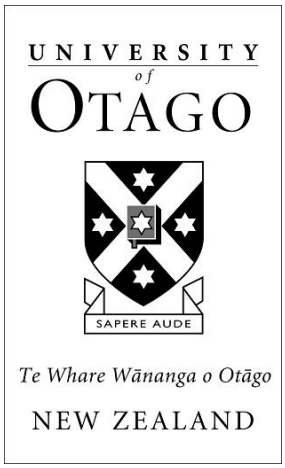
Living with dry mouth – Sjögren's patients' perspectives



Ngo Di Ying Joanna

Research Supervisors
Professor Anita Nolan
Doctor Shelagh Ferguson
Professor W. Murray Thomson

A thesis submitted for the degree of
Doctor of Clinical Dentistry (Special Needs Dentistry)
University of Otago, Dunedin
New Zealand

August 2014

Jesus answered, "*Everyone who drinks this water will be thirsty again, but whoever drinks the water I give them will never thirst. Indeed, the water I give them will become in them a spring of water welling up to eternal life*."

John 4:13-14 (New International Version)

Abstract

Aim

The aim of this qualitative study was to provide clinicians with insight into how dry mouth can impact on the daily lives of Sjögren's Syndrome (SS) patients. SS is an autoimmune exocrinopathy characterised by lymphocytic infiltration of exocrine glands in multiple sites, with dry mouth as a primary presenting symptom. Although quantitative studies have shown the negative impact of both dry mouth and SS on patients' quality of life, no qualitative diary and interview study has been undertaken to examine the specific impact of dry mouth on SS sufferers.

Methods

The revised international classification criteria (AECG) were used to identify participants from patients seen in the oral medicine clinic. After pilot study work to test the approach, the 10 main study participants were recruited. Diary entries and semi-structured interviews were used to explore how dry mouth affects the lives of SS sufferers. Owing to the exploratory nature of the research, thematic content analysis was applied, allowing the themes to arise naturalistically from the data without bias or elicitation.

Results

The main themes included: (1) the journey to diagnosis; (2) disease impact spectrum (of dry mouth amid other symptoms); (3) interactions with healthcare professionals (HCPs); and (4) the positive SS (and dry mouth) coping process.

Conclusion

The findings revealed patients' perspectives on diagnosis, coping with dry mouth and SS, and interaction with HCPs. Dry mouth is not a trivial symptom for SS sufferers; it has considerable impact on their day-to-day lives. HCPs need this understanding in order to be part of the Sjögren's journey.

Acknowledgements

First, I wish to thank my supervisors: Professor Anita Nolan, Dr Shelagh Ferguson, and Professor Murray Thomson, whose brains and effort, have helped me to put this thesis together. Anita, thank you for being the caring clinician who came up with the idea of understanding the dry mouth experience for SS patients. Despite being based in Auckland after the first year, Anita remained supportive. Shelagh, thank you for being adventurous enough to be roped in from the 'dark side' (the marketing department). Her social science background had taught me to conceptualise knowledge from a different perspective. Murray, thank you for being the dry mouth 'guru' on board. Murray's humble and diligent word-for-word editing has definitely honed my scientific writing skills. Collectively, my supervisors have taught me to have humour while enjoying what I do in life.

I am grateful to the New Zealand Dental Association/Ministry of Health (NZDA MOH) Oral Health Research Fund Assessment Panel for funding the study. Moreover, the University of Otago Dental School Department of Oral Diagnostic and Surgical Sciences, the University of Otago Department of Health Sciences, and the Gut Health Network have been generous in the funding for research presentations in the annual Special Care Dentistry Association in Chicago (2014), and the GUT health network meeting in Christchurch (2014).

I wish to express my heartfelt thanks to Jolin (and her family), my best friend, who spent many nights listening to my grumbling and presentation practice sessions. Without her honest admonishment, I would not have recognised the detrimental effects of stress on the QoL of the people around me. Thank you to my family, who provided continuing support from afar. Special thanks to my Mum, who taught me to always be positive no matter what, by smiling even though her mother (my grandmother) has been paralysed and bed-ridden for the past 2 years. Moreover, thank you to my little sister 'Bao' who has always believed in me. Last but not least, thank you God, for

bringing me to New Zealand, the land of 'milk and honey', and for bringing me peace, and trust during this thesis experience.

Table of contents

List of Tables

List of Figures

List of Appendices

List of Abbreviations

SS	Sjögren's Syndrome
AECG	Amererican-European Consensus Group
SLE	Systemic Lupus Erythematosus
RA	Rheumatoid Arthritis
QoL	Quality of life
HRQoL	Health-related quality of life
OHRQoL	Oral health-related quality of life
OHIP	Oral Health Impact Profile
OHIP-14	Shortened version of the Oral Health Impact Profile
SF-36	36-Item Short-Form Health Survey
GOHAI	General Oral Health Assessment Index
XI	The Xerostomia Inventory
SXI	Shortened Xerostomia Inventory
JTD	Journey to diagnosis
HCPs	Healthcare professionals
IHCPs	Interactions with healthcare professionals

DIS	Disease impact spectrum
PCP	The positive SS (and dry mouth) coping process

Chapter 1 Introduction

The body of the thesis has been organised into the literature review, the methodology, the SXI results (quantitative), the results and commentary (quantitative), and the discussion. The literature review has been distributed into four sections on dry mouth, SS, QoL, and qualitative work (on dry mouth and SS). Chapter 2.1 on dry mouth will provide an understanding of its definition, epidemiology, aetiology, impact, and management. The next chapter (2.2) on SS will be an overview of SS, with discussion on its definition, epidemiology, aetiology, pathogenesis, manifestations, diagnosis, and management (with focus on dry mouth). These two chapters will allow a background understanding of the conditions that the participants of the current research are experiencing. The next chapter (2.3) on QoL will provide an understanding of the effects that dry mouth and SS have on QoL. This will include a discussion on HRQoL and OHRQoL; their definitions, measurements, and related health models. Moreover, there will be a review of the use of HRQoL and OHRQoL indices in existing dry mouth and SS studies. The strengths and limitations of using these indices in conceptual models of HRQoL/OHRQoL will be discussed in relation to the Wilson and Cleary model revised by Ferrans et al (2005). This will lead to the need to integrate

other research methods (qualitative) in order to address some of those limitations. The following chapter (2.4) will be on qualitative research, with a focus on dry mouth and SS. The research paradigms informing quantitative and qualitative methods will be established (and compared). There will then be a review on the (limited) existing qualitative research on dry mouth and SS. There will be an identification of the gaps in the current knowledge of the experience of dry mouth in SS patients, thus leading to the research question.

Chapter 3 on methodology will discuss how the research was carried out, including the background work (for Maori consultation, ethical approval, and grant application), participant recruitment (for the pilot and main studies), data collection (both diaries, interviews, and the SXI), and data analysis (that has a main qualitative component and a minor quantitative component).

Results were collated into Chapters 4 and 5. Chapter 4 will report the statistical analysis of the SXI scores. Chapter 5 will discuss the results and commentaries of the findings from the thematic content analysis. It will be divided into four sections according to the themes: (1) the journey to diagnosis; (2) interactions with HCPs; (3) disease impact spectrum; and (4) the positive SS (and dry mouth) coping process.

The results are discussed in Chapter 6, and the strengths and limitations of the current study will be outlined before conclusions are presented with suggestions for future research. There will then be a overview of the thesis in Chapter 7.

Chapter 2 literature review

2.1 Dry mouth

2.1.1 Introduction

This chapter is about dry mouth, the condition on which this research is centred. First of all, dry mouth will be defined, followed by consideration of its epidemiology. There will then be an overview of the leading causes of dry mouth. The impact of dry mouth will briefly be introduced, in order to explain why it is a significant condition. A short review of the management of dry mouth will reveal the current lack of a cure. The implications of dry mouth for quality of life (QoL) will be built on in the succeeding chapters.

2.1.2 Defining dry mouth

There is some confusion about the definition of dry mouth in the literature because the terms "xerostomia" and "salivary gland hypofunction (SGH)" have been used interchangeably in relation to the subjective complaints and objective aspects of dry mouth (Hopcraft and Tan, 2010; Nederfors et al., 1997; Nederfors, 2000). Dry mouth can manifest as xerostomia or salivary gland hypofunction (Nederfors et al., 1997; Thomson, 2005). Xerostomia is the subjective symptom of dry mouth (this could be a result of a qualitative change in saliva). It is a sensation that is assessed only by directly questioning the individual (Fox et al., 1987). On the other hand, the objective sign of dry mouth is salivary gland hypofunction, in which the quantity of saliva produced is lowered. It can be determined by sialometry (Navazesh, 1993).

2.1.3 The epidemiology of dry mouth

Dry mouth is an important condition that has been shown to affect the quality of life of sufferers, and especially that of the older population (Gerdin et al., 2005; Ikebe et al., 2007; Locker, 2003b). Studies have also shown that it also affects younger people (Thomson et al., 2006b; Thomson et al., 2006c). A condition is an important health problem when it affects a significant number of the population. A look at the prevalence of dry mouth will show that dry mouth is not a trivial condition. Determining the epidemiology of dry mouth is complex due to: (1) the poor definition of dry mouth; (2) the different methods to measure dry mouth; (3) the samples used in most studies tend to be institutionalised older patients, who are often on multiple medications (which can confound its investigation). Epidemiological research is not the main focus of this study, but these issues are discussed for a complete overview of dry mouth as a condition.

2.1.3.1 Measuring dry mouth

In this section, the different methods to assess xerostomia and salivary gland hypofunction are described. Xerostomia is subjective, and therefore questions are used to assess xerostomia. These range from single-item methods to multi-item summated rating scales. There are advantages and disadvantages to each method. An understanding of the exisiting methods will provide background information on how the current qualitative research can complement the exisiting methods to assess

xerostomia. SGH is an objective sign measured using techniques to quantify saliva flow rate. In the diagnosis of Sjögren's syndrome (SS), both objective and subjective assessments are made (Vitali et al., 2002). Next, the prevalence of dry mouth based on different methods and sample populations will be discussed. This provides an idea on the significance of dry mouth as a condition in populations.

