

## RESEARCH ARTICLE

# Illness Experiences in Women with Oral Dryness as a Result of Sjögren's Syndrome: The Patient Point of View

Gonzalo Rojas-Alcayaga<sup>1,2\*</sup>, Andrea Herrera Ronda<sup>2</sup>, Iris Espinoza Santander<sup>1</sup>, Claudia Bustos Reydet<sup>2</sup>, Matías Ríos Erazo<sup>2</sup>, Pamela Wurmman<sup>3</sup>, Francisca Sabugo<sup>3</sup> & Rinie Geenen<sup>4</sup>

<sup>1</sup>Department of Oral Pathology, Faculty of Dentistry, Universidad de Chile, Santiago, Chile

<sup>2</sup>Behavioral Science Area, Institute for Research in Dental Science, Universidad de Chile, Santiago, Chile

<sup>3</sup>Rheumatology Section, Hospital Clínico Universidad de Chile, Santiago, Chile

<sup>4</sup>Department of Psychology, Utrecht University, Utrecht, the Netherlands

## Abstract

**Background:** Sjögren's syndrome and the associated dryness can have multiple consequences. The aim of the present qualitative study was to give an in-depth account of the life experiences of women with primary Sjögren's syndrome (pSS) and health-related behaviours, and to summarize these experiences in an integrated model.

**Methods:** Twelve women diagnosed with pSS who regularly attended the Hospital of the University of Chile participated in detailed interviews. The data were analysed using qualitative methods based on the principles of grounded theory.

**Results:** Selective coding identified three categories: illness experience, social interaction and psychological response. An integrated model was developed connecting these dynamic aspects and suggesting how they could lead to a life cycle crisis in cases of maladjustment. We found that problem-solving strategies, reconstruction of identity, acceptance and a social support may prevent this life cycle crisis.

**Discussion:** Xerostomia and other consequences of pSS can have a profound influence on daily life. However, the severity of the consequences depends on individual experiences with the illness, social influences and the psychological responses of the patient. Physicians, dentists and other healthcare professionals can help the patient by listening to their problems and exploring solutions based on a psychological approach.

## Keywords

Sjögren's Syndrome; health experiences; oral dryness

## \*Correspondence

Gonzalo Rojas Alcayaga, Facultad de Odontología, Universidad de Chile, Sergio Livingstone 943, Independencia, Santiago, Chile. Tel: +56 2 2978 1844; Fax: +56 2 2978 1702

Email: grojasalcayaga@yahoo.es

Published online 6 April 2016 in Wiley Online Library (wileyonlinelibrary.com) DOI: 10.1002/msc.1134

## Background

Sjögren's syndrome is a rheumatic autoimmune disease that impairs different bodily organs and tissues, particularly the lacrimal and salivary glands (Tincani et al., 2013). The systemic manifestations, although rare, can compromise musculoskeletal, cutaneous, pulmonary, gastrointestinal, renal,

neurological and haematological function (Carsons, 2001). The annual incidence rate of primary Sjögren's syndrome (pSS) is approximately 3.1 per 100,000 individuals; it is higher in women and those over the age of 55 (Kvarnström et al., 2015; Thomas et al., 1998), with a prevalence of 0.019–0.6% among European women (Anagnostopoulos

et al., 2010; Bowman et al., 2004b; Maldini et al., 2014). Similarly to patients with rheumatoid arthritis and systemic lupus erythematosus, patients with pSS experience a decline in several dimensions of health-related quality of life (HRQOL). Compared with healthy individuals, patients with pSS exhibit a decreased ability to engage in aerobic exercise, diminished general functional capacity, joint problems and higher levels of fatigue, pain, anxiety and depression (Bowman et al., 2004a; Strömbeck et al., 2003; Westhoff et al., 2011).

Studies using instruments such as the 36-Item Short Form (SF-36) Health Survey have found that many or all domains are affected (Meijer et al., 2009; Segal et al., 2009), most notably physical functioning, physical role, emotional role and general health (Stewart et al., 2008). Fatigue levels are high, with a prevalence of 54% of patients reporting severe fatigue (Overman et al., 2015). Sexual activity and sexual desire are also affected in patients with pSS, owing to vulvar and vaginal dryness (Maddali Bonghi et al., 2013). The effects of pSS on HRQOL are even more pronounced when symptoms of anxiety and depression are present. However, social support can mitigate the impact of depression (Inal et al., 2010). Furthermore, psychological, as well as physical, aspects of the disorder affect HRQOL (Champey et al., 2006). Although end-organ damage is uncommon in pSS, the degree of functional ability is high. Oral symptoms are considered in pSS diagnosis (Vitali et al., 2002), and both xerostomia (dryness of the mouth) and damage to oral tissues (such as tooth decay, yeast infections and gingivitis) are the main symptoms that affect patients (Sutcliffe et al., 1998) and could be important contributing factors to the impaired HRQOL.

