We asked potential participants to respond to a 'focus prompt'; an incomplete sentence, which they could complete as many times as they wished. The research team designed the focus prompt and it went through several iterations. The precise wording of the focus prompt aimed to capture barriers and facilitators to participation in daily activities for people with PSS, using lay terms. The focus prompt was:

People with Sjögren's could do more of the things they want to do and have to do if……….

This process generated a list of statements/ideas from all participants taking part in this stage of the study. A participant completing this exercise online could see statements provided by other participants who had completed the brainstorming activity previously. We added statements received by participants in both the face-to-face meeting and from postal replies to the online interface. Therefore, participants taking part online could also see the statements provided through the alternative data collection methods. Brainstorming was continued until data saturation was achieved(19, 24). This was the point where no further unique ideas were being generated through the brainstormed responses(32).

Stage 2. Statement reduction

In this second stage, the full list of statements was reduced to a shorter list of unique ideas by several members of the research team (KH, VD, TR). Firstly, we split statements containing more than one idea into separate statements. Next, we applied a key word to each statement, formed groups of statements containing the same key word and considered them in turn. We removed duplicate statements and combined those which described the same or overlapping idea(24). Subsequently, the refined statement list was reviewed for syntax and readability by the research team, two patients with PSS and an AHM.

Stage 3. Sorting activity

During the statement reduction process, similar statements were considered together. Applying a random number (1-94) to each statement, in effect, shuffled the statement list prior to the sorting activity. The statements were numbered and randomised within the software used for this GCM project (CS Global MAX^{TM}). The numbered statements were printed onto individual cards and participants were asked to sort them by creating piles of similar meaning statements. They were

asked to name each pile and to record the name of each name and numbers of the statements

contained within each pile. Those opting to take part online could sort statements into virtual piles.

Stage 4. Rating activity

Participants were given a list of the numbered statements and asked to rate them for importance on

a 1-5 Likert scale, where 1=relatively unimportant and 5=extremely important.

Stage 5. Data analysis

Sorting and rating data were analysed in the CS Global MAX^{TM} software. Multidimensional scaling

was applied to the sort data, which was arranged into a similarity matrix to position each statement

relation to others as a point on an x-y axis. This results in a 2D representation of the statements and

each statement is represented by a numbered point on a map. Multidimensional scaling produces a

stress value. This indicates the goodness of fit of the map with the raw data and stability within the

overall map. A stress value below 0.36 is preferred in concept mapping studies(33). Statements

which were frequently sorted together end up being closely located to each other on the map, as

participants considered them to be similar conceptually during the sorting activity. Hierarchical

cluster analysis (Ward's method) was next applied to the data. This results in clusters of statements,

which were examined by the authors, who agreed an overall cluster solution through discussion. The

software suggests labels for clusters based on the names participants give to their piles during the

sorting exercise and appropriate cluster names were selected using these suggestions.

Importance ratings were considered at cluster level in a pattern match, which demonstrates

differences between the importance ratings attributed by each participant group to the clusters.

Importance ratings were also considered at statement level in go-zones. These are scatter plots

comparing importance ratings for each statement within a cluster for two groups. To make a visual

two group comparison, groups with both the lived experience of PSS (patients and AHMs) were

combined and compared with the HCP group. A statement falling within the top right quadrant, of

the go-zone (demarcated by the mean importance ratings for each group) indicates it is a priority for both lived experience and HCP groups. Go-zones were generated for each cluster.

Ethics approval

Ethics approval was granted by granted by the Office for Research Ethics Committees Northern Ireland (13/NI/0190, IRAS Ref: 125562) and the study registered on the National Institute of Health Research Comprehensive Clinical Research Network's portfolio of non-commercial studies (Study ID: 15939).

