Six medico-psycho-social dimensions of a pedagogical model used to define clusters of patients with Sjögren's syndrome and intentionality to participate in a patient education programme

P. Antoine¹, S. Morell-Dubois¹, H. Maillard¹, G. Wojtasik¹, S. Sanges¹, D. Launay¹, V. Sobanski¹, A. Saraux², V. Devauchelle², É. Hachulla¹, M.-M. Farhat¹

¹University Lille, Inserm, CHU de Lille, Service de Médecine Interne et Immunologie Clinique, Centre de Rférence des Maladies Autoimmunes Systémiques Rares du Nord et Nord-Ouest de France (CeRAINO), U1286 - INFINITE - Institute for Translational Research in Inflammation, Lille; ²EA2216, ESPRI 29, IFR148, Département de Rhumatologie et de Pédiatrie, Hôpital de la Cavale-Blanche, Brest, and Université de Bretagne occidentale, Brest, France.

Abstract Objective

Sjögren's syndrome (SS) is an autoimmune disease with an impact on quality of life (QoL). The aim of patient education (PE) is to improve patients' QoL. The main objective of this study was to describe the medico-psycho-social characteristics defining the six spheres of an allosteric educational model in order to characterise clusters of patients with SS and intentionality for patients to participate in a programme of patient education.

Methods

A self-administered questionnaire was proposed to 408 patients with SS followed in the Department of Internal Medicine of the University Hospital of Lille, France with the aim of assessing the six spheres of the allosteric model: intentional, perceptual, affective, cognitive, infra-cognitive and meta-cognitive. Sub objectives were to determine factors that can influence intentionality to participate in a PE programme and to determine, using cluster analysis, similar characteristics of patients with SS.

Results

127 patients (31%) agreed to participate and were included in the study; 96% were women and the median age was 51 years (±14.5). They mostly reported dry syndrome and fatigue, had a good knowledge of SS, and presented anxiety symptoms. They mainly had problem-centred coping strategies, internal locus of control and low self-esteem. SS had an impact on their social interactions. Considering intentionality to participate in a PE programme, the patients were significantly younger, had a shorter duration of the disease, more frequently had disabled status, reported more fatigue, more self-reported symptoms and a poorer QoL. Two clusters of patients could be individualised, with one group including 75 (59%) patients presenting a higher global impact of the disease, including a more severe impairment for the scores of the perceptual, emotional and infra-cognitive spheres, worse physical QoL, and a higher intentionality to participate in a PE programme.

Conclusion

Our study described an SS population in terms of the different spheres of an allosteric model applicable to the practice of PE. A cluster of patients appeared to present more impact of the disease and more intentionality to participate in a programme of PE. There was no difference between the two groups in terms of the cognitive sphere (i.e. knowledge of the disease), thus indicating that motivation to participate in a PE programme is influenced by non-cognitive factors. Considering intentionality to participate in a PE programme, duration disease, age of the patient and QoL should be more considered to propose to patients to participate in a PE programme. Use of the allosteric model appears promising for future research in PE.

Key words Sjögren's syndrome, patient education, allosteric model, quality of life.

Pascale Antoine, MD Sandrine Morell-Dubois, MD Hélène Maillard, MD Géraldine Wojtasik, Ms Sébastien Sanges, MD David Launay, MD, PhD Vincent Sobanski, MD, PhD Alain Saraux, MD, PhD Valerie Devauchelle, MD Éric Hachulla, MD, PhD Méryem-Maud Farhat, MD

Please address correspondence to: Méryem-Maud Farhat CHU Lille, Hôpital Claude Huriez, Rue Michel Polonowski, 59000 Lille, France. E-mail: meryem.farhat@chu-lille.fr