Having a dry mouth makes it difficult to eat without additional liquid, diminishes taste capacity and may lead to a burning sensation in the mouth (Kamel et al., 2009). The low secretion of saliva also promotes the development of dental caries, often in unusual locations, as well as oral candidiasis (Tincani et al., 2013). Furthermore, sicca (dryness) symptoms may correlate with greater fatigue (Haldorsen et al., 2011) and pain (Hartkamp et al., 2011) in pSS.

Although the findings of studies that have evaluated HRQOL quantitatively in patients with this disease have made important contributions to the development of clinical strategies, the quantitative nature of these investigations has not enabled them fully to capture the experience of suffering from a chronic illness

such as pSS. Qualitative methodology is particularly focused on how the world is understood, experienced and produced by individuals' personal lives, their behaviours, their interactions, the dynamics of their social processes and their perspectives about their own worlds, and also by what people think and what this thinking means and implies (Vasilachis de Gialdino, 2009).

The aim of the present qualitative study was to give an encompassing in-depth account of the life experiences of women with pSS and health-related behaviours, and to summarize these experiences in an integrated model.

## Methods

### Design

The investigation was an exploratory relational study employing qualitative analyses derived from grounded theory, which shares many features with other qualitative methods such as phenomenological interviewing (Bristowe et al., 2015). We chose grounded theory over other methods because we wanted to develop an integrated model from the input of patients, with as little interpretation bias as possible. Grounded theory is a qualitative method to explore phenomena and lived experiences in people, with the primary propose of developing theory 'from the ground up'. It means that the researcher enters the field naïve, allowing theory to emerge from the data (interviews) through an iterative and clearly defined process of coding, conceptualization, categorization and theory development (Boeije, 2010; Bristowe et al., 2015). Grounded theory is frequently used in medical research to generate in-depth information (meanings, understanding and experiences) because prescribed steps ensure rigorous processing and avoid the fact that conclusions are biased by personal understanding, interpretation and analysis.

Data analysis was based on the constant comparative method. This is a general approach for analysing qualitative data using explicit coding and analytical procedures to generate and suggest categories, properties and hypotheses about general problems. Properties may be causes, conditions, consequences, dimensions, types or processes relating to the situation of interest (Glaser and Strauss, 1967).

The present study took place over eight months during 2013–14. The focus of the study was on the experiences, beliefs and values of patients with pSS in a hospital environment. As access to the ways of understanding and

living with the disease requires privacy to encourage an open, fluid, spontaneous dialogue, without perceived barriers, we chose a technique based on the conversation – specifically, an in-depth interview.

## Participants

Interviews were carried out in 12 women with pSS who regularly attended the Rheumatology and Dental Maxillofacial Service of the Clinical Hospital of the University of Chile. Female patients were chosen because the prevalence of pSS in women is about nine times as high as the prevalence in men (Qin *et al.*, 2015). The size of the purposeful sample was determined by the principle of theoretical saturation, which means that data collection is finished at the point at which additional data from interviews seems to be redundant in the light of the emergent model (Kostova *et al.*, 2014). All participants were adult Chilean women, aged over 45 years and with a specialist-confirmed diagnosis of pSS according to established criteria (Vitali *et al.*, 2002).

The patients were identified and invited to participate in the study by two rheumatologists in the Rheumatology Section and one dentist in the Dental Maxillofacial Service. The aim of the study – to get an insight into the life experiences of women with pSS and their health-related related behaviours – and the procedures were explained to the patients by the first author (GR) prior to the interview. Participation was voluntary and was initiated after informed consent that complied with the ethical standards of the Helsinki Declaration. The Ethics Committee of the Faculty of Dentistry of the University of Chile approved the study.

## Data collection and analysis

The first author (GR) interviewed 12 women with SS using a semi-structured questionnaire. A script (Box 1) that included the following topics was used: causal attributions of disease, perception of the progression of the disease, daily difficulties, own resources for illness management, social relationship, family and job impact, treatment experiences, emotional response to illness, and experience with the medical care. The script was developed by two of the authors (GR, IE) based on the background literature about pSS symptoms and HRQOL, and it was then discussed with the other researchers (AH, MR, CB).

## Box 1. Script of the in-depth interviews

---

### *Background information*

1. Can you tell me your name, age, address, work and education level?
2. What was your main reason for accepting the invitation to participate in this study?
3. When were you diagnosed with Sjögren's syndrome?

### *Core interview*

4. How did you first notice your illness? How did you realize that you were ill?
5. How has your life been affected by the disease?
6. What daily activities did you stop doing or modify because of the disease?
7. How has the disease affected your social relationships – i.e. the activities with your family, friends and partner?
8. Who do you share your illness experience with?
9. What is your opinion about the treatment?
10. What would you say to other people who have just been diagnosed with the same disease?
11. What new or important experiences have you obtained from your illness?
12. What do you think is the cause of this disease? Do you link the disease onset with something that happened?
13. What aspects of your medical care are the best and worst for you?