RESULTS

Participant characteristics

From the 371 patients invited to participate in the study 49% replied indicating they would like to take part and 33% of patients completed one or more stages of the GCM exercise. Flow diagrams of participants through the study can be seen in Supplementary Figure 1. 231 participants took part, including 121 PSS patients, 43 AHMs and 67 HCPs. The mean age of patient participants was 63 years (SD 10) and 64 (SD 9.5) for AHMs. Descriptive statistics demonstrating demographic data for both PSS and AHM participants and patient symptom scores can be seen in Table 1.

The HCP group included doctors (hospital doctors and general practitioners), therapists (physiotherapists, occupational therapists, psychologists, podiatrist), nurses and a service manager. A breakdown of professional groups within the HCP participants can be viewed in Supplementary Figure 2.

Statements – Stages 1 and 2

Brainstorming generated 463 statements, which were distilled to a final set of 94 unique statements.

Concept maps – generated from stages 3, 4 and 5

Multidimensional scaling resulted in a point map with a stress value of 0.18. A seven-cluster solution was agreed upon and contained the following named clusters: *Access and coordination of healthcare; Knowledge and support; Public awareness and support; Friends and family; Symptoms; Patient empowerment* and *Wellbeing*. The smallest cluster (Friends and family) contained 6 statements and the largest (Access and coordination of healthcare), 22 statements. The point cluster map can be seen in Figure 2.

Statements belonging to the Patient empowerment cluster received the highest priority ratings out of a possible 5, with a mean of 4.07 (SD 0.24), followed by the Symptoms cluster with a mean importance rating of 3.98 (SD 0.33) for each statement. The next most important clusters were Wellbeing (mean 3.91, SD 0.38); Access and coordination of healthcare (mean 3.89, SD 0.43); Knowledge and support (mean 3.74, SD 0.39) and Friends and family (mean 3.69, SD 0.30). The lowest rated cluster was Public awareness and support (mean 3.48, SD 0.36). Mean importance scores for each of the themed clusters, together with the mean rating scores for the individual statements within them can be viewed in Table 2.

Average importance rating scores for each cluster have been broken down by stakeholder group and can be seen in Figure 3. Importance is rated 1-5 with 5 being the maximum possible score.

Go-zones demonstrating the most important statements within the clusters as determined by all participants, as agreed by both health care staff and the combined PSS patient and household groups, can be seen in Figure 4. Priority statements are presented in the upper right quadrants of

each go-zone, which are demarked with mean importance scores for each cluster. The remaining go-zones can be viewed in Supplementary Figure 3.

Patient empowerment (8 statements): This cluster was rated the most important cluster by both the PSS and AHM group. Within this cluster, the statements #80 *'Felt they were being taken seriously'*, #25 *'Have support to manage their symptoms themselves'* and #29 *'There was a cure'* all fell within the top right priority area of the go-zone.

Symptoms (20 statements): Statements within this cluster all related to symptoms of PSS. Statements within the priority go-zone quadrant, all related to symptoms of fatigue, sleep disturbances, pain and oral or ocular dryness.

Wellbeing (16 statements): Statements falling within the priority quadrant include #21 *'Feel in control of their symptoms'*, #43 *'Have a positive attitude'*, #65 *'Learn to balance their activity and rest'*, #89 *'Develop good coping strategies'* and keeping both mind and body active (#90 and #91). There was some disagreement regarding the priorities of other statements within this cluster. The HCP group perceived #19 *'Could come to terms with their symptoms'*, #63 *'Could come to terms with their limitations'* and #52 *'Their mood was better'* as being priority statements, whereas those with the lived experience did not. Those in the lived experience group rated a good diet, better mobility and regular exercise as priorities, whereas the HCP group regarded these concepts as being less important.

Access and coordination of healthcare (22 statements): Statement #54 *'There is good communication between clinicians'* was rated as the most important within this cluster (See Table 2). Other priority statements indicate that 'one stop' clinics where a range of health care professionals can be seen in one visit (#7), professional support during flares of the condition (#14), clarity about who can be

contacted if symptoms do flare up (#1) and access to a consultant if required (#15) are all deemed as being important factors.

