Content Validity and Measurement Properties of the PROFAD-SSI-SF Patient-reported Outcome Measure in Patients with Primary Sjögren's Syndrome with Organ Involvement

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Supplementary materials

Supplementary Methods: Refining a disease model for pSS: qualitative analysis

CE discussion forum: secondary analysis

All data from transcripts were coded and analyzed using NVivo qualitative research software v11.0 (NVivo qualitative data analysis software, QSR International Pty Ltd, Victoria, Australia) in accordance with grounded theory analysis methods [1], where concepts emerge from participants rather than imposing an *a priori* theory. Transcripts were reviewed for themes related to the symptoms and impacts of pSS. A saturation analysis determined that 46 interviews were sufficient for capturing all relevant concepts and descriptors of pSS (**Table S1**).

KOL interviews

Interviews with KOLs helped to further refine the disease model and identify concepts of greatest importance to patients with pSS. Trained researchers conducted 90-minute, semi-structured telephone interviews with KOLs who had experience in treating patients with pSS or in patient advocate organizations; KOLs were recruited based on recommendations by the study sponsor. A total of five KOLs took part in the interviews: three rheumatologists with expertise in the treatment and management of patients with Sjögren's syndrome, and two patient advocates, representing the United States and Europe.

During the interview, KOLs reviewed the pSS disease model to confirm the existing content and identify any missing content that should be included. KOLs discussed the appropriateness of the procedures and tests used to establish a pSS diagnosis; commonly reported signs, symptoms and triggers; and the KOLs' perceptions of the

impact of pSS on patients' lives. Interview transcripts were coded and analyzed using thematic analysis and the pSS disease model was updated as needed.

Reference

1. Strauss, AL, and Corbin, JM. *Basics of qualitative research: Grounded theory procedures and techniques.* Newbury Park, CA: Sage Publications; 1990.

Table S1. Saturation grid from the saturation analysis of CE forum interview transcripts

Major theme	Code set 1 Interviews 1–12	Code set 2 Interviews 13–24	Code set 3 Interviews 25–35	Code set 4 Interviews 36–46
Journey to diagnosis	2	-	-	-
Symptoms				
Cutaneous dryness	3	0	2	0
Dysautonomia	7	3	3	0
Fatigue	4	0	0	0
Nasal dryness	0	1	1	0
Ocular dryness	1	0	0	0
Oral dryness	4	1	0	0
Otic dryness	3	0	0	0
Pain	7	1	2	0
Vaginal dryness	1	0	0	0
Other	12	9	5	0
Impact				
Activities of daily living	2	0	0	0
Cognitive	1	0	0	0
Emotional and mental health	10	1	6	0
Physical functioning	4	1	0	0
Productivity	2	0	0	0
Relationships	1	0	0	0
Social functioning	2	0	0	0
Other	10	2	2	1
Healthcare utilization	16	5	0	0
Medication use				
Anti-depressants	4	1	1	0
Anti-fungals	1	2	1	0
Anti-rheumatics	4	1	0	0
Eye drops	3	0	0	0
Heart medications	2	0	6	0
Hormones	2	1	0	0
Inhalers	0	0	6	0
Muscle relaxants	0	1	2	0
Pain medication	10	2	0	0
Sleep aids	1	1	1	0
Steroids	5	0	1	0
Other	15	3	5	1
Non-medication treatment	12	4	9	0
Side effects of medication	13	10	6	0
Self-reported comorbidities	51	12	16	1

Triggers/exacerbating factors	14	2	0	0
Total number of new codes:	229	64	75	3
New codes per set, %:	61.7	17.3	20.2	0.8

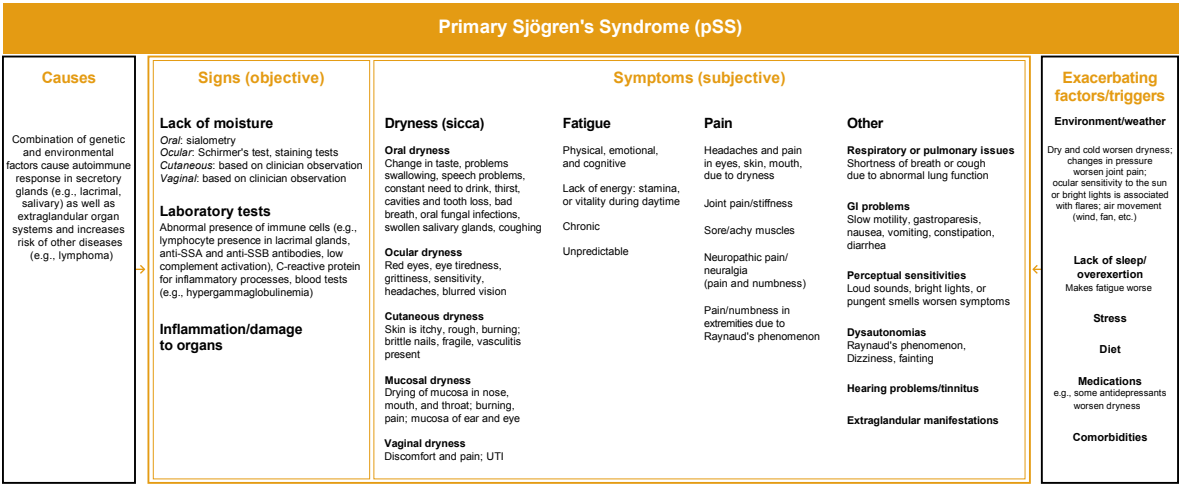
To assess saturation, interview transcripts were divided into quartiles and reviewed to determine the point in data collection at which each sub-theme was elicited. The number within each cell represents the number of new sub-themes elicited for its respective code set.

CE, concept elicitation.

Table S2. Cognitive debriefing interviews: Participant demographics (N=20)

	N (%)
Age, years	
35–44	7 (35.0)
45–64	7 (35.0)
65+	6 (30.0)
Sex	
Male	5 (25.0)
Female	15 (75.0)
Race/ethnicity	
White	13 (65.0)
African American	1 (5.0)
Hispanic	4 (20.0)
Asian	2 (10.0)
Highest level of education	
Some College	7 (35.0)
Associates Degree	2 (10.0)
College Graduate	10 (50.0)
Graduate Degree	1 (5.0)

Figure S1. Final disease model of pSS



Impact on functioning and well-being

	Dryness (sicca)	Fatigue	Pain	Other
Primary dryness impact				
Oral health problems	•			
Environmental limitations (cold, dry, etc.)	•			•
Need for adaptive behaviors (e.g., applying ointments, drinking water before bed, applying eye drops, etc.)	•			
Problems eating, swallowing	•		•	•
Sexual dysfunction (female)	•		•	
Sleep problems	•	•	•	
Physical functioning				
Difficulty with exertion	•	•	•	•
Limitations on ADLs	•	•	•	
Limitations on leisure activities	•	•	•	•
Need for frequent breaks/rest		•		
Social functioning				
Embarrassment in social situations	•			
Impact on relationships	•	•	•	
Limitations on social activities	•	•	•	
Vocational				
Limitations at work	•	•	•	•
Need for special accommodation	•	•	•	•
Psychological				
Cognitive impairment		•		
Emotional difficulties	•	•	•	•
Irritability	•	•	•	•
Mood (anxiety/depression)	•	•	•	
Poor self-perception due to limitations	•	•	•	•

ADL, activities of daily living; GI, gastrointestinal; SSA, Sjögren's syndrome A; SSB, Sjögren's syndrome B; UTI, urinary tract infection