### *Closing interview*

14. Would you like to say anything else?
  15. How did you feel during this conversation?
- 

The script was split into three sections. The first section included three general background questions and the goal was to introduce the interview and to generate a warm environment. The second section was the body of the interview, and this part included the ten main questions about the illness experience that were explored in-depth. Patients were allowed to introduce other topics. The last section included two questions. Participants were urged to come up with new topics. The goal was also to close the interview. Individual interviews, which lasted between 60 and 90 minutes each, were conducted in the same hospital where patients were being treated, in a private room with good lighting and noise isolation. All interviews were video-recorded, and the 20 hours of recordings produced were transcribed by an assistant who did not know the research question.

The interviews were analysed by two researchers (GR and AH) using the constant comparative method consisting of three steps: open, axial and selective coding (Boeije, 2010). In the open coding phase, significant statements about illness experiences were identified as a unit of meaning (incidents). Categories and their properties emerged from initial comparisons of codes. In the next step, we performed comparisons between

incidents and categories and their properties. Finally, properties and categories were integrated, discovering underlying uniformities from the original set.

## Results

Most of the 12 women included in the study were housewives and the number of years since their pSS diagnosis ranged from one to 12. The characteristics of the sample are shown in Table 1.

Open and axial coding established three categories relating to the experience of pSS:

- Illness experience: uncertainty, strangeness, loss and symptoms
- Social interaction: isolation versus integration
- The psychological response to the loss of health: adjustment versus maladjustment.

The central argument that connects the categories is that pSS can involve a life cycle crisis that is not clearly understood or supported by the social environment.

### Illness experience: uncertainty, strangeness, loss and symptoms

pSS is a disease that is hardly recognized by the patient. When it is diagnosed, the patients are stunned, without knowing exactly what they have and therefore not knowing how to react. Some relate it to cancer and others to total and permanent disability. Therefore, one of the first reactions is characterized by catastrophic thoughts and emotional reactions of fear, which in any case are based on the actual characteristics of the disease:

'I immediately asked the doctor: will I be disabled or how long will I live?' (AS, 63-year-old)

In addition, the sparse knowledge of the characteristics of pSS in patients' close social environment adds to the experience of strangeness. People in patients' social networks express ignorance about the condition and give it a connotation of a 'strange phenomenon', which increases patients' feeling of living with or having something very strange:

'The people to whom you mention the Sjögren, say it is something they have never heard of. This disease is rare. People say: "I have a dry mouth", but never tell [you that] they have Sjogren's syndrome'. (JO, 73-year-old)

The damage to the oral mucosa and the extreme reduction in saliva secretion created a longing for the same amount of saliva as before the illness. One patient recounted:

'My dream is to be able to spit and cry a lot of tears again'. (MM, 49-year-old)

Another patient remarked similarly:

'I would give all the gold in the world just to have a little more moisture [in my mouth].' (ML, 66-year-old)

However, other individuals gradually adjusted to hyposalivation. Adaptation may depend on the degree to which saliva secretion is affected, as well as on how quickly the hyposalivation progresses.

**Table 1.** Characteristics of the sample

Patients' initials	Age	Current activity	Years from diagnosis	Years with symptoms of oral dryness
AH	45	Housewife (retired)	12	13
MM	49	Housewife	5	7
MAO	58	Housewife (retired)	3	3
MO	61	Housewife (retired)	5	1
AS	63	Housewife (retired)	7	5
CO	64	Paramedic	2	3
SCC	65	Housewife (retired)	4	Many years (unspecified)
ML	66	Housewife (retired)	7	9
SV	70	Housewife	1	20
EI	70	Housewife	Unspecified	Many years (unspecified)
JO	73	Seamstress	4	Many years (unspecified)
GM	73	Housewife	1	1

pSS produced general, as well as oral, signs and symptoms. Oral symptoms included pain, a burning sensation in the mouth and dryness of the oral mucosa. The latter created difficulty in moving the lips and mouth:

'It gets so dry that sometimes my lips get stuck together, and I have to pull them apart because my lips won't part'. (MO, 61-year-old)

Complications of dry mouth also included partial or total loss of the sense of taste and smell:

'Sometimes I don't taste the flavour of food, and I also find that I am losing my sense of smell'. (SV, 70-year-old)

They also included difficulty in eating, swallowing and speaking:

'I talk with my sisters, for I don't know how long, and immediately they notice that I start to ... that my mouth is dry and my tongue gets stuck and I can't articulate speech well'. (MM, 49-year-old)