Knowledge and support (13 statements): The most important statements within this cluster were those relating to the need for more research to develop and test treatments and research to understand the causes of the disease. These were closely followed by statements relating to education on PSS for both healthcare professionals and patients.

Friends and family (6 statements): The most important statement within this cluster was #41 *'Have supportive family and friends'*. It was also deemed important that family could understand PSS symptoms (#83) and were able to explain to others what they could or could not do (#94).

DISCUSSION

This study has identified factors, which stakeholders have deemed to interfere with performance of the daily activities and participation in people with PSS. These factors have been grouped into conceptual cluster themes through the sorting process undertaken by stakeholders and subsequent multiple dimensional scaling and cluster analysis. The stress value of the point map generated during the multidimensional scaling process was low (0.18) demonstrating stability within the concept map (ideal stress values should fall below 0.39)(19). The factors were also prioritised at cluster level with individual priority factors identified within each cluster through ascertaining the mean rating scores of the individual statements within each of the clusters.

To our knowledge only one published study has explored patients' viewpoints on determinants they consider to interfere with their performance of daily activities and quality of life(34). In this qualitative focus group study, the authors found three broad domains containing 86 concepts. These domains were 'the physical dimension', 'psychological and emotional challenges' and 'social life and daily living'. The most commonly reported factors were from the physical domain, and specific factors within this domain included pain, dryness, shortness of breath and constipation(34).

However, only a small number of patients were involved (n=20), consequently, additional concepts may have been missed. Moreover, the authors did not use a structured approach to identify priority factors for intervention and did not seek the perspectives of close family members of the patients and healthcare providers. Our study has addressed these limitations. Indeed, our data have identified additional concepts not previously reported such as access to and coordination of healthcare.

The PSS participants who took part in this study had a mean age of 63 years, similar to the average age of UKPSSR participants (61 years)(7), which indicates that our sample was representative of this cohort. The average number of years since diagnosis in this study was 10. It is conceivable that 'newly' diagnosed PSS patients may have different needs and a separate study is required to address this.

Our data show that the most important themed cluster was Patient empowerment. Patient empowerment is a process where people improve their capacity to utilise their own resources to navigate their health care and live well with their chronic conditions(35). Others have described the potential for patient empowerment as occurring at three levels: At a patient level (e.g. patients' rights, responsibilities and opportunities) a healthcare provider level (e.g. through individual focused empowering intervention(s)) and at a healthcare system level (e.g. provision of group empowering intervention(s)(36). The Patient empowerment cluster was located centrally in the map, indicating a connection between this themed cluster and the surrounding clusters. Priority statements within the Patient empowerment cluster demonstrate that in order to empower patients, healthcare professionals need to take them seriously (#80) and support them to manage their symptoms themselves (#25). Multidisciplinary education has been shown to empower rheumatoid arthritis patients to manage their condition and reduce disease activity in the longer term(37). We hypothesise that addressing these factors, for example through individualised interventions supporting patients to manage their oral and ocular dryness symptoms and with non-

pharmacological interventions such as exercise(38, 39), pain management(40, 41) and cognitive behavioural therapy for sleep disturbances(4); patients may feel more patient empowered. Modes of delivering these interventions need to be considered and digital technologies, such as the use of mobile applications, can be utilised to empower patients to take charge of their own health(42) and used as an adjunct to face-to-face care.

The go-zone statements within the Wellbeing cluster include potential facilitators to self-managing symptoms such as fatigue and pain, including balancing activities and rest (#65) and developing good coping strategies (#89). These could be incorporated into a complex non-pharmacological behaviour change intervention package and ultimately empower patients to self-manage their symptoms(36). It is interesting that within the Wellbeing cluster, there was also some disagreement between the HCP and lived experience groups. HCPs considered mood as being an important factor, whereas the lived experience group did not prioritise this. Other studies have demonstrated a relationship between mood and quality of life(7), fatigue(43) and pain(44). However, these studies have not been able to determine whether mood is a consequence of these symptoms or a causal factor. Our study shows that although HCPs regard mood as a priority, patients prioritised other symptoms first. This might be due to patients viewing their low mood as being a consequence of these symptoms(34).