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Introduction

Sjögren's syndrome (SS) predominantly affects women, with a sex ratio of 9 women to 1 man, and has a peak incidence at 50 years of age. It is a chronic autoimmune disease that can be primary or secondary (i.e. associated with another systemic autoimmune disease). SS develops and progresses very slowly, so that diagnosis is often delayed. It is only very rarely life-threatening and the average survival of affected patients does not differ significantly from that of a general population. SS is a disease with a broad spectrum of clinical manifestations. SS patients in 80% of cases suffer from dry syndrome. Xerophthalmia is one of the most troublesome symptoms and can severely impair quality of life (OoL) (1). Glandular involvement is not limited to the lacrimal and salivary glands but affects all exocrine glands (2). Extra-glandular involvement is present in about 25% of patients, with preferential involvement of the musculoskeletal, digestive, pulmonary and/or haematological systems. Asthenia is a very common symptom in patients with SS, occurring more frequently than in the general population (3, 4). Indeed, in the study by Urbanski et al. assessing fatigue in patients with SS, the mean Fatigue Severity Scale (FSS) score was 4.82±0.24 (indicating severe fatigue) and 75% of patients suffered from fatigue (5), a higher proportion than that found in other autoimmune diseases (6). The study by Brito-Zeron et al. demonstrated the association between fatigue and an increased incidence of anxiety and sleep disorders, depression, and non-specific symptoms such as myalgia, arthralgia and malaise (7). Several studies have shown that patients with SS have greater impairment of their QoL, as assessed by the 36item Short Form Health Survey (SF-36), compared to the general population (8-10). Liu et al. demonstrated a negative correlation between QoL in patients with SS and anxiety and depressive symptoms as assessed by the Hospital Anxiety and Depression Scale (HADS) questionnaire (10). In the study by McCoy et al. (11), the top three symptoms or signs that patients with SS hoped new treatments would address were dryness, fatigue and a reduction in lymphoma or blood cancer risk and the top aspects of QoL that patients reported were impaired by SS were sex life, hobbies, social activities and extracurricular activities, job/career or ability to work, and finding the correct word during conversations.

Patients with SS suffer mainly from moderate to severe depression and/or anxiety (8). A meta- analysis showed a higher prevalence of depression in patients with SS compared to the general population (12). Similarly, the study by Cui *et al.* found that mean HADS-A (HADS-Anxiety) and HADS-D (HADS-Depression) scores were higher in patients with SS than in a control group (13).

The chronicity and impact of SS make the role of patient education (PE) essential. In 1998, the World Health Organization (WHO) encouraged the introduction of PE to "help patients acquire or maintain the competencies they need to manage as well as possible their lives with a chronic disease (...) It comprises organized activities, including psychosocial support, designed to make patients aware of and informed about their disease and about health care, hospital organisation and procedures, and behaviour related to health and disease, so that they (and their families) understand their disease and their treatment, collaborate with each other and take responsibility for their own care as a means of maintaining or improving their quality of life".

Various educational models to practice PE can be used to propose the better environment to let patients acquiring new skills. Giordan *et al.* described a so-called "allosteric" model based on the exploration of six dimensions of the person (14, 15):

- Intentional: the desire to learn.
- Perceptual: the body's experience.
- Emotional: the emotional experience.
- Cognitive: knowledge related to the disease.
- Infra-cognitive: thought reflexes, reasoning, coping mechanisms.
- Meta-cognitive: opinion, especially about the social environment and the health care system.

This exploration should lead to the establishment of a motivational environment that facilitates the implementation of tools that are favourable to behaviour change (14-16). Exploring the six dimensions of the person in patients with SS appears as a mandatory prerequisite for creating a suitably adapted PE programme. To our knowledge, this model has yet not been used in PE applicative research.

The main objective of this study was to explore the six medico-psycho-social characteristics, as defined in the aforementioned allosteric educational model of patients with SS. Sub objectives were to determine factors that can influence intentionality to participate in a PE programme and to determine, using cluster analysis, similar characteristics of patients with SS.

Patients and methods

This was a single-centre, cross-sectional, observational study. Patients aged between 18 and 65 years and diagnosed with SS according to the ACR-EULAR 2016 criteria, followed in the internal medicine department of Lille University Hospital, France, were included, subject to their having given informed consent to participate in the study. The study complied with the ethical code of the Declaration of Helsinki. An observational, questionnaire-based study, with self-questionnaires all validated in French which took approximately 50 minutes to complete, assessed demographic and medico-psycho-social data relating to the six dimensions of the allosteric model. Patients were included in 2020 following their consultation with their health care provider as part of their routine follow-up.

Demographic data was self-collected including gender, age group, marital status, education level, professional status, occupation, worker status, year of diagnosis, complications of the disease and treatments.