The combination of physical deterioration with the emotional reaction of loss revealed the perception of limitation across domestic, work and social realms. Symptoms impose serious limitations in various household activities, which patients live with, with varying degrees of difficulty, and experience as a loss of their usual capabilities:

'There are meals [that] I [am unable to] taste; there are foods [that] I [cannot tell] if they are salty or not ... As I [have been a] housewife since I [was] a girl, I know the amount of salt, [and] all that ... but there are foods that I can't [experience as] tasty 'though everyone says that it is delicious, but I [cannot] taste ... ' (EI, 70-year-old)

### **Social interaction: Isolation versus integration**

Patients' perceptions of the quality of their social interactions depended on factors such as family support, the characteristics of the physical surroundings in which the social interactions take place and the possibility of

an adequate performance of oral functions and other physical activities. As one patient commented:

'There were cigarette butts, and I coughed for more than an hour; people think it's for another reason – they don't think it's because of that [irritation of the airways by cigarette smoke]'. (MAO, 58-year-old)

Difficulties in maintaining social relationships tended to create social isolation, which reinforced the negative aspects of the disease and led to depressed mood or even depression. In one case, a patient commented:

'My life isn't like it was before. I live a very limited life – it's shutting myself up in the house and coming to the hospital'. (MAO, 58-year-old)

Social support was fundamental because feeling understood and supported allowed individuals to maintain their activities and sustain their social networks:

'It's just that you can't isolate yourself. All of my friends know about the problem, so when I go over to their houses, if we are drinking tea, they know that I am going to drink two or three cups; I mean, they are all prepared – in that aspect, I have a lot of support'. (ML, 66-year-old)

By contrast, when patients perceived a lack of understanding from others, they tended to isolate themselves:

'I was ashamed to go out; I would be talking and I had to have a glass of water with me and everyone asked me why I had it [the dry mouth]'. (MM, 49-year-old)

Withdrawal from social activities and the isolation they may fall into is not because of rejection by others, but rather as a result of the discomfort or embarrassment that patients may experience for presenting certain symptoms, functional limitations or aesthetic defects, or of the frequent explanations that they have to give about suffering with SS:

'I have often stopped going out because I'm embarrassed, because sometimes I talk and I'm bleeding from the gums and it is annoying. Another thing is that sometimes my breath is nasty, very nasty, although I did brush my teeth'. (EI, 70-year-old)

Participating in a social activity functions as a protective element and motivates patients to cope with the disease. Thus, the suffering of pSS does not preclude the ability to enjoy other aspects of life and to avoid an over-identification as a sick person:

'I also participate in a spiritual activity ... worshipping God ... all we do is ask for the humanity... but it is something that satisfies me very much. I have participated for many years in this ... yes, everybody talks there, talks about life'. (JO, 73-year-old)

### The psychological response to the loss of health: adjustment versus maladjustment

The psychological response to pSS was related to the loss of health, especially the oral functions, which produced feelings of sadness, abandonment and powerlessness. This reaction was also due to the characterization of pSS as an incurable, progressive and degenerative disease and was supported by information obtained both from objective and valid sources, such as doctors and the health system, and also from informal sources in the community. The portrayal of the disease as ill defined and enigmatic led individuals to ascribe its origin to magical-religious, climatological or emotional conditions, such as excessive crying or guilt. As one patient recounted:

'It was when my daughter died, so of course then, and after, after a while, I felt that my eyes were all dry'. (SV, 70-year-old)

In regard to climatological issues, a patient stated:

'It was right in the summer, when it was so hot ... and even in the heat I had to go out to see him [her husband, when he was sick in a hospital], and when I returned, it was hot, so that's why I blamed the heat'. (GM, 73-year-old)

The psychological response of sadness and discouragement was exhibited as grief for the loss of previous good health, which challenged the personal resources available. Patients who accepted the reality of the condition and focused on positive elements adopted problem-solving strategies to treat the symptoms (e.g. seeking a means to moisten the mouth and eyes, and

changing patterns of physical activity) or restructured their social, work and domestic activities:

'So far, I have accomplished many things, like having my mouth a little more moist, so I think that, with more time, I'm going to get back to where it was before – well, not at the same level, but ... I have faith that it's going to happen'. (MM, 49-year-old)

Another patient commented:

'There are many people who just lie down and die, and they sink with a health problem – I do not!' (ML, 66-year-old)

These responses reflected an effort to accept the new condition and eventually restructure their identity.