2.1.3.1.1 Measuring Xerostomia

The subjective symptom of dry mouth, xerostomia, can be measured by a (1) single-item question, or (2) multi-item approaches, including (a) batteries of items, or (b) summated rating scales.

(1) Single-item approaches

A single question (global item) is a statement which requires the respondent to integrate their experiences, perceptions, and behaviours into an overall summary judgement. An example of a single question is "Does your mouth feel distinctly dry?" (Österberg et al., 1984). There are limitations to this method, because a single-item inevitably categorises a patient as xerostomic or non-xerostomic according to an arbitrary cut-off point (Hopcraft and Tan, 2010). Chronic dry mouth is a condition that can vary with time, and such single questions will be likely to cause the respondent to wonder whether the question refers to "now", or "usually?" (Thomson, 2014). Different questions can yield quite different prevalence rates within the same sample. For example, the prevalence of dry mouth in a Japanese elderly population was reported at 9% when asking about dry mouth in relation to eating, and a different prevalence of 38% was obtained when questioning the same sample about dry mouth in relation to waking (Ikebe et al., 2007).

Fox et al. developed four questions (two related to difficulties in swallowing, one related to eating, and one related to amount of saliva) in an attempt to gain a more comprehensive assessment of xerostomia (Fox et al., 1987). However, there is concern that each of the four questions may yield a different prevalence estimate. No guidance was given by the authors as to whether the four items can be used in a battery or scale (see below). In another attempt to include more aspects of xerostomia, Närhi employed a series of 16 items ranging from difficulty in speaking to oral burning and itching sensations (Närhi, 1994). Despite the expansion from a single-item questionnaire to a

series of items, the responses were still limited to ‘yes’ and ‘no’ answers that do not capture the severity and changes in time associated with xerostomia. Such limitations can be overcome with qualitative diary methods, although these are not able to be used in determining the condition’s prevalence (described in a later chapter).

(2) Multi-item approaches

The multi-item approaches include: (a) batteries of items; and (b) summated rating scales. Each will next be discussed accordingly.

(a) Batteries of items

Batteries are a list of items with a ‘yes’/‘no’ response format. During analysis, the total number of positive responses is used as an index score. For example, Locker used a list of seven questions and a simple ‘yes’/ ‘no’ response format to group nursing home residents into three categories: no xerostomia (0 positive responses); mild xerostomia (1 to 2); or marked xerostomia (3 to 7) (Locker, 2003b). The first question was “During the past four weeks, have you had a dry mouth or tongue during the daytime?” The next six questions also asked about the experiences in the past month with regard to dry mouth-related problems (difficulty talking, difficulty chewing, difficulty swallowing) or behaviours undertaken to alleviate dry mouth (needing to drink water during the daytime, needing to drink water with meals, needing to chew gum to relieve dryness). This battery was found to have acceptable internal consistency reliability. However, no comparison with a single item xerostomia item was made in order to judge the validity of the battery.

Batteries of items can be useful for exploring the determinants of xerostomia. However, they may not relate to the experience of dry mouth with just a ‘yes’/ ‘no’ response. This will be further elaborated in a later chapter on the effects of dry mouth and SS on quality of life.

In a variation of the battery-type approach, Pai et al (2001) made a modification in the type of response scored. Instead of a ‘yes’/‘no’ response, a visual analogue scale (VAS) response was employed. In a VAS response, the respondent places a mark on a line to indicate the point representing his/her position between two extremes. This spectrum of responses, instead of an absolute response, possibly provides a more

naunced assessment of xerostomia and allows for the monitoring of symptom alterations. That eight-item VAS xerostomia questionnaire was employed to improve the diagnosis and monitoring of SGH and its changes over time in relation to xerostomia (Pai et al., 2001). However, xerostomia and salivary gland hypofunction can exist independent of each other (as described in a later section). The problem with the VAS is that the items were put together from clinical experience or the literature in an arbitrary manner. The less relevant items included may result in error and variance. This issue is addressed by summated rating scales (Thomson, 2014).

(b) Summated rating scales

A summated rating scale is a multi-item scale that is a more refined development of the batteries of items, in which the items have been shown to psyhchometrically correlate with the underlying construct (in this case, xerostomia). The items in the summated rating scale are generated through a series of steps: conceptual development of underlying theory; development of an item pool, which encompasses a list of all issues relevant to xerostomia (from literature, clinical experience, qualitative interviews with sufferers); item pool reduction and psychometric testing (to determine whether the items do actually relate to xerostomia); and then field testing and validation. The theory behind using such a scale to measure dry mouth is to be able to place respondents on a continuum that represents the range of experience of the condition.

This allows for subtle differences in dry mouth to be explored (Thomson, 2014).

Least possible Most possible

2.1.3.1.2 The Xerostomia Inventory

The Xerostomia Inventory (XI), a multi-item continuous scale instrument is an example of a summated rating scale. It was developed as part of a study of the question of whether the use of xerogenic drugs among older people is associated with greater caries experience. The XI contains 11 items (derived from both quantitative and qualitative methods) aimed to capture the broad experience of xerostomia. The items were generated in a few stages: (1) a literature search revealed single items that have been previously used; (2) these single items were used to develop a framework for semi-structured interviews that were undertaken with four chronic sufferers of

xerostomia; (3) content analysis was used to identify dominant themes that were developed into 19 potential XI items that reflected many manifestations of the xerostomia experience grounded in the experiences of xerostomia sufferers; and (4) these 19 items were then field tested and reduced to 11 items that represented a mix of experiential and behavioural aspects of the condition (Thomson et al., 1999b). The five possible responses for each item range from 'never' to 'very often', capturing the severity of the condition, and avoiding the dichotomising of respondents into 'xerostomic' or 'non-xerostomic' categories. Each response is given a numerical value ranging from 1 to 5 according to its severity, and the XI score is calculated as the sum of the 11 items. The XI also allows changes in severity to be monitored over time. So far, the XI is the most comprehensive measure that addresses the individual awareness and the consequences of xerostomia. The XI was validated in a prospective cohort study comparing patients about to undergo radiotherapy (for head and neck cancer) to a convenience sample of middle-aged and older individuals (Thomson and Williams, 2000). Further testing of the responsiveness of the XI indicated that a change in score of 6 or more to be clinically meaningful (Thomson, 2007). The XI has been translated into several languages, including Dutch (Bots et al., 2005), Mandarin (He et al., 2013), Portuguese (da Mata et al., 2012), Turkish (Eltas et al., 2012), and Spanish (Martín-Piedra et al., 2011). The XI was subsequently shortened to a 5-item continuous scale (Appendix 1) and tested using samples from Australia, The Netherlands, Japan, and New Zealand. The conclusion of that work was that the shortened XI (SXI) is valid for measuring xerostomia symptoms in clinical and epidemiological research (Thomson et al., 2011).

It is important to note that none of these multi-item methods collects information on psychological traits or emotions such as anxiety or fatigue. Researchers seeking to explore these aspects should seek the appropriate instrument such as qualitative interviews or diary methods, and use it concurrently with the xerostomia measure of choice. The SXI will be used in this research as a description of symptom manifestation (xerostomia) for each participant. This will be described in a later chapter.

2.1.3.1.3 The measurement of SGH

SGH is an objective sign that can be measured clinically. The normal daily production

of saliva is between 0.5 and 1.5 litres. The three major salivary glands (parotid, submandibular and sublingual) contribute 90% of total salivary flow. The minor salivary glands (distributed all over the mouth) contribute to the remaining 10% of salivary flow (Navazesh and Kumar, 2008). The secretions of the parotid glands are mainly serous in nature, contributing mostly to stimulated saliva. The secretions of the submandibular glands are mixed (serous and mucous) in nature, while the secretions of the sublingual glands are mostly mucous. The submandibular and sublingual glands are the main contributors to unstimulated saliva. The rest of it is produced by mucin from the numerous minor salivary glands. Unstimulated saliva functions to lubricate and protect the oral mucosa (Jensen and Vissink, 2014). SGH can be estimated by measuring stimulated or unstimulated salivary flow. The steps are: (1) to measure salivary flow; and then (2) determine whether the patient has SGH by comparing his/her flow to a threshold value. Unstimulated saliva is the resting basal flow that is present in the mouth for the majority of each day. It coats and protects the mucosa and oral tissues. Stimulated saliva, on the other hand, is in the mouth for about 2 hours each day for alimentary functions (Sreebny, 2000). Salivary stimulation is commonly achieved using citric acid or chewing of a piece of paraffin. Measuring unstimulated salivary flow is more representative of the *in vivo* situation. Salivary flow can be evaluated by collecting saliva from individual glands or by assessing whole salivary flow (total secretions of major and minor glands). Whole salivary flow (usually using the spit technique) is a more practical method to collect saliva (Thomson, 2005). Because salivary flow varies throughout the day, the objective of salivary flow measurement is patient standardisation. A base reference recording to compare within patient is ideal (Humphrey and Williamson, 2001). In the Amererican-European Consensus Group (AECG) revised international classification criteria for SS (Appendix 2.) (this will be further discussed in the next chapter on SS), a collection time of 15 minutes for unstimulated whole saliva is required (Vitali et al., 2002). This extended collection time in longer than the usual 5 minutes and may be due to the fact that patients with SS can have such a low salivary flow that such a long collection time is needed to produce a measureable amount of saliva. An unstimulated whole salivary flow rate of <0.1ml/min is considered to be less than normal, whereas a stimulated flow rate of <0.5ml/min is considered abnormal (Sreebny and Schwartz, 1997).

2.1.3.2 The prevalence of dry mouth

The prevalence of dry mouth is not easy to estimate due to the factors described previously: (1) the poor definition of dry mouth because of interchangeable use of xerostomia and SGH to describe subjective and objective aspects of dry mouth; (2) the different methods of measuring both xerostomia and SGH; and (3) convenience samples of institutionalised older people being used for epidemiology studies. In addition, some studies assume that everyone who experiences xerostomia has SGH, and vice versa. The evidence suggests that the two are not necessarily concurrent.