Our data also suggest that by addressing the structure of healthcare systems and how patients can access them could influence patients' ability to function better. For instance, PSS patients may see different specialists because of the diverse symptomatology of the disease. Allowing patients' access to several specialists within a single clinic would improve patient access and facilitate communication between clinicians.

Our data are presented as priorities within each go-zone and these may be helpful when designing services and interventions for PSS patients. However, it is important to stress that individual patients have different priorities and a personalised approach is essential. In order to provide a personalised approach, holistic and multidisciplinary care is required. Embedding access to multi-disciplinary

support within clinical services, (and addressing the priority statements within the Access & coordination of healthcare cluster) would facilitate individualised care.

This study is not without limitations. Only 33% of patient participants invited to take part in the study, went on to complete one or more stages of the GCM activities. We therefore cannot rule out possible selection bias. However, the mean age of participants in this study (63 years) is similar to the age of a recent study which included the majority of the UKPSSR cohort (mean of 61 years)(7). Secondly, 13% of the PSS patients who took part in this study were male, which is slightly greater than the proportion of males (9%) reported in a recent meta-analysis of PSS studies which included 7888 participants (45). We therefore compared differences in importance ratings for each cluster between males and female PSS participants (males, n=10, females, n=83) by generating a further pattern match. This revealed no differences in importance ratings between males and females (r=0.99). Therefore, despite a relatively greater proportion of male PSS patients taking part in this study, there was a very high level of agreement between males and females and the increased male representation did not influence the overall priority scores.

In conclusion, our study has identified several key areas, as targets for planning future interventions to support improvements in daily function and participation in PSS patients. Empowering patients by taking their health concerns seriously and supporting them to self-manage their condition is the greatest priority.

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Table 1: Demographic information and symptom scores for patients and demographic and Carer Strain Index scores for adult household members

PSS Patients	
Mean age in years (SD)	63.01 (9.96)
Mean number of years since diagnosis (SD)	10.15 (7.21)
Sex	87% Female
Live with another adult	73.50%
Dependents living at home	18% Yes
Employment status	5.7% Unemployed
	17% Employed P/T
	14.8% Employed F/T
	46% Housewife/husband
	57.9% Retired
Receiving disability benefits*	22% Yes
HADs Anxiety	7 (6)
HADs Depression	6 (5.7)
Pain VAS	37.3 (27.4)
Fatigue VAS	54.6 (29.2)
Mental Fatigue VAS	38.1 (28.7)
Dryness VAS	56.7 (30)
CFQ	43.2 (18)
Improved HAQ	17.2 (36.7)
Adult Household Members	
Sex	37.2% Female
Mean age in years (SD)	62.7 (11.4)
Mean number of years since diagnosis of household member (SD)	10.7 (7.9)
CSI	1 (3)

*Scores reported as medians (IQR) unless otherwise stated. U/E – Unemployed, P/T – Part-time, F/T – Full-time, HADs – Hospital Anxiety and Depression Scale (score range 0-21), VAS - visual analogue scale (score range 0-100), CFQ - Cognitive Failures Questionnaire (score range 0-100), Improved HAQ – Health Assessment Questionnaire (score range 0-100), *Disability Living Allowance, Attendance Allowance, Personal Independent Payments, Independent Living Fund, Employment and Support Allowance or Incapacity Benefit, CSI – Carer Strain Index (score range 0-13*

Table 2: Mean importance ratings for the clusters and each of the numbered statements within each cluster