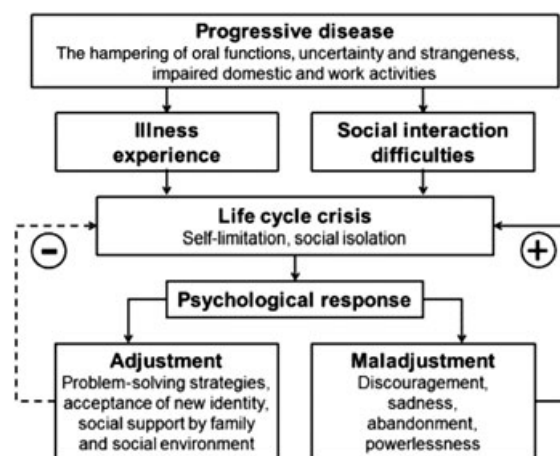
The emotional impact of the disorder motivated a search for understanding from family members, as partners, children, other family members and friends could provide patients with crucial social support, such as emotional support and assistance with treatment. One patient commented:

'My entire family is aware of the problem, and they all try to help me not to talk too much, keep me calm'. (ML, 66-year-old)

### Core category: Life cycle crisis

Figure 1 presents a dynamic model summarizing the consequences of xerostomia for emotional and social functioning. The central argument that connects the categories is that pSS involves a life-cycle crisis that is not clearly understood or supported by the social environment. This interruption of the life cycle may involve individuals' identity and individual functioning as well as social participation – for example, in leisure activities, work and relational functioning. Two core dimensions of the life cycle crisis are: *self-limitation and social isolation*.

*Self-limitation* is experienced by patients in everyday activities, social interaction and emotional expression. In the present study it was related to the possible restructuring of identity. Both the oral and general symptoms they experienced led patients to perceive their lives and social environments differently and made it difficult to maintain their previous social roles. Individuals experienced the loss and grief from not being *'the same*



**Figure 1.** An integrated model summarizing the life experiences of women with primary Sjögren's syndrome and health-related behaviours. The model shows that the core category *life cycle crisis* is modulated by illness experience and social interaction in relation to the Sjögren's syndrome. When uncertainty and strangeness are experienced by a patient with low social interaction, a life cycle crisis occurs, with self-limitation and social isolation as its main features. Psychological maladjustment brings on these experiences. A positive adjustment occurs when problem-solving strategies are used by patients, they accept their new identity and a good social support network is available. Thus, adaptive psychological responses reduce the experiences of self-limitation and social isolation and cause the life cycle to continue without too much interruption

as before'. The resulting questioning of self-concept and effect on self-esteem will have had an impact on identity.

To work through the grief, individuals had to adopt problem-solving strategies, restructure and accept their new their identity and perceive themselves as effective, in spite of their physical limitations. This is the situation for some patients – that, in spite of physical symptoms, they carry on with work or social activities because they feel useful to others.

In this adjustment process, social networks that provided emotional support and treatment assistance were fundamental. Psychological adaptation was more likely to occur when family and friends provided understanding and support. *Social isolation* is a frequent issue in these patients, mainly when they do not accept their new health status or when the people surrounding them do not understand the pSS condition. However, if patients restructure their identity and feel accepted by others, social integration is possible.

## Discussion

The aim of the present study was to explore the illness experience of patients with pSS, using a qualitative approach to identify the main psychosocial issues involved. Analysis of the information supplied by participants identified three main categories: *experience of the illness, psychological response and social interaction*.

The category *experience of the illness* was described as the uncertainty and strangeness associated with living with a complex disease marked by reduced saliva secretion. The category *psychological response* involved feelings of sadness and abandonment due to the characterization of the disease and the associated xerostomia as an incurable and progressively worsening condition. Finally, difficulties with speech, swallowing and taste affected *social interaction* necessitating the restructuring of social roles. The above categories contributed to the identification of a core category: *vital cycle break due to incurable disease*. This break has two dimensions: *self-limitation and social isolation*.

Self-limitation was perceived to be related to the social isolation and interference with social role due to the difficulty in engaging in activities that required the secretion of saliva, such as speaking, chewing, swallowing and tasting food. This finding is consistent with the results of studies that used measures such as the SF-36 to evaluate HRQOL in which patients with pSS reported poorer social functioning due to physical and emotional difficulties compared with control groups. They also found that the quality of life of individuals with xerophthalmia and xerostomia was more affected in all domains, notably in reduced intimate/sexual contact, fewer opportunities for social interaction, decreased productivity at work and fewer employment opportunities (Ikebe et al., 2007; Strömbeck et al., 2003; Stewart et al., 2008). These

results are in agreement with those of the present study, which found that suffering from xerostomia affected social roles and sexual intimacy with a partner.

Self-limitation and social isolation were preceded by the difficulty of working through grief to restructure identity. If the person continued to believe that she had to be the same as she was prior to the illness, she would see herself as self-limited and thus become frustrated in both personal and social domains. However, the interviews indicated factors that assisted these individuals to restructure their identity: the immediate social support of family and friends and the characterization of pSS, which was shaped by information from the community as well as clinicians. Unlike diseases such as AIDS, Alzheimer's disease and diabetes, pSS is a non-fatal disease with a low prevalence in the population. It thus lacks the social impact to generate the knowledge that would make it understandable to patients and others. Without a clear understanding of the disorder, individuals with the illness feel like outsiders, and this inhibits the grief process.