The intentional sphere was assessed by a closed question on the patients' interest in participating in a PE programme. In the case of a positive answer, a list of workshops was proposed. In addition, two visual analogue scales (VAS) were used to quantify patients' willingness



Fig. 1. The six dimensions of the allosteric model and evaluation criteria. PE: patient education.

to learn and to participate in a workshop. The perceptual sphere was explored by two specific validated self-questionnaires: PROFAD-SSI (17, 18), assessing respectively somatic and mental fatigue, arthralgia, vascular anomalies, xerostomia, xerophthalmia and vaginal dryness, with significant impairment with a score higher than 5 and the EULAR Sjögren's Syndrome Patient Reported Index (ESSPRI) (18, 19) assessing fatigue, dryness and pain with significant impairment with a score higher than 5. The Validated Patient Global Assessment of disease activity (PtGA) self-questionnaire assessed the patient's disease activity (1 VAS scored out of 10) (20). Pain intensity and impact was assessed by the validated BPI self-questionnaire (15 VAS rated out of 10 and 2 open items) (18, 21). Fatigue was explored via the validated Multidimensional Assessment of Fatigue (MAF) self-questionnaire (14 VAS, 2 closed items at 4 degrees) scored out of 50 (22), and via the validated FSS self-questionnaire specific to rheumatism (9 closed items at 7 degrees ranging from strongly disagree to strongly agree, scored out of 7) with significant impairment indicated by a score higher than 4 (23).

The emotional sphere was assessed by the validated HADS self-questionnaire (14 closed items with 4 levels distributed in two scores, HADS-A for anxiety and HADS-D for depression, with a definite symptomatology indicated by a score higher than 10) (24), and by evaluation of the impact of the diagnosis announcement and potential complications on the patient's morale and anxiety usin g 4 closed items with 4 degrees.

The cognitive sphere was explored by 9 "true/false" items giving a knowledge score out of 9, focusing on patients' conception of their disease. Treatment compliance was self-assessed by a closed item with 4 degrees of compliance (compliance estimated as poor, mediocre, good, and very good).

The exploration of the infra-cognitive sphere included the evaluation of patients' coping strategies by the validat-

ed Cousson Coping Self Questionnaire (27 closed items with 4 degrees) giving 3 types of coping strategies (25); perceived control of the disease by the Wallston scale (validated self-questionnaire of 18 closed items with 6 degrees) giving 3 types of locus (internal, external and due to chance) (26) and self-esteem by the validated Rosenberg self-questionnaire (10 closed items with 4 degrees, rated out of 40, with very low self-esteem for a score below 25, low between 25 and 30, average between 31 and 33, high between 34 and 39 and very high if above 39) (27).

The meta-cognitive sphere was evaluated by measuring patients' satisfaction with the information received at the time of diagnosis and during follow-up on: diagnosis, disease course, complications of the disease, treatments with their indications and side-effects (8 closed items with 4 degrees: very insufficient, insufficient, sufficient, very sufficient) and the repercussions of SS on daily life (social, professional, familial, sentimental and sexual) (13 closed items), from which a social interaction score was established, rated out of 8. Some questionnaires explored the different spheres simultaneously: the brief illness perception questionnaire (IPQ-BREF), a validated self-questionnaire, assessed whether the illness was perceived by patients as threatening (9 VAS scored out of 80) (28); the validated SF-36 self-questionnaire which explored QoL (36 closed items, divided into two scores, the physical score and the mental score, each scored out of 100: the higher the score, the better the QoL) (29-31) and the validated self-assessment questionnaire assessing health-related QoL in primary SS (QoL-pSS), which specifically assessed QoL in SS (25 closed items with 4 degrees, scored out of 96 for women and 92 for men: the higher the score, the worse the QoL) (32).

All assessment parameters and selected questionnaires are summarised in Figure 1.

Statistical analysis

Statistical analyses were performed using R software, v. 3.6.2. All statistical tests were two-sided with a p-value of 5%.

Table I. Description of demographic and medico-psycho-social self-assessed variables and distribution of the population according to patients' interest in participating in PE.