2.1.3.2.1 Varying estimates

The variability in estimate of the prevalence of xerostomia due to different questionnaire formats and sample selection bias has been addressed in a systematic review by Orellana et al (2006). Based on mainly elderly Scandinavian population samples, the reported prevalence of xerostomia has ranged from 0.9% to 64.8%. The variation may be explained as a consequence of differences in diagnostic process (Orellana et al., 2006). The review criticised the lack of consensus in the literature regarding the definition of xerostomia, and showed how different types of single-item questions used may yield different prevalence estimates in the same population. Also, most of the studies were based on an older population. Suggestions were made for future research to investigate xerostomia in younger populations. Similarly, an earlier epidemiology paper by Thomson (2005) demonstrated the varying estimates of xerostomia. In this study, the reported estimates of xerostomia prevalence from epidemiological studies of representative samples of non-institutionalised older populations ranged from 12% to 39% (Thomson, 2005).

2.1.3.2.2 Ageing and dry mouth

Even though it has been generally accepted that ageing has no significant impact on salivary flow rates, the prevalence of xerostomia appears to be greater in older people. This may be due to the polypharmacy experienced with a concurrent increase in age-related medical conditions (Turner and Ship, 2007). The misconception that xerostomia affects only older people has been challenged by findings from the Dunedin Multidisciplinary Health and Development Study, a longitudinal study of health and behaviour in a complete birth cohort, where xerostomia was reported in

10% of the 972 32-year-old participants (Thomson et al., 2006b). There is still much to know about the natural history of dry mouth, from the pre-clinical stage to its clinical manifestation, and then to persistence, remission, relapse or resolution (Thomson, 2005).

2.1.3.2.3 Sex and dry mouth

There appears to be a sex difference in xerostomia, whereby the reported prevalence of xerostomia is lower in men (10 to 26%) than in women (10 to 33%) (Hopcraft and Tan, 2010). However, there is no apparent gender difference in younger adults (Thomson et al., 2006b). This could mean that there may be some changes related to menopause, resulting in an increased experience of dry mouth in older women (Agha‐Hosseini et al., 2012).

2.1.3.2.4 The relationship between xerostomia and SGH

The relationship between xerostomia and SGH is complex. Logically, xerostomia can occur due to a reduction in salivary flow in SGH, but both the subjective (xerostomia) and objective (SGH) components are known to occur independently of each other (Locker, 1995). The few studies conducted on both xerostomia and SGH within the same population show that the two are largely separate conditions. In a systemtic review, the prevalence of xerostomia prevalence ranged from 8 to 42%, while the prevalence of SGH prevalence ranged from 12 to 47%. The prevalence of both conditions existing together is only about 2 to 6% (Hopcraft and Tan, 2010). Similarly, in a longitudinal study of a population-based sample of 700 older South Australians, the prevalence of xerostomia was 20.5% and the prevalence of SGH was 22.1%, but only 5.7% of participants had both conditions (Thomson et al., 1999a).

In general, xerostomia and SGH affects a significant proportion of the population, especially older people. As the global population continues to age (Lutz et al., 2008), dry mouth will become an increasingly prominent problem in the future.

2.1.4 The aetiology of dry mouth

The common causes of dry mouth include xerogenic drugs, radiotherapy to the head and neck for cancer, and systemic diseases such as SS and various associated connective tissue disorders (Guggenheimer, 2003).

Xerogenic drugs are the most common cause of dry mouth investigated in epidemiological studies, and it is an area which has been accompanied by the greatest number of avoidable mistakes (Thomson, 2014). The role of medications in the occurrence of dry mouth is very relevant to this research because SS patients are often on multiple (possibly xerogenic) medications for the autoimmune effects of the disease. A look at dry mouth-inducing medications reveals an inexhaustable list. These drugs usually have anticholinergic or sympathomimetic actions that affect the neural control of salivary glands, a cytotoxic effect on salivary glands, or a diuretic effect that depletes fluids (Scully and Felix, 2005a). Sreebny and Schwartz identifed more than 400 drugs in 42 drug categories which they asserted capable of inducing xerostomia or SGH (Sreebny and Schwartz, 1997). The utility of such broad lists may be limited because most of the categories of medication included are based on case reports and clinicians' opinions instead of the findings of clinical or epidemiological studies. There is no indication of whether the medications listed cause xerostomia or SGH, and there is a lack of information on the xerogenicity of each specific medication, whether alone or in combination. Analysing the xerogenicity of medications is complicated by the difficulties of capturing information on medication exposure and choosing the analytical approach to be taken. Furthermore, polypharmacy is common in older people, and dry mouth may be a side-effect of the underlying medical conditions being treated (Thomson, 2005).

Irradiation for malignant tumours in the head and neck region can cause dry mouth by direct damage to the salivary glands. Systemic diseases such as diabetes mellitus and chronic renal failure can result in dehydration and hence xerostomia. Systemic diseases that affect the salivary glands can cause salivary dysfunction, resulting in dry mouth. These include sarcoidosis, hepatitis C virus infection, and SS. In SS, dry mouth is a prominent and consistent oral finding (Fox, 2007). Dry mouth in SS patients is a focus of the current research, and it will be elaborated on in the next chapter. The common aetiology of dry mouth is summarised in Table 1 (Guggenheimer 2003).

Table 1: Causes of dry mouth (after Guggenheimer 2003)

Medications	Antihistamine, Antihypertensives, Antidepressants, Anticholinergics,

	Antipsychotics, Sedatives, Analgesics, Muscle relaxants, Diuretics, Anticonvulsants, etc.
Sjögren's Syndrome	Primary and Secondary
Connective Tissue Diseases	Rheumatoid Arthritis, Systemic Lupus Erythematosus, Systemic Sclerosis, Mixed connective tissue disease
Other Conditions	Radiotherapy, Chronic Active Hepatitis, HIV, Graft vs Host disease, Renal Dialysis, Anxiety, Depression, Diabetes Type 1 and 2

In addition, there are physiological causes of dry mouth, such as anxiety (due to sympathetic activity), mouthbreathing, and (rarely) salivary gland agenesis (Scully and Felix, 2005a). Dry mouth in a patient can be due to more than one factor. For example, in a patient with a chronic systemic disease such as SS, depression can be a commonly associated issue (Valtýsdóttir et al., 2000; Westhoff et al., 2012). In such a scenario, dry mouth in the patient may be due to the salivary gland dysfunction, the anti-depressant-induced dry mouth, or the depression itself (Anttila et al., 1998).

2.1.5 The impact of dry mouth

With an understanding of the prevalence and aetiology of dry mouth, this next section will discuss the effects of dry mouth. These include physical, emotional, and social impacts. This provides a background understanding for the focus of this research – the experience of dry mouth.

The function of saliva needs to be understood to explain the effects of chronic dry mouth. In healthy adults, up to 1.5 litres of saliva is produced daily. Salivary function can be oraganised into five major categories that serve to maintain oral health and create ecologic balance: (1) lubrication and protection; (2) buffering action and clearance; (3) maintenance of tooth integrity; (4) antibacterial activity; and (5) taste and digestion. Unstimulated saliva keeps the oral mucosa moist and maintains oral health. Stimulated saliva that is produced in response to sensory stimuli (together with

mechanical chewing) aids in the digestion process. Saliva also facilitates speech, cleanses food residues in the mouth, enhances taste, and neutralises potentially damaging food acids (Humphrey and Williamson, 2001). The impact of dry mouth on oral health is drastic. These include infections, pain, nocturnal disturbances, denture wearing difficulties, difficulty in eating, speech problems, and more. The impact of chronic dry mouth will be highlighted later on, using the clinical manifestations of SS as an example.

The impact of dry mouth extends beyond the oral cavity into the daily lives of sufferers. One way of assessing this is by examining the effect of dry mouth on the oral-health-related quality of life (OHRQoL) of sufferers (Locker and Allen, 2007). A study of 225 institutionalised older people in Toronto (Locker, 2003b) that used two different OHRQoL scales the General Oral Health Assessment Index, or GOHAI; (Atchison and Dolan, 1990), and the short-form Oral Health Impact Profile, or OHIP-14 (Slade, 1997). It found xerostomia be an important influence on the well-being and QoL of the population. In a Swedish older population study (Gerdin et al., 2005), both xerostomia and SGH were found to be significantly associated with OHRQoL. As part of the Dunedin study, xerostomia was found to be strongly and independently associated with poorer OHRQoL among 32-year-olds. This finding suggested that xerostomia is not a trivial condition for anyone, whether relatively healthy young adults or institutionalised older adults (Thomson et al., 2006a). More investigation aimed at improving the understanding of the impact of dry mouth on daily lives is essential. Qualitative research is one approach to improve knowledge in this area.

2.1.6 The management of dry mouth

Dry mouth is not an easy condition to manage. The management of dry mouth, and its consequences (as previously described) will be discussed in this section. This will provide background knowledge of the dry mouth management which the participants in this research will have undergone.

In general, the goals in the management of dry mouth are to manage underlying systemic condition(s), alleviate symptoms, and institute preventive measures. This may involve increasing the amount of exisiting saliva or replacing lost secretions in order to control the development of caries and treat specific oral infections such as candidiasis (Tsubota, 2012).

2.1.6.1 Management of underlying systemic condition(s)

In order to manage dry mouth, the underlying cause needs to be understood (and rectified if possible). For example, patients taking xerogenic medications may have the drugs changed for an alternative if possible. However, this may be impractical in view of the multiple group of medications that induce dry mouth, and the polypharmacy that is especially common in the older population. Should the underlying cause of dry mouth be a systemic disorder such as diabetes mellitus, treatment should be aimed at the systemic disease. In SS, there is no cure for the condition that often results in damage to the salivary glands. The management of dry mouth in SS will be a life-long journey of palliation.