Statements and clusters	Importance (1-5)
Cluster 1: Patient empowerment	**4.07**
29 There was a cure	4.45
80 Felt they were being taken seriously	4.34
25 Have support to manage their symptoms themselves	4.22
58 Take their medication as prescribed	4.07
24 Have confidence to seek advice when needed	4.02
22 Look after their physical, emotional and spiritual wellbeing	4.00
92 Were taught relaxation techniques	3.74
59 Have support with memory and concentration difficulties	3.73
Cluster 2: Symptoms	**3.98**
84 Their eyes were less dry	4.37
50 Have less pain	4.34
18 Were less fatigued	4.34
76 Fatigue was better managed/treated	4.34
32 Their vision was not impaired	4.32
61 Their eyes were more comfortable	4.31
71 Were able to sleep better	4.23
68 Have healthy teeth and/or comfortable dentures	4.17
69 Their throat was less dry	4.16
34 Swallowing was easier	4.10
82 Mouth and lips were less dry	4.07
72 Gastrointestinal (stomach and bowel) problems were managed	3.93
8 Were less prone to getting infections	3.89
31 Skin problems were treated	3.76
74 Were less breathless	3.64

77 Have more feeling in their mouth and lips	3.64
3 Did not have mouth sores or ulcers	3.61
36 Didn't have sexual problems	3.54
27 Their skin was less dry	3.46
26 Their vagina was less dry	3.40
Cluster 3: Wellbeing	**3.91**
91 Keep their mind active	4.41
43 Have a positive attitude	4.41
90 Keep their body active	4.36
21 Feel in control of their symptoms	4.24
89 Develop good coping strategies	4.17
47 Exercise regularly	4.01
65 Learn to balance their activity and rest	3.96
46 They have better mobility	3.90
19 Could come to terms with their symptoms	3.84
6 They have a good diet	3.79
63 Could come to terms with their limitations	3.78
49 Could improve their concentration	3.75
52 Their mood was better	3.73
10 Were less stressed or worried	3.69
30 Could continue to drive	3.68
5 Could go out in the sun	2.86
Cluster 4: Access & coordination of healthcare	**3.89**
54 There is good communication between clinicians	4.45
2 Have access to a range of good drug treatments	4.39
14 Have professional support during a flare up of symptoms	4.37
1 Know who to contact when their symptoms flare up	4.32
85 Associated conditions are diagnosed and treated	4.31
15 Can see a consultant when needed	4.25

93 Their healthcare is better coordinated	4.20
4 Knew the range of available treatment options	4.18
79 Diagnosis was quick	4.17
66 There was more funding for specialist rheumatology services	4.12
7 There were "one stop" Sjögren's clinics with all relevant health care professionals available	4.02
62 Have access to a specialist nurse	3.91
40 Have an individualised treatment plan	3.90
45 There was better management of the side effects of drugs	3.88
56 Health care professionals would raise sensitive topics (e.g. sex and vaginal dryness) during consultations	3.71
57 Professionals could direct them to support groups and charities	3.66
28 Have access to psychological support	3.49
70 Have access to occupational therapy	3.46
9 Have access to physiotherapy	3.39
88 Have access to complementary therapies or alternative remedies	3.23
44 There were diaries for recording symptoms and problems to bring to appointments with health care professionals	3.16
81 Have access to hydrotherapy	2.96
Cluster 5: Knowledge & support	**3.74**
20 There was more good research to test and develop treatments	4.45
13 There was more good research to understand the underlying causes	4.38
53 There was education on Sjogren's for healthcare professionals	4.28
37 There was education on Sjogren's for patients	3.97
75 There was information available on exercise and Sjogren's	3.67
60 Have access to support and advice from other people with Sjogren's	3.66
78 Have help with dental costs	3.65
67 Felt a family member or supporter would be welcome at their appointments	3.55
48 Have access to appropriate aids and adaptations in their homes	3.51
23 Felt a family member or supporter could be included in their care planning	3.48
11 Have Sjogren's advice leaflets	3.44
38 Could access support to help set personal goals	3.44