Variables	Population	Interest in participating in PE		p
	n=127	Yes n=87	No n=40	Г
Damagagahu				
Age (Years)	51 + 14.5	48 + 12	54.5 + 15	0.05*
Duration of illness (Years)	7 ± 10.5	6 ± 9	9.5 ± 12.3	0.01*
Gender (Female)	122 (96.1)	84 (96.6)	38 (95)	0.65
Non-smoker	88 (69.3)	62 (71.3)	26 (65)	0.65
Civil status				
Married - cohabitation	88 (69.3)	57 (65.5)	31 (77.5)	
Single	18 (14.2)	14 (16.1)	4 (10)	0.43
Divorced	13 (10.2)	11 (12.6)	2 (5)	
Widowed	8 (6.3)	5 (5.8)	3 (7.5)	
Level of education				
Higher education	65 (51.2)	45 (51.7)	20 (50)	0.09
High school	44 (34.7)	31 (35.6)	13 (32.5)	
College	9 (7.1)	8 (9.2)	1 (2.5)	
Technical education	9 (7.1)	3 (3.5)	6 (15)	
Professional status				
Full-time working	42 (33.1)	30 (34.5)	12 (30)	
Working part-time	22 (17.3)	13 (14.9)	9 (22.5)	
Retired	16 (12.6)	9 (10.3)	7 (17.5)	
Inactive On disability pansion	10(12.0) 15(11.8)	13(14.9) 12(14.0)	3(7.5)	
On sick leave	13(11.0) 11(8.7)	13(14.9) 5(58)	$\frac{2}{6}(15)$	
Working part-time on a therapeutic h	11(0.7)	3(3.5)	0(13)	
Never worked	2(1.6)	1(1.2)	1(2.5)	0.17
Disabled worker status	38 (29.9)	31 (35.6)	7 (17.5)	0.04*
Symptomatic	119 (93.7)	83 (95.4)	36 (90)	0.26
Currently taking treatment	65 (51.2)	44 (50.6)	21 (52.5)	0.85
Cognitive sphere				
Knowledge score	8 + 1	8 + 1	8 + 1	0.35
Treatment compliance	52 (80)	36 (81.8)	16 (76.2)	0.74
Quantification forgetting treatment /v	veek			
none	53 (81.5)	36 (81.8)	17 (81)	
1 time	4 (6.2)	3 (6.8)	1 (4.8)	
2 times	6 (9.2)	4 (9.1)	2 (9.5)	0.76
3 times	1 (1.5)	0 (0)	1 (4.8)	
\geq 4 times	1 (1.5)	1 (2.3)	0 (0)	
Perceptual sphere				
Profad-SSI score	3.8 ± 2.8	4.1 ± 2.1	2.7 ± 2.7	< 0.001*
ESSPRI score	6.3 ± 3	6.7 ± 2.7	4.7 ± 4.8	0.001*
BPI score pain severity	4.3 ± 6.5	4.5 ± 6.5	3.3 ± 5.5	0.07
BPI score pain-related discomfort	3.9 ± 6.4	4.1 ± 6.9	3 ± 5.1	0.1
MAF score	34.7 ± 14.4	36.6 ± 12.5	28.5 ± 17.8	0.001*
FSS score	5.3 ± 2.8	5.4 ± 2.1	4.4 ± 3.5	0.02*
PIGA score	0 ± 3.3	0 ± 3	5.5 ± 4	0.06
Emotional sphere				
HADS-A score	10 ± 7	11 ± 7.5	10 ± 5.3	0.06
HADS-D score	7 ± 6	8 ± 6	6 ± 7	0.06
Diagnosis' impact on anxiety				
None	23 (18.1)	17 (19.5)	6 (15)	0.64
Moderate	59 (46.5)	37 (42.5)	22 (55)	
High	35 (27.6)	25 (28.7)	10 (25)	
very high	10 (7.9)	8 (9.2)	2 (5)	
Diagnosis' impact on morale	10 (14.0)	12 (14.0)	5 (10.5)	
None	18 (14.2)	13 (14.9)	5 (12.5)	0.00
Moderate	08(33.3)	43 (49.4)	25 (62.5)	0.62
Important Very important	29 (22.8) 12 (0.5)	22 (23.3) = 0 (10.3)	(1/.3)	
Very important	12 (9.3)	9 (10.3)	5 (1.5)	
Impact on anxiety of risk of complica	ations	21 (24.1)	8 (20)	
Moderate	29 (22.8) 55 (12.2)	21 (24.1) 36 (41.4)	o (20) 10 (47 5)	0.47
Important	26 (20.5)	16 (18.4)	10(47.3)	0.47
Very important	17 (13.4)	14(16.1)	3(7.5)	