2.1.6.2 Managing the symptoms of dry mouth

Synthetic saliva substitutes (such as dry mouth gel or sprays) contain carboxymethylcellulose, a mucopolysaccharide, glycerate polymer base or mucins, which can provide temporary mucosa wetting. More recently, 1% malic acid spray has been explored as a topical sialogogue to improve xerostomia in patients who are on antidepressants and anti-hypertensive medications (Martín-Piedra et al., 2011). Sugar-free candies and chewing gums that contain xylitol are intended to stimulate salivary flow. Patients with dry mouth can also make adjustments to their diet to avoid dry or acidic food, and can sip water with their meals. Patient education is also important so that they can avoid factors that may increase oral dryness (such as caffeine or alcohol), and to keep the mouth moist (sip on water).

2.1.6.3 Managing the consequences of dry mouth

There are measures to prevent the consequences of dry mouth. These include a non-cariogenic diet, a high level of oral hygiene, and the regular use of topical fluoride agents (toothpaste, gels, rinses, varnishes), and amorphous calcium phosphate. Dental examinations every 4-6 months with accompanying radiographs are also recommended. Sialologues that are cholinergic drugs (such as pilocarpine) can be prescribed to promote saliva production. This is especially so in the management of SS patients (Felix et al., 2012; Napeñas et al., 2009).

There is no cure for dry mouth; rather, there is only palliative management to alleviate the symptoms. Although acupuncture has been noted to be effective in the treatment of

dry mouth after head and neck radiotherapy (Johnstone et al., 2001), further research needs to be done in this area. The measures to manage dry mouth are summarised in Table 2 (Napeñas et al., 2009).

Table 2: Overview of the management of dry mouth (after Napeñas et al., 2009)

Manage underlying systemic conditions
Multidisciplinary management with other healthcare providers
Management of symptoms
Diet and habit modifications
Frequent and regular sips of water
Avoidance of dry, hard, sticky, acidic foods
Avoidance of excess caffeine and alcohol
Salivary substitutes and lubricants
Artificial saliva, gels, rinses, sprays, bedside humidifier (sleeping hours)
Sialogogues
Pilocarpine 5-10mg orally TDS
Cevimeline 30mg orally TDS
Acupuncture

Management of the consequences of dry mouth

Prevention

- Increased frequency of dental examination
- Topical fluoride application

Treatment of oral conditions

- Dental caries - Restoration, topical fluoride
- Oral candidiasis - Chlorhexidine rinse and antifungal medication
- Poor-fitting prosthesis - Denture adhesives

In Table 2, the management is divided into the underlying cause of dry mouth, the symptoms of dry mouth (diet and habit modifications, salivary substitutes, sialogogues, acupuncture) and the underlying consequences of dry mouth (prevention and treatment). The items in the Table act as a general guideline, because there are different brands and medications available in different countries. Also, each individual tailors the management of dry mouth to suit their lifestyle and preferences. Many of these strategies to alleviate the symptoms of dry mouth may only have a short-term effect. This will be further discussed in the Results chapter.

2.1.7 Conclusion

In conclusion, dry mouth is a condition that is complex and relatively prevalent. Much research has been done to quantify and understand the cause of dry mouth. Its impact on sufferers is pertinent in view of its negative effect on OHRQoL and the lack of cure. Dry mouth is the one of the most common findings in the diagnosis of SS. The following chapter will describe SS in relation to dry mouth.

2.2 Sjögren's Syndrome (SS)

2.2.1 Introduction

This chapter is an overview of SS, beginning with its definition, followed by its epidemiology. Next, the current understanding of the aetiology and pathogenesis of SS will then be summarised. There will then be consideration of the clinical perspective of the: (1) manifestations, (2) diagnosis, and (3) management of SS (with focus on dry mouth). These will allow a background understanding of the disease the participants of this research are experiencing. The impact of SS on QoL will follow in the next chapter.

2.2.2 The definition of SS

In 1933, the Swedish ophthalmologist Henrik Sjögren reported clinical and histological findings on the triad of dry eyes (keratoconjunctivitis sicca), dry mouth and rheumatoid arthritis. This triad became known as SS. SS has been defined as a chronic autoimmune disorder characterised by lymphocytic infiltration of exocrine glands in multiple sites (Al-Hashimi, 2005; Fox, 2005; von Bultzingslowen et al., 2007). The main exocrine glands that can be affected are the lacrimal and salivary glands, which end up with reduced secretory functions (Kassan and Moutsopoulos, 2004). Although dry eyes and dry mouth are classical symptoms of SS, the exocrine involvement may extend to the skin, vagina, respiratory system, or gastrointestinal systems (Al-Hashimi, 2005) (to be discussed in the later section on the clinical manifestations of SS). Primary SS, or "sicca syndrome", is the association of dry mouth with dry eyes only. By contrast in secondary SS, dry mouth and dry eyes occur in association with other autoimmune disorders such as rheumatoid arthritis (RA), systemic lupus erythematosus (SLE), or progressive systemic sclerosis (SSc) (Al-Hashimi, 2001; 2005; Fox, 2005; Scully, 2010).

2.2.3 The prevalence of SS

The few epidemiological studies of SS have yielded heterogeneous findings due to differences in diagnostic criteria and study design (Binard et al., 2007). SS has been found to be most common in middle-aged females. The American-European Consensus Group (AECG) (Appendix 2.) Revised International Classification criteria for SS (Vitali et al., 2002) is the most accepted diagnostic criteria in clinical and research fields (to be discussed in conjunction with the diagnosis of SS). In a review of SS epidemiology by Mavragani and Moutsopoulos (2010), the reported prevalence of SS ranged from 0.1% to 4.8%. The observed variability was suggested to reflect the different diagnostic criteria applied (whether the European Criteria, the San Diego Criteria, the Copenhagen Criteria, or the AECG), the diverse geographic study origin, as well as the different sampling methods, and sex distribution (Mavragani and Moutsopoulos, 2010). The estimated prevalence of SS from available data was suggested to be 0.5% to 1% in the general population of Europe (Binard et al., 2007). The findings are similar to those from population based studies in Scandinavia (Theander and Wollheim, 2012), Turkey (Kabasakal et al., 2006), and the UK

(Bowman et al., 2004).

The effect of different diagnostic criteria used in the same sample on prevalence estimates can be observed in a study on a Chinese community population of 2066 adults (Zhang et al., 1995). In this study, the prevalence of SS was reported to be 0.77% according to the Copenhagen criteria (Manthorpe et al., 1985), and 0.33% by the modified San Diego criteria. This is because the latter is a more stringent set of criteria, and so fewer cases were identified.

An interesting example of geographic origin affecting the contribution of genetic and environmental factors to SS is seen in a Japanese study of 1008 Nagasaki atomic bomb survivors (Hida et al., 2008). In this study, the prevalence of SS was found to be 2.3% (applying the AECG criteria). However, SS prevalence in these atomic bomb survivors was found not to be associated with radiation dosage. Instead, an observed association between radiation dose and hyposalivation supported the possibility that radiation exposure damaged salivary gland function.

Similarly, in a systematic review by Binard et al, the estimated prevalence of SS varied widely from 0.1% to 3.6% (Binard et al., 2007). Nevertheless, 14 of the 17 prevalence estimates were lower than 2%, and 12 were lower than 1%. The studies conducted in older populations tended to result in higher prevalence estimated. This could be due to the fact that dry mouth is a more common symptom in the older population, as discussed in the previous chapter (the contribution of dry mouth to the diagnosis of SS will be considered later in this chapter).

SS is found in all age groups but more commonly in middle-aged women (Garcia-Carrasco et al., 2002), and rarely in children and adolescents (Sardenberg et al., 2010). The female preponderance is large, with the female-to-male ratio noted to be about 9:1 (Fox, 2005). In New Zealand, this could mean that there are 20,000 to 40,000 sufferers, most of whom are middle-aged women. There could be more, though, because the broad spectrum of clinical manifestations makes SS an under-diagnosed condition (Al-Hashimi, 2005), with many of the symptoms presenting as deceptively non-specific (such as, fatigue and joint pain) (Al-Hashimi, 2001).

2.2.4 The aetiology and pathogenesis of SS

The aetiology of SS is multifactorial, involving a complex interplay of genetic, environmental, hormonal, and immunological factors. This is depicted in Figure 1.

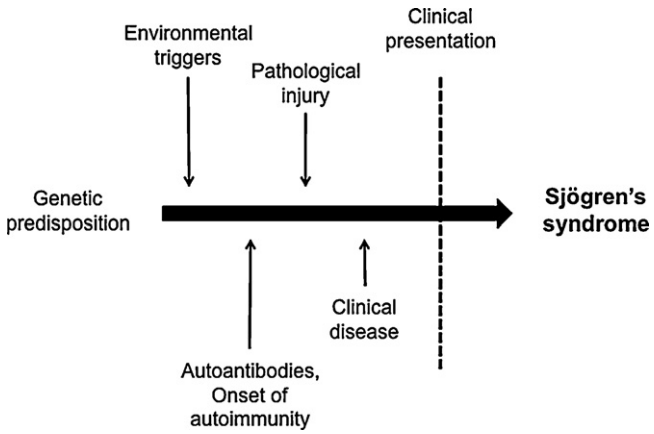


Figure 1. Proposed aetiopathogenic events prior to diagnosis of SS (Jonsson and Brun, 2010)

Each of these aetiological factors will be discussed in turn.

2.2.4.1 Genetic predisposition

A genetic predisposition was implied from observations of a higher incidence of SS among patients with a family history of the disease, with a link between human leukocyte antigen (HLA) class I and class II and the development of SS (Loiseau et al., 2001; Reveille et al., 1984). This is supported by the observation of the co-occurrence of SS in twins (Bolstad et al., 2000). More recently, a large-scale association study of 395 SS patients observed the importance for SS of genes (HLA, IRF5-TNPO3, DDX6-CXCR5, and TNIP1) that are involved in both the innate and adaptive immunity (Lessard et al., 2013).