12 There were appropriate aids and adaptations in the community	3.17
Cluster 6: Friends & family cluster	**3.69**
41 Have supportive family and friends	4.12
83 Family could understand the symptoms	3.83
94 Can explain to others what they can and cannot do	3.78
55 Could easily describe Sjogren's to others	3.66
33 Friends and family include them in events	3.65
86 On a bad day people could tell by looking at them how they are feeling	3.10
Cluster 7: Public awareness and support	**3.48**
17 There was education about Sjogren's for people who fund services	4.01
87 Those unable to work and/or needed support to function, were eligible for benefits	3.92
64 Employers were aware of things they could do in the workplace that are helpful for people with Sjogren's	3.85
51 There was education on Sjogren's for family members	3.59
16 There was education about Sjogren's for the general public	3.38
35 Public spaces were more Sjogren's friendly e.g. heated/lit/airconditioned differently	3.28
39 Have a disabled parking badge	3.14
73 Public transport was accessible	3.09
42 Have assistance with shopping, cleaning etc.	3.04

Figure 1: The 5 Stages of Group Concept Mapping

Figure 2: Point Cluster Map Showing the 7 Themed Clusters

Figure 3: Pattern Match Depicting the Mean Importance Ratings by Participant Group

Figure 4: Go-zones Showing the Most Important Statements Within the Most Important Clusters

Supplementary Figure 1: Flow diagrams demonstrating recruitment of participants through the study

Supplementary Figure 2: Breakdown of professional groups within the health care professional participants

Supplementary Figure 3: Go-zones showing the most important statements within the lower priority clusters

Stage 1: Ideas generation/brainstorming

Participants complete an incomplete sentence (the focus prompt) as many times as they wish. Any many people take undertake this activity, a list of many statements is produced.

Stage 2: Statement reduction

Duplicate statements are removed and the remaining statements are corrected syntax, grammar and readability.

Stage 3: Sorting activity

Participants are asked to sort similar meaning statements into piles and to give each pile a name.

Stage 4: Rating activity

Participants are asked to rate each statement for importance on a 5 point scale.

Stage 5: Analysis

Multidimensional scaling and hierarchical cluster analysis are applied to the sorting data to produce concept maps with themed clusters. The rating data is applied to individual statements to create 'go-zones' and to the themed clusters to create 'pattern matches'.

Patient empowerment

r = 0.56

Health care staff (n=61)

4.48
4.11
2.74

Patients and household members (n=124)

2.82   4.05   4.6

25. Have support to manage their symptoms themselves
29. There was a cure
80. Felt they were being taken seriously

Symptoms

r = 0.85

Health care staff (n=61)

4.48
3.91
2.74

Patients and household members (n=124)

2.82   4.02   4.6

18. Were less fatigued
32. Their vision was not impaired
34. Swallowing was easier
50. Have less pain
61. Their eyes were more comfortable
68. Have healthy teeth and/or comfortable dentures
69. Their throat was less dry
71. Were able to sleep better
76. Fatigue was better managed/treated
84. Their eyes were less dry

Wellbeing

r = 0.71

Health care staff (n=61)

4.48
3.99
2.74

Patients and household members (n=124)

2.82   3.87   4.6

21. Feel in control of their symptoms
43. Have a positive attitude
65. Learn to balance their activity and rest
89. Develop good coping strategies
90. Keep their body active
91. Keep their mind active

Access & coordination of healthcare

r = 0.72

Health care staff (n=61)

4.48
3.89
2.74

Patients and household members (n=124)

2.82   3.88   4.6

1. Know who to contact when their symptoms flare up
2. Have access to a range of good drug treatments
4. Knew the range of available treatment options
7. There were "one stop" Sjogren's clinics with all relevant health care professionals available
14. Have professional support during a flare up of symptoms
15. Can see a consultant when needed
54. There is good communication between clinicians
79. Diagnosis was quick
85. Associated conditions are diagnosed and treated
93. Their healthcare is better coordinated