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Variables	Population n=127	Interest in participating in PE		
		Yes n=87	No n=40	
Impact on morale of risk of complication	s			
None	31 (24.4)	22 (25.3)	9 (22.5)	
Moderate	50 (39.4)	34 (39.1)	16 (40)	0.98
Important	32 (25.2)	21 (24.1)	11 (27.5)	
Very important	14 (11)	10 (11.5)	4 (10)	
Infra-cognitive sphere				
Problem-centred coping score	25 ± 9.5	25 ± 9	25 ± 9	0.23
Emotion-centred coping score	19 ± 9.5	20 ± 11	18.5 ± 8	0.26
Search for social support	18 ± 8	20 ± 8	17 ± 8.3	0.16
Internal locus score	2.5 ± 0.5	2.5 ± 0.5	2.5 ± 0.3	0.20
Random locus score	2.3 ± 0.5	2.3 ± 0.5	2.3 ± 0.4	0.2
External locus score	2.3 ± 0.5	2.3 ± 0.5	2.3 ± 0.7	0.68
Rosenberg score	30 ± 8.5	29 ± 8.5	31 ± 9	0.81
Meta-cognitive sphere				
Social interaction score	5 ± 2	5 ± 2	6 ± 2	0.08
At diagnosis, sufficient information recei	ved on:			
Diagnosis	70 (55.1)	44 (50.6)	26 (65)	0.18
Course of the disease	45 (35.4)	27 (31)	18 (45)	0.16
Complications	40 (31.5)	24 (27.6)	16 (40)	0.22
Treatment	58 (45.7)	36 (41.4)	22 (55)	0.18
Adverse effects	47 (37)	32 (36.8)	15 (37.5)	1
During follow-up, estimated sufficient int	formation on:			
Diagnosis	70 (55.1)	46 (52.9)	24 (60)	0.56
Evolution	44 (34.7)	29 (33.3)	15 (37.5)	0.69
Complications	42 (33.1)	26 (29.9)	16 (40)	0.31
Treatment	57 (44.9)	38 (43.7)	19 (47.5)	0.70
Adverse effects	53 (41.7)	36 (41.4)	17 (42.5)	1
Cross-sectional items				
SF-36 physical score 4	8.9 ± 30.9	47.2 ± 30.8	56 ± 38.3	0.10
Mental score SF-36 4	7.1 ± 25.3	46 ± 24.4	56.3 ± 25.8	0.02*
Qol-PSS score	48 ± 16.5	51 ± 12.5	45 ± 21.5	0.006*
IPQ-BREF score	50 ± 14.5	49 ± 15	50 ± 14	0.54
Intentional sphere				
VAS score for willingness to participate in PE	5 ± 7.5	8 ± 5	0 ± 1.3	<0.001*
VAS score for desire to learn skills	7 ± 5	8 ± 3	1.5 ± 4.3	<0.001*

*Statistically significant.

Quantitative variables are expressed as median \pm interquartile range; qualitative variables are expressed as number of patients (percentage).

All data were described in the study population: mean (standard deviation) or median [interquartile range (IQR)] if not normally distributed for quantitative variables; numbers (percentages) for qualitative variables. The characteristics of intentionality to join a PE programme were compared using Kruskal-Wallis tests for quantitative variables and Fisher exact tests for qualitative variables. Principal component analysis (PCA) was performed to represent graphically the correlations between the different scores collected, with variables represented by points within a circle. Multiple univariate tests can be responsible for an inflation of the alpha risk. However, multiple test adjustments in such exploratory study, in addition for a rare disease, are not strictly required. Considering that the different spheres are interconnected, we did not perform a multivariate analysis regarding interest to participate in a PE programme.

Cluster analysis was carried out by bottom-up hierarchical clustering using Ward's minimum variance from the selected medical-psycho-social variables, which were centre-reduced. The medical-psycho-social variables collected to characterise the dimensions of the allosteric model were selected, excluding the intentional sphere. For the main analysis, we assessed the number of clusters using the visual distance criterion at the horizontal intersection at the highest level of dissimilarity on the dendrogram (*i.e.* where the sum of the lengths of the vertical branches was the largest) (33). In an exploratory approach, we used another criterion (Dindex) to estimate the number of clusters. Stability was assessed using the Jaccard index [a Jaccard similarity index <0.5 indicates a weakly stable cluster, while a coefficient >0.75 is in favour of a good stability of the cluster (34)] in a resampling process (by bootstrapping) (35).

Results

The study was proposed to 408 patients, and 127 (31%) participated. The patients' demographic and medicopsycho-social characteristics used in assessing the six spheres are presented in Table I.

Of the 127 patients, 122 (96.06%) were women and the median age was 51 years (± 14.5); the median duration of the disease was 7 years (± 10.5) ; 119 patients (94%) self-reported being symptomatic: 80 patients (63%) described myalgias, 62 (49%) muscle weakness, 56 (44%) inflammatory arthralgia, 45 (35%) Raynaud's phenomenon, 38 (30%) dyspnoea, and 33 (26%) a previous or current episode of parotitis. Complications described by the patients were as follows: ophthalmological for 58 patients (46%), stomatological for 47 (37%), pulmonary for 24 (19%), neurological for 22 (17%), haematological for 13 (10%) and renal for 5 (4%).

Regarding intentionality, 87 patients (68%) were interested in participating in a PE programme.