2.2.4.2 Environmental triggers

A viral initiating factor has been suggested, based on animal studies which found that mice infected with cytomegalovirus infection developed SS-like signs (Fleck et al.,

1998). Other viruses implicated in the development of SS are the Epstein-Barr virus, the hepatitis C virus (HCV), and the human T-lymphotropic virus type-1 (Gessain et al., 1985; Haddad et al., 1992; Saito et al., 1989; Terada et al., 1994). It is hypothesised that viruses can initiate autoantibody production because the amino-acid sequence and structural similarities between foreign and self-molecules. An immune response directed against the virus can elicit an autoimmune response, resulting in cell and tissue destruction (Bayetto and Logan, 2010). This is further supported by how HCV and Human Immunodeficiency Virus (HIV) infections can induce SS-like symptoms, such as dry mouth (Scully and Felix, 2005b).

2.2.4.3 Hormonal factors

The high prevalence of SS in middle-aged women suggests an association between hormonal change and the disease. The immune regulatory effect by sex hormones have been suggested to play a part in the development of SS, with oestrogen and prolactin functioning as immune response stimulators, and androgen acting as an immune response inhibitor. Oestrogen has a role in lymphocyte proliferation, antibody production, and cell apoptosis. Prolactin stimulates T-cell proliferation, interleukin-2 receptor expression, and antibody production (McMurray, 2001). There is evidence that higher levels of oestrogen may be involved with the development and proliferation of anti-Ro/SSA and anti-LA/SSB autoantibodies that are key markers for SS diagnosis. A possible explanation of the higher incidence of SS in middle-aged women could be that oestrogen levels increase, and androgen levels decrease during menopause (Taiym et al., 2004). However, it is also important to take into consideration that the diagnosis for SS is often delayed by years because of the vague symptoms. By then, the women who are diagnosed are usually in middle age. More research on this is required.

2.2.4.4 Autoimmunity in SS

SS is characterised by the mononuclear cell infiltration of exocrine tissues, and the presence of autoantibodies against ribonucleoprotein particles SS-A/Ro and SS-B/La. The salivary and lacrimal glands are the main targets of T-cell mediated chronic inflammation, resulting in glandular atrophy and dysfunction. This presents as ‘sicca syndrome’. Other organs are also affected, resulting in systemic manifestations (that will be described later under the clinical features of SS). The pathogenesis of SS is a result of the autoimmune process that is not completely understood. The context of dry

mouth in SS will be used to illustrate the involvement of autoantibodies in the SS pathogenesis. This will be followed by a description of the general flow of events.

Anti-Ro/SSA and anti-La/SSB are the most common autoantibodies found in SS patients. It is unknown whether these important markers have a direct pathological effect on SS. However, these autoantibodies have been found to be: (1) in higher concentrations in SS patients; (2) detected before symptom onset; and (3) associated with specific signs and symptoms such as dry mouth, dry eyes, hypergammaglobulinemia, lymphopenia, and severe salivary gland disease (Halse et al., 2000; Jonsson et al., 2007; Jonsson et al., 2013).

The interaction of neural and immune factors has been found to affect the secretory responses of glands. This immune-mediated neural pathogenesis has been suggested by studies that describe the presence of antimuscarinic acetylcholine receptor-3 (m3AChR) autoantibodies in patients with SS (Deák et al., 2012; Waterman et al., 2000). These autoantibodies influence the normal function of muscarinic M3 receptors that regulate secretion in the lacrimal and salivary glands (Fox and Michelson, 2000). The knowledge is applicable to the use of muscarinic agonists, such as pilocarpine in the management of dry mouth in SS.

The pathogenesis of dry mouth (and dry eyes) in SS remains unclear. It is hypothesised that SS begins in the salivary (or lacrimal) glands, initiated and/or caused by an exogenous agent, probably some type of retrovirus; after initiation, a straightforward sequence of events follows. First, salivary gland epithelial cells are disrupted, then T lymphocytes migrate to (and are activated in) the glands. Following this, hyper-reactive B cells get the help they need and start to produce SS (anti-Ro/SSA and anti-La/SSB) and rheumatoid factor (RF) autoantibodies. B cells play a central role in the pathogenesis of SS. This is reflected in the observation of B cell infiltration of salivary glandular and extra-salivary tissue in SS, and the association of SS with B-cell lymphomas (O'Neill and Scully, 2013). Abnormal B cell distribution has also been suggested to be linked to the clinical evolution of primary SS to B cell lymphoma (Tobón et al., 2010). Next, cytokines and interferon pathways are activated. Local and systemic cytokine dysregulation occurs, with an increase in pro-inflammatory

cytokines such as interferon-gamma, tumour necrosis factor-alpha, interleukin-6, and B cell-activating factors. Finally, these processes lead to the structural destruction and loss of acinar cells (Konttinen and Kasna-Ronkainen, 2002). As the condition progresses, fibrosis replaces normal acinar cells in the glands, gradually rendering them non-functional.

Owing to the inflammatory and immune processes involved in the pathogenesis of SS, treatment has been focused on immunomodulatory/immunosuppressive therapies. These will be discussed later, in the section on the management of SS.

2.2.5 The clinical features of SS, with a focus on dry mouth.

SS is a chronic autoimmune disorder characterised by lymphocytic infiltration of exocrine (especially the lacrimal and salivary) glands. This can lead to structural destruction and ultimately, loss of gland function. Although dry eyes and dry mouth are the classical symptoms of SS, any organ or mucosal surface may be involved during the disease course. Therefore, patients may experience a wide spectrum of systemic clinical manifestations.

2.2.5.1 Dry eyes

Dry eyes are a prominent ocular manifestation of SS. The compromised lacrimal gland function causes diminished tear production. This may result in damage to the corneal conjunctiva epithelium (Kassan and Moutsopoulos, 2004). Accordingly, dry eyes may manifest as a gritty sensation, soreness, itchiness, eye fatigue, photosensitivity, ocular discharge, and intolerance to contact lenses (Al-Hashimi et al., 2001).

2.2.5.2 Dry mouth

Dry mouth is a common oral manifestation in SS patients. The effects of chronic dry mouth on SS patients are related to the functions of saliva. As outlined in the previous chapter, dry mouth can manifest as a xerostomia (a subjective sign), or SGH (an objective symptom). The manifestations can be due to the dry mouth itself, or to its consequences. In the previous chapter on dry mouth, the functions of saliva were discussed in relation to the impact of dry mouth. In summary, saliva plays an essential role in the numerous functions of the mouth, such as protecting the oral cavity (lubrication, buffering, and antibacterial activity), taste, digestion, and speech. The

impact of dry mouth (both xerostomia and SGH) on the SS patient is because the many important functions of saliva are compromised.

2.2.5.2.1 Xerostomia in SS individuals

As described earlier, the subjective symptom of dry mouth, xerostomia, can be measured by different questions. These questions can assess a range of aspects of xerostomia. For example, the lack of saliva may cause SS individuals to experience significant difficulties in eating some foods (Hay et al., 2001), especially without fluids. In an American study of 169 SS patients, 93.5% reported xerostomia, 55.6% reported a sore mouth, 41.4% reported a change in taste, 25.4 % reported to have difficulty in swallowing, 16.6% reported to have difficulty in chewing, and 13% reported a change in smell (Al-Hashimi et al., 2001). Similarly, in a review by Soto-Rojas and Kraus (2002), the subjective symptoms of oral dryness common in patients with SS were found to lead to: (1) problems with mastication; (2) alterations in taste sensation; (3) pain in the salivary glands; (4) speech difficulties; (5) nocturnal discomfort; and (6) an increase in fluid intake (Soto-Rojas and Kraus, 2002).

2.2.5.2.2 SGH in SS individuals

The objective sign of dry mouth (SGH) can manifest as clinical signs of dryness, or the consequences of a lack of saliva.

2.2.5.2.2.1 Clinical signs of dryness

Saliva plays an important role in the lubrication and protection of the tongue, lips, and buccal mucosa (Napeñas and Rouleau, 2014). Interestingly, in the early stages of SS, the mouth may sometimes appear moist due to sialorrhoea (Mignogna et al., 2005). However, as the disease progresses, dry mouth in SS individuals has been shown to deteriorate with time (Skopouli et al., 2000). The usual pooling of saliva in the floor of the mouth becomes absent. As the disease advances, the mouth can appear to have: (1) dry, cracked, and peeling lips; (2) a dry and coarse tongue; (3) cracks in the corners of the mouth; (4) an erythematous tongue; and (5) oral ulcers (Soto-Rojas and Kraus, 2002). Extreme SGH can cause the tongue to stick to the palate, and this may lead to a “clicking” sound in the speech of individuals with SS (Kassan and Moutsopoulos, 2004). The lack of lubrication by saliva may cause SS sufferers to sip water frequently,

and to have denture retention difficulties. These clinical signs demonstrate the possible chronic discomfort that SS sufferers may experience.

2.2.5.2.2.2 Increase in dental caries and erosion

In SS patients, chronic salivary gland inflammation leads to the loss of function, manifesting as SGH (Kassan and Moutsopoulos, 2004). Dental caries is a multifactorial (time, diet, substrate, and bacteria) disease described as the dissolution of hydroxyapatite crystals by acids produced from the bacteria in dental plaque. (González et al., 2013; Napeñas and Rouleau, 2014). Saliva contains immunoglobulins (IgA), histatins, defensins, lysozymes, cytokines, growth factors, mucins, and other components that can inhibit bacteria and fungi (Soto-Rojas and Kraus, 2002; Yan et al., 2011). In addition, saliva contains organic and inorganic components that can buffer pH changes. Furthermore, saliva contains proline-rich proteins that contribute to the formation of the dental pellicle, and to tooth surface mineralisation. It has also been shown that SGH in SS patients has been associated with high (cariogenic) bacterial levels of *Lactobacillus acidophilus* and *Streptococcus mutans* (Kolavic et al., 1997; Lundström and Lindström, 1994). Moreover, SGH has been associated with a longer time for sugar clearance and hence, greater levels of dental caries (Leone and Oppenheim, 2001). Dry mouth in SS may therefore give rise to greater smooth surface dental caries experience (on the cervical, incisal, and cuspal areas) and generalised dental erosion (Soto-Rojas and Kraus, 2002). As a consequence, SS patients have been found to have a higher DMFT (with associated higher dental expenses) in spite of better and more regular oral health care practices than the general population (Bøge Christensen et al., 2001). This was further confirmed by a large-scale study by Fox et al (2008), whereby SS individuals were shown to have a higher rate of dental care utilisation than control individuals (Fox et al., 2008). SS (together with other syndromes with salivary dysfunction) has also been reported to be associated with a greater risk of tooth wear because of the diminished buffering capacity due to the lack of saliva (Young et al., 2001). The impact of dry mouth on the QoL of SS patients will be explored in the next chapter.