The course of chronic illnesses tends to worsen if only limited social support is available. It was observed that patients with pSS had less social support than healthy people or individuals with lymphoma (Karaiskos *et al.*, 2009). A previous study found that middle-aged women who did not interact with friends, and single men who lived alone were more likely to develop depression, revealing the importance of social support for maintaining good mental health (Tanaka *et al.*, 2011). In addition, the same study found that patients with chronic illness and those who perceived their health as poor were also at greater risk of depression. Finally, a study found that negative social support, such as not recognizing or acknowledging a condition or even invalidating a patient's experience, is associated with poor health (Kool *et al.*, 2013). The present study showed that patients emphasize the importance of social support and low invalidation rates by their social environment; patients perceived themselves as less self-limited when family and friends who were with them understood the nature of the illness.

Consistent with other research (Perno, 2007), the present study found that xerostomia was not a minor condition or irrelevant to the daily life of individuals with pSS. On the contrary, patients reported that it clearly affected their daily life, HRQOL and ability to live a satisfying life. The healthcare team thus plays a critical role in an individual's psychological response to the illness. They are expected to provide appropriate

treatment that includes clear and precise information concerning the disorder, as well as emotional support that allows the patient's concerns to be heard. Keystones of education in people with a chronic disease are the concepts of the expertise, independence and empowerment of patients (Snow *et al.*, 2013). It is important for healthcare professionals to encourage and help the patient to adjust to the challenges of daily life because patients' self-limitations interfere not only with medical treatment, but also with the opportunity to live a productive and satisfying life.

Qualitative research could be an important link between medical science, clinical practice, and patients and doctor experiences in health care (Lempp and Kingsley, 2007). In-depth interviews are an opportunity to access the subjective world of patients in an open way and to discover unknown aspects of their general life and the impact of diseases (Taylor and Bogdan, 1987). Usually, in the clinical setting, there is not enough time to let patients speak about their doubts or preferences, and this hampers the ability of physicians and dentists to discuss the important issues for management of the disease (Rahman and Tasnim, 2007). While fatigue, body pain, dryness of the mouth and eyes, and tissue damage are aspects of the disease that are well recognized by physicians and dentists, patient behaviours and psychosocial issues should also be a frequent topic of conversation by physicians and other health professionals (Rahman and Tasnim, 2007). In-depth knowledge of the patient's perspectives is helpful for realizing new management strategies (Bergsten *et al.*, 2009) and treatment planning, and to improve adherence and patient satisfaction (Zakrzewska, 2013). In some situations, the beliefs of patients with pSS could be considered as inadequate or misleading, and physicians and health professionals may try to modify them. However, patients' beliefs should be heard and validated because the patient point of view is an extraordinary resource that could provide a better approach, not only in the diagnosis and treatment of the disease, but also to improve HRQOL.

A potential limitation of any qualitative study is researcher bias. In the present study, results were interpreted by researchers without patients being involved in the coding process. However, themes were identified independently by two researchers and discussed thoroughly to prevent missing any relevant themes. Another possible limitation was the fact that no external peer review was considered in the coding process. However, the model was discussed with co-authors, enabling a broader



perspective to be reached, and the final model was attained as a consensus. Patients were involved only as respondents in the interviews; they were not part of the research team that developed the interview and coded the results. Another limitation of the present study was that only women participated. Some findings about illness experience are closely related to the female gender, such as housekeeping role problems and specific sexual problems. These and others fields of experiences could be associated with self-esteem or self-concept.

To summarize, the present study showed that uncertainty and strangeness associated with a progressive and harmful disease, impaired work and domestic activities and the hampering of oral functions foster a possibility of a life cycle crisis in cases of maladjustment, while problem-solving strategies, acceptance of one's new identity and social support may prevent this situation. Social isolation and self-limitation are the main consequences when the psychological response is characterized by discouragement, sadness, abandonment and powerlessness. This integrated model suggests that not only are the physical aspects important for the suitable control of pSS, but also that the psychosocial issues must be considered in anamnesis. Physicians, dentists and other healthcare professionals can help the patient by listening to their problems and exploring solutions based on a psychological approach. Health professional staff can be part of the patient's social network and provide the patient with cognitive, behavioural, informative and emotional support, avoiding the perception of abandonment and social isolation. These patients require counselling about active coping, with problem-solving strategies that allow an adjustment to their new life conditions.

## Acknowledgements

This study was supported by Domeyko's Funds for Investigation, University of Chile.

## REFERENCES

Anagnostopoulos I, Zinzaras E, Alexiou I, Papathanasiou AA, Davas E, Koutroumpas A, Barouta G, Sakkas LI (2010). The prevalence of rheumatic diseases in central Greece: A population survey. *BMC Musculoskeletal Disorders* 11: 98.