Regarding the perceptual sphere, the median Profad-SSI was 3.76 (IQR 2.76), with no significant discomfort due to the specific symptomatology of SS, while the median ESSPRI was 6.33 (IQR 3), indicating significant discomfort.

Regarding the emotional sphere, 63 patients (50%) had anxiety symptoms and 31 (24%) had depressive symptoms.

Regarding the cognitive sphere, among the 65 patients currently taking treatment, compliance was judged as being "good" by 52 patients (80%).

Regarding the infra-cognitive sphere, the highest median score on the Cous-



son Coping Scale was problem-centred coping at 25 (IQR 9.5), followed by emotion-centred coping [median score of 19 (IQR 9.5)]. On the Rosenberg self-esteem questionnaire, 24 patients (19%) had very low self- esteem, 19 had average self-esteem (15%) and 3 (2%) had very high self-esteem.

Regarding the meta-cognitive sphere, the median social interaction score was 5 (IQR 2), reflecting a significant impact on the social environment.

The median SF-36 physical score was 48.85 (IQR 30.89) and the median mental score was 47.08 (IQR 25.31). The median IPQ-BREF score assessing the patient's global view of their disease was 50 (IQR 14.5).

The results of the PCA are presented in Figure 2. There was a correlation between the results of the self-questionnaires assessing the perceptual, emotional, and QoL spheres (with an inverse correlation for the SF-36).

Compared to the group of patients with no interest in PE, interested patients were significantly younger, with a shorter disease duration, were more frequently recognised as disabled workers, were more symptomatic (Profad-SSI score, ESSPRI score, MAF score, FSS score), with a poorer QoL (SF-36 mental score and Qol-PSS score), and greater intentionality in participating in PE and in learning new skills.

Cluster analyses

An optimal number of two clusters resulted from the analysis of the 21 selected variables: Cluster 1 (C(1)) and Cluster 2 (C(2)) (Fig. 3). For C(1), the Jaccard index was 0.84, and for C(2), it was 0.67. The characteristics of these two clusters are summarised in Table II. C(1) was composed of 52 patients (40.94%) and C(2) of 75 patients (59.06%). More patients had self-reported complications of the disease in C(2) than in C(1) (58 patients (77.33%) vs. 28 patients (53.85%); p<0.01) and more patients had disabled worker status (31 patients (41.33%) vs. 7 patients (13.46%); p<0.01).

There was a significant difference between the two clusters concerning the perceptual, emotional, infra-cognitive, and intentional spheres and impairment of quality of life.

- Regarding the perceptual sphere, C(2) patients had greater discomfort due to SS symptomatology than C(1) patients on all perceptual questionnaires.
- Regarding the emotional sphere, C(2) patients had anxiety symptomatology (median HADS-A score 12 (IQR 5)) and depressive symptomatology (median HADS-D score 10 (IQR 5)), whereas C(1) patients had neither anxiety nor depressive symptomatology.
- Regarding the infra-cognitive sphere, patients in both clusters had a predominantly problem-centred coping strategy (medians respectively of 25 (IQR 9.5) and 25 (IQR 8.25)). The median of the internal locus score was the highest of all medians in C(1) (2.67 (IQR 0.33)) while they were superimposable for the three types of locus in C(2) (2.33 (IQR 0.5 to 0.67)). Self-esteem was lower in C(2) patients, with a significantly lower median Rosenberg questionnaire score (28 (IQR 8) *vs*. 34.5 (IQR 6.25); *p*<0.01).
- QoL was more impaired in C(2) patients than in C(1) patients. The median of the physical and mental sub-dimensions of the SF-36 was lower (36.88 (IQR 18.96) vs. 69.32 (IQR 22.58); p<0.01 and 40.42 (IQR 16.3) vs. 66.2 (IQR 19.53); p<0.01) as was the median of the Qol-PSS score (54 (IQR 12) for C(2) and 36.5 (IQR 18.25) for C(1); p<0.01).
- Regarding the intentional sphere, more patients were interested in participating in a PE programme in C(2) than in C(1) (58 patients (77.33%) vs. 29 patients (55.77%); p=0.0121).

Regarding the cognitive sphere, the median knowledge score was similar in both groups.

Regarding the meta-cognitive sphere, there was no significant difference between the two clusters in terms of the social interaction score.

Discussion

Our study describes the medico-psychosocial characteristics of the six spheres



Fig. 3. Dendrogram of the bottom-up hierarchical classification. In blue are the clusters of the principal component analysis and in orange the clusters of the exploratory analysis. C: cluster.

defining the allosteric model in PE of patients with SS. To our knowledge, this is the first study evaluating all these medico-psycho-social characteristics classified according to these different spheres for patients suffering from SS. Demographic data that we found in our in-study population, and in particular the sex ratio and mean age of patients, are superimposable on those reported in the literature (8, 36).