2.2.5.2.2.3 Increase in oral infections

As mentioned, SGH results in poorer antibacterial and antifungal defence. SS patients have been found to have a high risk of oral candidiasis, and a high frequency of

recurrent *Candida* infections. In addition, azole resistance patterns among their *Candida* spp. were found, supporting the necessity for drug susceptibility testing for such patients (González et al., 2013; Napeñas and Rouleau, 2014; Yan et al., 2011). Intra-orally, *Candida* infection may present as a spectrum, consisting of: (1) pseudomembranous candidiasis; (2) acute atrophic candidiasis (erythematous); (3) chronic hyperplastic candidiasis; (4) median rhomboid glossitis; (5) chronic atrophic candidiasis (denture stomatitis), or (6) angular cheilitis. Both intra-oral and extra-oral *Candida* infections are often found in SS patients (Napeñas and Rouleau, 2014).

2.2.5.2.2.4 Swelling of the salivary glands

Swollen salivary glands (especially parotid glands) are common in SS patients. In the AECG criteria, one of the diagnostic questions (under oral symptoms) includes recurrent or persistently swollen salivary glands. Swelling of the salivary glands may begin unilaterally, then proceeding to become bilateral. It can also be chronic or episodic (Kassan and Moutsopoulos, 2004). In general, SS patients develop progressive major salivary gland swelling (typically bilateral), and it may be accompanied by discomfort. Such swellings are usually recurrent and benign. The pathophysiology of such a benign swelling is that of lymphocytic infiltration of salivary tissue, with associated parenchymal changes (Carr et al., 2012). A sudden swelling of a single gland suggests an infection (related to SGH). However, asymptomatic involvement of multiple glands with lymphadenopathy, of long-standing nature, may be an alert for lymphoma (Fox et al., 2008; Napeñas and Rouleau, 2014; Theander et al., 2011). In the event of a suspected lymphoma, appropriate investigations (such as magnetic resonance imaging of the involved glands) are required to establish a diagnosis. The greater risk of lymphoma in SS patients will be discussed in a later section.

2.2.5.2.2.5 Burning sensation

The sensation of oral burning (glossodynia) is a common oral manifestation of dry mouth in SS patients (Al-Hashimi et al., 2001). This can be attributed to fungal infections or SS-associated neuropathies. The differential diagnosis should also include anaemia, allergies, oral lesions, and burning mouth syndrome (Napeñas and Rouleau, 2014). The clinical manifestations of dry mouth common to SS patients are summarised in Table 3 (Soto-Rojas and Kraus, 2002).

Table 3: Signs and symptoms of oral dryness common in SS patients (after Soto-Rojas and Kraus, 2002)

Signs
Dry, cracked, and peeling lips; dry and coarse tongue
Cracks in the corners of the mouth
Dental decay, cervical, atypical (in incisal and cusp areas)
Dental erosion
Erythematous tongue
Swelling of the salivary glands
Mucositis
Oral candidiasis
Oral ulcers
Symptoms
Difficulties while swallowing and chewing dry foods
Sensitivity to spicy foods
Altered, salty, bitter, and metallic taste in mouth
Burning sensation
Lack of (or diminished) taste perception
Pain in salivary glands
Coughing episodes
Voice disturbances/speech difficulties
Increased liquid intake
Nocturnal discomfort

Table 3 provides a good overview of the signs and symptoms of dry mouth experienced by SS patients (Soto-Rojas and Kraus, 2002). Recently, Napeñas and Rouleau (2014) summarised the oral complications of SS with a few additional points on gastroesophageal reflux disease, periodontal disease, and headaches (Napeñas and Rouleau, 2014). There is evidence to support a higher rate of gastroesophageal reflux disease in SS patients (Mandl et al., 2007). In addition to SGH, this may further contribute to greater dental erosion in these patients. There seems to be conflicting evidence with regards to the association of periodontal disease or headaches with SS (Napeñas and Rouleau, 2014). Future research is needed to establish or discount these associations.

2.2.5.3 Systemic manifestations of SS

SS is a systemic condition with broad manifestations. It can be associated with organ-specific or systemic autoimmunity. It is not the intent of the current research to cover all of the systemic manifestations of SS in depth. However, the common ones (fatigue, musculoskeletal pain, and dry skin) will be briefly described. To conclude this section, a Table listing the various manifestations is included to provide a better understanding of the breadth of symptoms that may be experienced by SS sufferers.

2.2.5.3.1 Fatigue

Chronic, extreme fatigue has been found in SS patients. In a study of 94 American SS patients (applying the AECG-criteria), fatigue was found to be present in 67%, as measured by the Fatigue Severity Scale (Krupp et al., 1989; Segal et al., 2008). In a recent qualitative study, SS patients described the fatigue to be an ever-present, fluctuating, and non-relievable lack of vitality that is beyond their control. This forced these SS patients to scale down their everyday lives in different ways (Mengshoel et al., 2013). It is worth pointing out that this particular study is an example of how qualitative research can substantiate quantitative work and improve understanding of a condition.

2.2.5.3.2 Musculoskeletal and dermatologic involvement

Musculoskeletal manifestations (such as myalgias, arthralgias, and intermittent non-

erosive mild polyarthritis affecting mainly small joints) are common in SS patients (Pons-Estel et al., 2012). In secondary SS patients, RA may be an additional factor contributing to musculoskeletal involvement. SS sufferers commonly experience dryness in the skin and mucosal membranes. Cutaneous manifestations consist of xerosis (dry skin), angular cheilitis, eyelid dermatitis, pruritus (itch), cutaneous vasculitis, and erythema annulare (ring-shaped redness) (Soy and Piskin, 2007).

2.2.5.3.3 Other extraglandular signs and symptoms

SS can manifest as many other different conditions, these include haematological disorders, pulmonary interstitial diseases, kidney failure, peripheral and central neuropathies, and gastrointestinal tract disorders (Baldini et al., 2012b; Delalande et al., 2004; Ito et al., 2005). Not only does SS have a physical impact, the condition has been associated with anxiety and depression (Valtýsdóttir et al., 2000). This may be due to a downturn in life quality associated with the multiple symptoms. The effect of SS on QoL will be reviewed in the next chapter. The other extraglandular manifestations of SS are listed in Table 4 (Bayetto and Logan, 2010).

Table 4: Symptoms and conditions associated with extraglandular manifestations of SS (after Bayetto and Logan, 2010)

Malaise
Fatigue
Fibromyalagia
Fever
Arthralgia
Synovitis
Raynaud's phenomenon
Peripheral neuropathy
Autoimmune thyroiditis
Renal tubular acidosis
Chronic hepatitis
Vasculitis
Gastrointestinal symptoms
Respiratory diseases
Psychosis
Lymphadenopathy
Lymphoma

Table 4 may not cover all the possible systemic involvement, but it shows adequately the breadth of the different systems and organs that can be affected by SS, a systemic autoimmune disease. An alternative way of classifying these manifestations was later presented in another study on SS outcome measures by Seror et al (2012). The clinical features of SS were divided into two facets: (i) the benign, subjective but disabling manifestations such as dryness, fatigue, articular and muscular pain, affecting almost all patients; and (ii) potentially severe systemic manifestations, such as synovitis,

vasculitis, skin and renal involvement, neurological feature, and lymphoma (Seror et al., 2012). As a result of the multiple clinical features, there have been many tools developed to assess the various domains. Of those, Seror et al (2012) have found the European League Against Rheumatism (EULAR) Sjögren's Syndrome Patients Reported Index (ESSPRI) (Seror et al., 2011), and the EULAR Sjögren's Syndrome Disease Activity Index (ESSDAI) (Seror et al., 2010) to both be feasible, valid, and reliable instruments.

2.2.5.4 The prognosis of SS

The initial presentation of SS has been found to determine the subsequent outcome. Purpura, decreased C4 complement levels, and mixed monoclonal cryglobulinemia are adverse prognostic factors associated with greater overall mortality of SS patients than in the general population (Skopouli et al., 2000). The most serious condition associated with SS is malignant lymphoma; the risk of developing non-Hodgekin's lymphoma has been reported to be 44 times higher for primary SS sufferers than for age- and sex-matched controls (Kassan et al., 1978). In a retrospective cohort study of 55 SS patients, 9% had developed malignant low grade B-cell lymphoma over a period of 12 years (Zufferey et al., 1995).

2.2.6 The diagnosis of SS

This section provides some of the history behind the development of the AECG criteria (used in this research) that have been widely adopted by the scientific community. The difficulties associated with SS diagnosis will be introduced, leading to the development of the AECG criteria. The aspects of the criteria involving the mouth (dry mouth and labial gland biopsy) will be touched on, followed by a discussion of their strengths and drawbacks.