Bergsten U, Bergman S, Fridlund B, Alfredsson L, Berglund A, Petersson IF, Arvidsson B, EIRA Study Group (2009). Patients' conceptions of the cause of

their rheumatoid arthritis: A qualitative study. *Musculoskeletal Care* 7: 243–55.

Boeije H (2010). *Analysis in Qualitative Research*. London: SAGE Publications.

Bowman SJ, Booth DA, Platts RG (2004a). Measurement of fatigue and discomfort in primary Sjögren's syndrome using a new questionnaire tool. *Rheumatology* 43: 758–64.

Bowman SJ, Ibrahim GH, Holmes G, Hamburger J, Ainsworth JR (2004b). Estimating the prevalence among Caucasian women of primary Sjögren's syndrome in two general practices in Birmingham, UK. *Scandinavian Journal of Rheumatology* 33: 39–43.

Bristowe K, Selman L, Murtagh FE (2015). Qualitative research methods in renal medicine: An introduction. *Nephrology, Dialysis, Transplantation* 30: 1424–31.

Carsons S (2001). A review and update of Sjögren's syndrome: Manifestations, diagnosis and treatment (2001). *American Journal of Managed Care* 7: 433–43.

Champey J, Corruble E, Gottenberg JE, Buhl C, Meyer T, Caudmont C, Bergé E, Pellet J, Hardy P, Mariette X (2006). Quality of life and psychological status in patients with primary Sjögren's syndrome and sicca symptoms without autoimmune features. *Arthritis and Rheumatism* 55: 451–7.

Glaser BG, Strauss AL (1967). The constant comparative method of qualitative analysis. In Glaser BG, Strauss AL (eds). *The Discovery of Grounded Theory. Strategies For Qualitative Research*. New Brunswick, NJ: Aldine Transaction.

Haldorsen K, Bjelland I, Bolstad AI, Jonsson R, Brun JG (2011). A five-year prospective study of fatigue in primary Sjögren's syndrome. *Arthritis Research and Therapy* 13: R167.

Hartkamp A, Geenen R, Kruize AA, Bossema ER, Godaert GL, Bootsma H, Bijlsma JW, Derksen RH (2011). Serum dehydroepiandrosterone sulphate levels and laboratory and clinical parameters indicating expression of disease are not associated with fatigue, well-being and functioning in patients with primary Sjögren's syndrome. *Clinical and Experimental Rheumatology* 29: 318–21.

Ikebe K, Matsuda K, Morii K, Wada M, Hazeyama T, Nokubi T, Ettinger R (2007). Impact of dry mouth and hyposalivation on oral health-related quality of life of elderly Japanese. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics* 103: 216–22.

Inal V, Kitapcioglu G, Karabulut G, Keser G, Kabasakal Y (2010). Evaluation of quality of life in relation to anxiety and depression in primary Sjögren's syndrome. *Modern Rheumatology* 20: 588–97.

Kamel UF, Maddison P, Whitaker R (2009). Impact of primary Sjögren's syndrome on smell and taste: Effect on quality of life. *Rheumatology* 48: 1512–4.

Karaikos D, Mavragani CP, Makaroni S, Zinzaras E, Voulgarelis M, Rabavilas A, Moutsopoulos HM (2009).