The main originality of this work is the cluster approach to define groups of patients using these medico-psychosocial spheres. Two clusters of patients could be individualised, with one group presenting a higher global impact of the disease, including a more severe impairment for the scores of the perceptual, emotional and infra-cognitive spheres, worse physical QoL, and a higher intentionality to participate in a PE programme. This information can lead to consider these aspects to propose PE to groups of patients.

Regarding the intentional sphere, we found that the wish to participate in a

PE programme was positively influenced by reported fatigue, self-reported non-specific symptoms, and poorer QoL, factors of motivation that health professionals should take into account when seeking to improve the practice of PE. Regarding the cognitive sphere, patients had a good knowledge of SS without differences between patients with or without interest to participate in a PE programme, suggesting that differences in patients' willingness to participate in PE were influenced by noncognitive factors.

Regarding the perceptual sphere, patients complained of dry syndrome and fatigue. A parallel can be found between the results reported by McCoy *et al.* (11) and results of our study, emphasising the need to consider these aspects in our medical practice as health care providers, particularly when practising PE.

Regarding the emotional sphere, half of the patients in our study presented anxiety symptoms. Our patients showed predominantly problem-centred coping and internal locus of control, which is

quite consistent with the study of Anderson et al. (41). Most of our patients had low self-esteem, a finding that has not, to our knowledge, been described in the literature about patients with SS. Low self-esteem is correlated with anxiety and depression, and thus with the emotional sphere, as already demonstrated for patients suffering from fibromyalgia (42), systemic lupus erythematosus (43) or systemic scleroderma (44). Our patients suffered from anxiety symptoms, with a higher prevalence than that of depressive symptoms, as found in the literature (13, 37, 45). This psychological impact can influence patients' work disability (46) and therefore their professional status, an important aspect of the meta-cognitive sphere. We also found a definite impact of the disease on social interactions for our population (this is consistent with the fact that coping centred on supporting others was rarely found in the population studied).

Regarding the meta-cognitive sphere, patients considered the information they

Table II. Description and comparison of Cluster 1 and Cluster 2

Variable	Cluster 1 (C1) n=52	Cluster 2 (C2) n=75	р
Jaccard Index	0.84	0.67	
Demographics			
Age (years)	52 ± 17.25	50 ± 13.5	0.77
Duration of illness (years)	7 ± 8.25	8 ± 10.5	0.27
Gender (female)	48 (92.3)	74 (98.7)	0.16
Disabled worker status	7 (13.6)	31 (41.3)	< 0.01*
Complications	28 (53.8)	58 (77.3)	<0.01*
Cognitive sphere			
Knowledge score	8 (1)	8 (1)	0.06
Perceptual sphere	2 12 1 00		0.01*
Protad-SSI score	2.13 ± 1.88	4.76 ± 1.89	<0.01*
ESSPRI score	4.33 ± 2.67	7.67 ± 1.83	<0.01*
BPI score pain severity	0 ± 2.56	5.75 ± 2.5	<0.01*
BPI score pain-related discomfort	0 ± 2.5	5.5 ± 3.25	<0.01*
FSS score	3.28 ± 2.36	0.33 ± 1.39	<0.01*
PIGA score	4 ± 2	1 ± 2	<0.01**
HADS A score	7 + 4	12 + 5	<0.01*
HADS-D score	4+5	12 ± 5 10 + 5	<0.01*
Infra-cognitive sphere			
Problem-centred coping score	25 ± 8.25	25 ± 9.5	0.928
Emotion-centred coping score	16 ± 9	22 ± 10	< 0.01*
Search for social sustain score	17.5 ± 10.25	19 ± 7	0.41
Internal locus score	2.67 ± 0.33	2.33 ± 0.67	< 0.01*
Random locus score	2.33 ± 0.5	2.33 ± 0.5	0.83
External locus score	2.33 ± 0.67	2.33 ± 0.5	0.37
Rosenberg score	34.5 ± 6.25	28 ± 8	<0.01*
Meta-cognitive sphere			
Social interaction score	5.5 ± 2	5 ± 2	0.14
At diagnosis, sufficient information on:			
Diagnosis	33 (63.5)	37 (49.3)	0.15
Course of the disease	25 (48.1)	20 (26.7)	0.01*
Potential complications	21 (40.4)	19 (25.3)	0.08
Treatment	29 (55.8)	29 (38.7)	0.07
Adverse effects	24 (46.1)	23 (30.7)	0.09
During follow-up, sufficient information on:	22 (61 5)	20 (50 5)	0.00
Diagnosis	32 (61.5)	38 (50.7)	0.28
Evolution	24 (46.1)	20 (26.7)	0.04*
Potential complications	22 (42.31)	20 (26.7)	0.08
Treatment	28 (53.8)	29 (38.7)	0.10
Ireatment self-reported adverse effects	26 (50)	27 (36)	0.14
Cross-sectional items	(0.22, (22, ()	26.00 . 10.06	0.01*
Physical score SF-36	69.32 (22.6)	30.88 ± 18.90	<0.01*
Oct DSS accus	00.2 ± 19.33	40.42 ± 10.3	<0.01*
IPO-BREF score	50.3 ± 18.23 44 + 10.25	54 ± 12 54 + 13	<0.01*
Intentional sphere		·· · 10	-0.01
VAS score willingness to participate in PF	45 + 6	7 + 6 5	<0.01*
VAS score willingness to learn skills	5+6	8+4.5	<0.01*
Interest in participating in a PE programme	29 (55.8)	58 (77.3)	0.01*
1 1 0	()	()	