2.2.6.1 The development of the AECG criteria

As discussed in the previous sections, SS is a heterogeneous autoimmune disease that has a wide variation of clinical manifestations. Secondary SS can be associated with different connective tissue disorders (such as RA, SLE, SSc). Even though much is known about its pathogenesis and aetiology, some uncertainties remain. The complexity of SS has made it difficult to identify a homogeneous group of patients with a common aetiopathogenesis or prognosis in order to elaborate

classification/diagnostic criteria for the disease (Baldini et al., 2012b). Over the years, leading experts in the field have proposed several different classification criteria sets for SS (Fox et al., 1986; Manthorpe et al., 1985; Vitali et al., 1993). In 1993, the Preliminary European Classification criteria for SS were proposed (Vitali et al., 1993). These criteria were based on a six-item set. Any four of the six items were considered to be required for a diagnosis of SS. These diagnostic criteria had good sensitivity (94%) and specificity (94%) for the diagnosis of SS (Baldini et al., 2012b) The items included were: (i) ocular symptoms, (ii) oral symptoms, (iii) ocular signs (iv) histopathologic features of focal sialadenitis observed by labial gland biopsy, (v) salivary gland involvement, and (vi) the presence of autoantibodies. The main criticism of the Preliminary European Classification criteria is that misclassification can occur because the combination of items (i), (ii), (iii), and (v) may also be met by patients with sicca symptoms without the (iv) histology evidence of SS pathogenesis or (vi) specific disease markers autoantibodies (anti-Ro/La antibodies). Moreover, there may be misclassification of SS patients who are without subjective symptoms, because two of the six criteria items are devoted to subjective complaints (Vitali et al., 2002). To overcome these limitations, the AECG revised international classification criteria for SS maintained the previous six items but introduced the obligatory rule that, for a definite diagnosis of SS, either the minor salivary gland biopsy (reflecting iv) or serology for anti-Ro/SSA and anti-La/SSB (reflecting vi) had to be positive. Moreover, each constituent item was defined more precisely. The AECG criteria are most commonly applied in the diagnosis of SS in clinical trials, epidemiological studies, and clinical practice due to their high specificity and sensitivity (Baldini et al., 2012b; Vitali et al., 2002).

2.2.6.1.1 Dry mouth and labial gland biopsy in the AECG criteria

Xerostomia and SGH comprise 2 of the 6 AECG criteria. The assessment of xerostomia uses subjective questions. SGH can be observed by parotid sialography or scintigraphy, and measured by determining the unstimulated whole salivary flow. This involves collecting all of the saliva produced during 15 minutes. A final volume of <1.5ml (equivalent to an overall flow of <0.1ml/min) is considered to be abnormal.

The labial salivary gland biopsy for SS diagnosis was first introduced in 1968 (Chisholm and Mason, 1968). It is a procedure that is performed under local anaesthetic. The surgeon makes a shallow elliptical incision usually in the lower lip down to the muscle layer in order to harvest approximately 6-8 minor salivary glands. The incision is then closed with sutures (to be removed after 4 to 5 days). The harvested glands are then sent for histopathological examination. The AECG criteria defined a positive minor salivary gland biopsy to be at least 1 focus of lymphocytes within 4mm^2 of glandular tissue (focus score $\geq$ 1 per 4 mm^2), to indicate focal lymphocytic sialodenitis. Over the years, the technique has been revised to become less invasive, with smaller incisions and the use of resorbable sutures. There are associated complications, including: (1) localised numbness (which may last a few months or can be permanent); (2) external haematoma; (3) local swelling; (4) formation of granulomas (indicating inflammation); (5) internal scarring and cheloid formation; and (6) post-surgical pain (Delli et al., 2014).

2.2.6.1.2 Strengths and limitations of the AECG criteria

The AECG criteria allow homogeneity of patients enrolled in clinical trials or categorised in epidemiological studies. However, the drawback is that an invasive technique is mandatory simply to fulfill the classification criteria (blood test/labial salivary gland biopsy or both). Also, there has been criticism that the AECG criteria do not cover the broad clinical and immunological heterogeneity of glandular involvement. The sensitivity of the AECG criteria was questioned in a study by Ramos-Casals et al (Ramos-Casals et al., 2010a). In this study, 507 patients diagnosed with primary SS (using the 1993 Preliminary European Classification criteria) during 1984 to 2008 were followed up every 6-12 monthly (until 2008). Of these, 221 (44%) did not fulfill the 2002 AECG criteria but were found to have similar outcome (prevalence of sicca features, overall systemic involvement, anti-nuclear antibodies, complement levels, development of B-cell lymphoma, or survival) over the follow-up. Ramos-Casals et al went on to propose introducing two different definitions: “Sjögren’s disease” and “Sjögren’s syndrome, respectively for patients fulfilling or not fulfilling the AECG criteria (Ramos-Casals et al., 2010a). So far, this differentiated definition has not really been used in the literature.

2.2.6.1.3 More recent developments in SS diagnosis

The more recent American College of Rheumatology Criteria for SS classification has been developed (Shiboski et al., 2012). This classification requires far greater objective laboratory evidence of SS and could be regarded as being less sensitive but more specific than the AECG criteria. The reason is that the newer classification will determine the inclusion criteria for clinical trials on biologic immunomodulating agents. Therefore, the threshold for diagnosis is high due to drug toxicity. However, patients with severe signs and symptoms of the disease might not attain the positive laboratory markers.

There has been research to investigate the viability of a new biological clinical variable to be used for the diagnosis of SS. Based on the B cell involvement in the pathogenesis of SS, a study comparing 25 primary SS patients (diagnosed with AECG criteria) against 136 disease controls suggested that the distribution of B-cell subsets may provide a potential diagnostic tool (Binard et al., 2009). This is a promising area that still requires more research.

The difficulty in establishing highly sensitive and specific criteria for the diagnosis of SS is because of the heterogeneity and insidious onset of SS. The seemingly vague and wide breadth of possible signs and symptoms (and associated conditions) in turn impact on the length of time taken for diagnosis. In an American study of 547 SS patients (diagnosed using the AECG criteria), the mean time to diagnosis was 7 years (Segal et al., 2009). The authors also attributed the delayed diagnosis to the non-specific nature of the presenting symptoms, or to poor physician awareness of SS in general.

2.2.7 The management of SS

Because the aetiology and pathogenesis of SS are still not fully understood, there is no definitive cure for SS. In accordance with the B-cell mediated pathogenesis of SS, current research is exploring the use of biologic therapy (such as the use of monoclonal antibodies) to target B cells in SS. The objective of the management of SS is symptomatic relief and the prevention of secondary complications (Guggenheimer, 2003; Jonsson et al., 2011).

2.2.7.1 The general management of SS

Nonsteroidal anti-inflammatory drugs and antimalarials (such as hydroxychloroquine) may be used for the joint pain or mild arthritis. The common symptom of fatigue is usually not responsive to pharmaceutical treatment. Biologic therapy has a potential to benefit patients with SS. The most appropriate use of biologics appears to be in SS patients with severe systemic involvement, erosive arthritis, and high inflammatory activity. However their use early in the disease has the potential to prevent disease progression (Brito-Zerón et al., 2013; Gottenberg et al., 2013). Tumour necrosis factor-alpha blockade has not been proven to be effective in SS. However, B-cell depletion using rituximab (a monoclonal antibody against B-cells) has been shown to be of benefit, mainly in relation to the extraglandular features (arthritis, arthralgia, myalgia, and extreme fatigue), and (to some extent) dry mouth where there is still residual salivary function (O'Neill and Scully, 2013). A double-blind, randomised, placebo-controlled trial of 30 SS patients (diagnosed using the AECG criteria) showed that treatment with rituximab is effective in improving whole saliva flow rate, lacrimal gland function, and immunological levels (B cell and RF) (Meijer et al., 2010). In addition, rituximab is effective in the treatment of SS-associated (extrasalivary) lymphomas, with a poorer response found in salivary lymphomas. Rituximab is administered by intravenous infusion (single or periodic), and has notable potential side-effects. These include infusion allergic reactions and infections. It is important to therefore weigh the full risk/benefit ratio when considering the use of biologics for any SS patients. Clinical use is best performed and monitored in conjunction with rheumatologists well-trained in biologic therapies. Further studies of rituximab in SS are ongoing, and new agents under clinical trials include belimumab (a monoclonal antibody against B-cells) (O'Neill and Scully, 2013).

2.2.7.2 The dental management of SS patients

The dental management of SS patients targets the signs and symptoms associated with oral dryness. Each patient requires an individualised treatment regime with a heavy preventive focus. The management involves: (1) managing the symptoms of dry mouth and (2) the consequences of dry mouth (as covered in the previous chapter). Some additional dental management of the consequences of dry mouth (that are pertinent to SS patients) will be highlighted here. The higher risk of caries can be managed by

regular preventive fluoride therapy (including high fluoride toothpaste, gel, varnish and mouthrinse), microbial control (chlorhexidine gel or mouthrinse that is alcohol free), casein phosphopeptide-amorphous calcium phosphate (CPP-ACP) (tooth mousse), baking soda mouth rinse, hygiene education, diet counselling, oral moisturisers, salivary substitutes, sugar-free chewing gums, and sialogogues (such as pilocarpine and cevimeline) (Hamburger, 2001; Ramos-Casals et al., 2010b; Walsh, 2008). It is especially important to preserve the dentition of SS patients because of the inability to retain removable prosthesis as a result of inadequate saliva. Pilocarpine is a cholinergic parasympatheticomimetic agonist that binds to M3 receptors to stimulate exocrine glands (salivary and lacrimal). The associated increase in exocrine secretion may alleviate the symptoms of dry mouth. However, the benefits rely on there being residual unaffected portions of the salivary glands, and therefore they are most effective during the early stages of SS. The side-effects of pilocarpine may include headache, nausea, vomiting, and sweating (Tsifetaki et al., 2003). Oral candidiasis is a common complication of dry mouth. The predisposition to oral infections can be managed by topical anti-fungal treatments in the form of rinses, ointments, and pastilles. Systemic antifungal therapy is reserved for immunocompromised patients. Dentures should be soaked in chlorhexidine or sodium hypochlorite to prevent denture stomatitis (Napeñas et al., 2009). In recent years a novel technique to manage dry mouth, the intraoral electrostimulation device, has been researched on. This device consists of stimulating electrodes, an electronic circuit, and a power source. It delivers electrostimulation through the oral mucosa to the lingual nerve in order to enhance the salivary reflex. The device was tested on 114 SS patients in a prospective, randomised, multicenter trial. The device was found to alleviate oral dryness, discomfort, speech and sleeping difficulties, and to increase salivary output (Strietzel et al., 2011). However, this can work only with patients who have residual salivary gland function, and the comfort and practicality of using such devices need to be considered.