- Stress, coping strategies and social support in patients with primary Sjögren's syndrome prior to disease onset: A retrospective case-control study. *Annals of the Rheumatic Diseases* 68: 40–6.
- Kool MB, van Middendorp H, Lumley MA, Bijlsma JW, Geenen R (2013). Social support and invalidation by others contribute uniquely to the understanding of physical and mental health of patients with rheumatic diseases. *Journal of Health Psychology* 18: 86–95.
- Kostova Z, Caiata-Zufferey M, Schulz PJ (2014). The impact of social support on the acceptance process among RA patients: A qualitative study. *Psychology and Health* 29: 1283–302.
- Kvarnström M, Ottosson V, Nordmark B, Wahren-Herlenius M (2015). Incident cases of primary Sjögren's syndrome during a 5-year period in Stockholm County: A descriptive study of the patients and their characteristics. *Scandinavian Journal of Rheumatology* 44: 135–42.
- Lempp H, Kingsley G (2007). Qualitative assessments. *Best Practice and Research. Clinical Rheumatology* 21: 857–69.
- Maddali Bonghi S, Del Rosso A, Orlandi M, Matucci-Cerinic M (2013). Gynaecological symptoms and sexual disability in women with primary Sjögren's syndrome and sicca syndrome. *Clinical and Experimental Rheumatology* 31: 683–90.
- Maldini C, Seror R, Fain O, Dhote R, Amoura Z, De Bandt M, Delassus JL, Falgarone G, Guillevin L, Le Guern V, Lhote F, Meyer O, Ramanoelina J, Sacré K, Uzunhan Y, Leroux JL, Mariette X, Mahr A (2014). Epidemiology of primary Sjögren's syndrome in a French multiracial/multiethnic area. *Arthritis Care and Research* 66: 454–63.
- Meijer JM, Meiners PM, Huddleston Slater JJ, Spijkervet FK, Kallenberg CG, Vissink A, Bootsma H (2009). Health-related quality of life, employment and disability in patients with Sjogren's syndrome. *Rheumatology* 48: 1077–82.
- Overman CL, Kool MB, Da Silva JA, Geenen R (2015). The prevalence of severe fatigue in rheumatic diseases: An international study. *Clinical Rheumatology* 2015 Aug 15. [Epub ahead of print].
- Perno M (2007). Xerostomia and quality of life. *International Journal of Dental Hygiene* 5: 60–1.
- Qin B, Wang J, Yang Z, Yang M, Ma N, Huang F, Zhong R (2015). Epidemiology of primary Sjögren's syndrome: a systematic review and meta-analysis. *Ann Rheum Dis* 74: 1983–9. DOI: 10.1136/annrheumdis-2014-205375.
- Rahman A, Tasnim S (2007). Twelve tips for better communication with patients during history-taking. *Scientific World Journal* 7: 519–24.
- Segal B, Bowman S, Fox P, Vivino F, Murukutla N, Brodscholl J, Ogale S, McLean L (2009). Primary Sjögren's syndrome: Health experiences and predictors of health quality among patients in the United States. *Health and Quality of Life Outcomes* 7: 46–54.
- Snow R, Humphrey C, Sandall J (2013). What happens when patients know more than their doctors? Experiences of health interactions after diabetes patient education: A qualitative patient-led study. *BMJ Open* 11: e003583.
- Stewart C, Berg K, Cha S, Reeves W (2008). Salivary dysfunction and quality of life in Sjögren's syndrome. A critical oral–systemic connection. *Journal of the American Dental Association* 139: 291–9.
- Strömbeck B, Ekdahl C, Manthorpe R, Jacobsson LT (2003). Physical capacity in women with primary Sjögren's syndrome: A controlled study. *Arthritis and Rheumatism* 49: 681–8.
- Sutcliffe N, Stoll T, Pyke S, Isenberg DA (1998). Functional disability and end organ damage in patients with systemic lupus erythematosus (SLE), SLE and Sjögren's syndrome (SS), and primary SS. *Journal of Rheumatology* 25: 63–8.
- Tanaka H, Sasazawa Y, Suzuki S, Nakazawa M, Koyama H (2011). Health status and lifestyle factors as predictors of depression in middle-aged and elderly Japanese adults: A seven-year follow-up of the Komo-Ise cohort study. *BMC Psychiatry* 7: 11–20.
- Taylor SJ, Bogdan R (1987). La entrevista en profundidad. In Taylor SJ, Bogdan R (eds). *Introducción a los métodos cualitativos de investigación*. Madrid: Paidós.
- Thomas E, Hay EM, Hajeer A, Silman AJ (1998). Sjögren's syndrome: A community-based study of prevalence and impact. *British Journal of Rheumatology* 37: 1069–76.
- Tincani A, Andreoli L, Cavazzana I, Doria A, Favero M, Fenini MG, Franceschini F, Lojaco A, Nascimbeni G, Santoro A, Semeraro F, Toniati P, Shoenfeld Y (2013). Novel aspects of Sjögren's syndrome in 2012. *BMC Medicine* 11: 93.
- Vasilachis de Gialdino I (2009). Los Fundamentos ontológicos y epistemológicos de la investigación cualitativa. [Ontological and Epistemological Foundations of Qualitative Research]. *Forum Qualitative Sozialforschung/Forum: Qualitative Social Research* 10 (2) Art. 30. Available at <http://nbn-resolving.de/urn:nbn:de:0114-fqs0902307> [November 5, 2015].
- Vitali C, Bombardieri S, Jonsson R, Moutsopoulos HM, Alexander EL, Carsons SE, Daniels TE, Fox PC, Fox RI, Kassan SS, Pillemer SR, Talal N, Weisman MH, European Study Group on Classification Criteria for Sjögren's Syndrome (2002). Classification criteria for Sjögren's syndrome: A revised version of the European criteria proposed by the American–European Consensus Group. *Annals of the Rheumatic Diseases* 61: 554–8.
- Westhoff G, Dörner T, Zink A (2011). Fatigue and depression predict physician visits and work disability in women with primary Sjögren's syndrome: Results from a cohort study. *Rheumatology* 51: 262–9.
- Zakrzewska JM (2013). Multi-dimensionality of chronic pain of the oral cavity and face. *Journal of Headache and Pain* 14: 37.