*Statistically significant.

Quantitative variables are expressed as median \pm interquartile range and qualitative variables are expressed as number of patients (percentage).

The 21 clustering variables are underlined. Bold highlighting of values indicates a significant impact of SS.

were given by the health care team to be sufficient, revealing that information is not sufficient for patients to improve their well-being and reduce all the consequences the disease can have on the medico-psycho-social dimensions. As in the literature, our patients had an impaired QoL (8, 9, 37). It has been shown that, for patients with SS, QoL is impacted by the dry syndrome, fatigue, and pain (representing the perceptual sphere) (9, 37), as in our study with a high prevalence of self-reported nonspecific symptoms. Hackett et al were able to highlight links between fatigue and pain in patients with primary SS (38). Taken together, these results lead to the notion of a psychosomatic approach which designates an interdisciplinary medical field exploring the relationships among social, psychological processes, pathological physical manifestations, whether they are functional or organic. It is possible to link fatigue to the infra-cognitive sphere, as shown by Kier et al., who looked at the interaction between fatigue and salutogenesis in lupus patients. Salutogenesis is a model of health developed by the medical sociologist Antonovsky, which, in contrast to pathogenesis, focuses on the factors that promote health (39, 40).

More patients had disabled worker status in one of the two clusters of patients in our study. According to WHO, disability is "a complex phenomenon, reflecting the interaction between features of a person's body and features of the society in which he or she lives". However, the impact on social interactions was similar and significant in both clusters. Indeed, in the study by Dumusc *et al.*, the percentage of patients with SS with a disability pension increased from 16% at the time of diagnosis to 41%two years after diagnosis (47).

Our study suffers from several limitations. It was a single-centre study that focuses only on patients with SS followed up at Lille University Hospital. Data would need to be confirmed by other studies. There were selection biases: the inclusion of patients on a voluntary basis led to a self-selection bias, with patients recruited in consultation not necessarily being representative of the whole SS population. Moreover, the representativeness of the population is questionable given the moderate response rate (31%). A possible explanation for this low rate is that the recruitment was carried out following a classic follow-up consultation and a relatively long time was needed to complete the questionnaires (about 50 minutes), which may have led to a loss of motivation to participate in the study for potential participants. Other factors may also have influenced the intention to participate in patient education workshops within a programme, such as comorbidities and disease activity that we did not evaluate in the study. Some spheres, notably the emotional sphere or quality of life, can be influenced by many factors other than the disease, such as the practice of a sporting activity and the socio-economic environment.

Conclusion

Our study described an SS population in terms of the different spheres of an allosteric model applicable to the practice of PE. Cluster analysis enabled us to identify interesting patterns in the description of the population according to the characteristics used, and revealed a group of patients with a more global impact of the disease on the medico-psycho-social dimensions who showed a greater interest in participating in a PE programme. There was no difference between the two groups in terms of the cognitive sphere (i.e. knowledge of the disease), thus indicating that motivation to participate in a PE programme is influenced by non-cognitive factors. We know that PE is dependent on the patient's willingness to participate, thus placing intentionality at the heart of the practice of PE. Considering intentionality to participate in a PE programme, duration disease, age of the patient and QoL should be more considered to propose to patients to participate in a PE programme. Use of the allosteric model appears promising for future research in PE.

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