2.2.8 Conclusion

SS is a disease that is relatively uncommon, but not rare. It has a complex network of aetiology and pathophysiology that still has room for more research. The clinical manifestations of SS are broad. It is not unusual for several years to pass by before patients are diagnosed, and the biggest challenge is to improve therapy and diagnosis of SS. However, there is more to treating dry mouth in SS patients than just

symptomatic relief. The effects of dry mouth and SS on the quality of life of patients will be discussed in the next chapter.

2.3 Measuring and understanding the effect of dry mouth and SS on quality of life

2.3.1 Introduction

This chapter is about the effect of dry mouth and SS on people's quality of life (QoL). The background behind the link between QoL and health is based on the progression of health models. First, there will be a consideration of the advancement from the 'biomedical' model to the 'biopsychosocial' model. Next, the terms health related quality of life (HRQoL) and oral health related quality of life (OHRQoL) will then be defined. This will be followed by a brief highlight of the implications of these concepts in healthcare policy and practice. This will convey the importance of understanding the effects that dry mouth and SS have on QoL. Subsequently, there will be a discussion of the common indices used to measure HRQoL and OHRQoL. This will be followed by a review of the use of these indices in dry mouth and SS studies. Later, conceptual models of HRQoL will be used to link clinical variables (measured by health indices) to health outcomes. After that, there will be a discussion of the strengths and limitations of quantifying health outcomes in a conceptual model. The need to integrate other research methods (qualitative) in order to address some of the limitations will then be discussed in the next chapter.

2.3.2 Health models

In order to understand the relationship between health and QoL, there will be an overview of the underlying health model. In the mid-20th century, the concept of health progressed from a biomedical model towards a biopsychosocial model. Around that time, the World Health Organization (WHO) defined health as "a state of complete physical, mental, and social wellbeing, and not merely the absence of disease and infirmity" (World Health Organization, 1948)

2.3.2.1 The biomedical model

Traditionally in healthcare, the biomedical model has been implicit. The philosophy behind this model is that 'the self' and 'the body' are separate entities, where the body is viewed as a machine, and health and illness are strictly biological and objective. The

patient's subjective experience of health and disease are not considered. In this model, health is defined solely by the lack of disease (Wade and Halligan, 2004).

2.3.2.2 The biopsychosocial model

Over the past three decades, the biopsychosocial model has challenged the biomedical model. The philosophy behind this model is that the social and physical environments in which we live in are major determinants of health status. The patient is viewed as a person whose subjective experience of health and disease is important. In this model, health is defined in terms of optimal functional, social, and psychological well-being. This paradigm shift moved from a reductionist perspective of health (with only a biological aspect) to a more holistic thinking of health. The biopsychosocial model (Figure 2) proposed by Engel included both psychosocial and physiological components as potential contributors to ill-health (Engel, 1977).

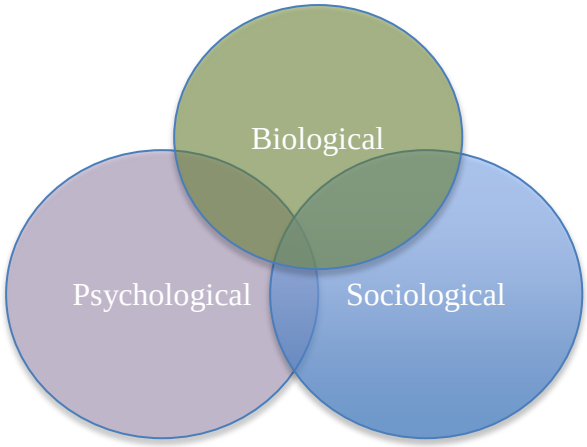


Figure 2. The biopsychosocial model (Engel 1977)

It is therefore important to develop ways to measure and understand perceptions, emotions, social, and functional aspects of the individual's experience of health and disease.

2.3.3 HRQoL and OHRQoL

The concepts of HRQoL and OHRQoL stem from the understanding of health to consist of biological, psychological, and social aspects. Although it is based on the same background health model, OHRQoL is a subset of HRQoL, and they will both be defined separately.

2.3.3.1 HRQoL

The concept of HRQoL regards health as a continuum that includes the negatively valued aspects (including death) and the more positively valued aspects of life (such as role function or happiness). The terms “health status”, “functional status”, and “QoL” are three concepts that are used interchangeably to describe HRQoL (Guyatt et al., 1993). Stewart and Ware have suggested that health perceptions integrate different components of health, values, expectations and beliefs (Stewart and Ware, 1992). Perceptions of QoL vary among individuals and are dynamic within each person, depending on his/her expectations and experiences of health. For example, different individuals experiencing the same clinical condition were found to report different QoL because of their different expectations (Carr et al., 2001). In addition, people with chronic disabling disorders may perceive their QoL as being better than healthy individuals (Allison et al., 1997), demonstrating the concept that the presence of disease may not always lead to poor QoL. Albrecht and Devlieger interviewed 153 persons with disabilities and found that 54.3% reported having excellent or good QoL (Albrecht and Devlieger, 1999). In accordance with Engel’s biopsychosocial model, HRQoL considers the physical, psychological, and social aspects of health (including global perceptions of function and well-being) (Berzon et al., 1993). The advantage of HRQoL measures is that they can be compared across populations with different diseases. However, in considering oral health, HRQoL measures are not sensitive to differences in oral health (Allen et al., 2001). OHRQoL measures are more likely to detect subtle changes in oral health than HRQoL measures.

2.3.3.2 OHRQoL

OHRQoL is a multidimensional construct that refers to the extent to which oral disorders disrupt an individual’s normal functioning. It can be considered to be a facet of HRQoL (Baker et al., 2007).

Traditionally, oral disease has been measured using clinical indices such as the Community Periodontal Index of Treatment Needs (CPITN) (Ainamo et al., 1982) and the decayed, missing, or filled teeth index (DMFT) (Klein et al., 1938). These are important objective measures for identifying the presence or absence of disease. However, they demonstrate only the end of the disease process, without reflecting the

impact of the disease process on social function or psychological well-being, as perceived by the individual. Corresponding to changing health perceptions from the biomedical model to the biopsychosocial model (as described earlier), it is important to understand the multi-dimensional nature of health. Recent research has been directed towards QoL because physiologic measures provide information for clinicians, but are of limited relevance to patients. This is because the physiologic measures often correlate poorly with functional capacity and well-being, the areas with which patients are most concerned (Guyatt et al., 1993).

Locker described a conceptual model (Figure 3) for measuring oral health. It considers the physical, psychological, and social aspects of disability that may be experienced by patients due to oral disease (Locker, 1988). The domains in this model are based on the WHO's classification of impairments (disturbances at the organ level), disabilities (disturbances at individual level), and handicaps (disadvantages of the interaction of the individual with the environment), with the aim of improving information on the consequences of disease (World Health Organization, 1980).

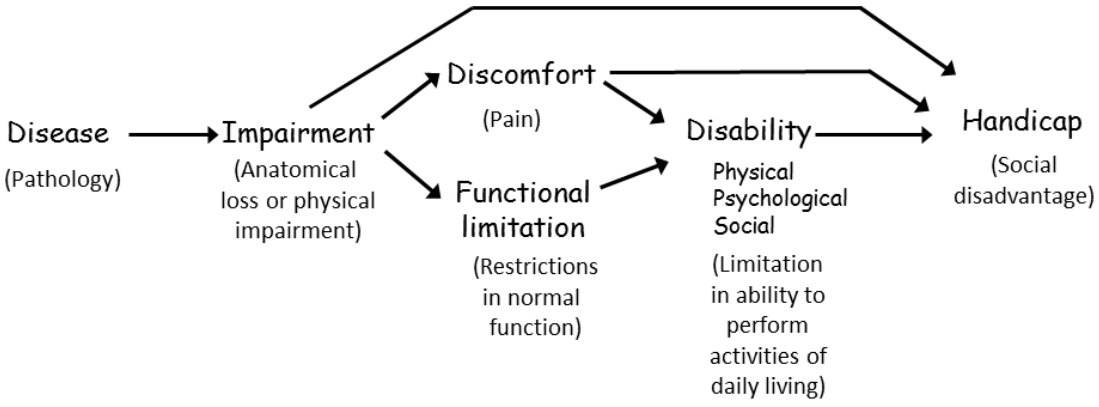


Figure 3. Conceptual model for measuring oral health (Locker 1988)

Locker and Allen (2007) defined OHRQoL as the impact of oral disorders on aspects of everyday life that are important to an individual, with those impacts being of sufficient magnitude, whether in terms of severity, frequency or duration, to affect the individual's perception of his/her life overall (Locker and Allen, 2007).

2.3.3.3 The Wilson and Cleary model

Wilson and Cleary proposed a conceptual model (Figure 4) of patient outcomes to link clinical variables with HRQoL (Wilson and Cleary, 1995). The model encompasses disease, health, and QoL; it combines concepts from both the biomedical model and the biopsychosocial model. The biomedical model is represented in the causal relationships among the clinical levels: (1) physiological/biological variables; (2) symptom status; (3) functional status; (4) health perceptions; and (5) quality of life. The biopsychosocial model is represented by the characteristics of the individual and environmental, as mediators of health outcome. Examples of how this mediation may occur are depicted by words along the arrows in the Wilson and Cleary model. The model demonstrates the importance of including both the usual clinical variables and environmental and individual characteristics in considering the factors associated with QoL (Wilson and Cleary, 1995). It is important to note the dynamic and complex relationship among biological, social and psychological concepts in the model; each level in the model (boxes) may be independent to another, and arrows may indicate a uni- or bi-directional relationship.

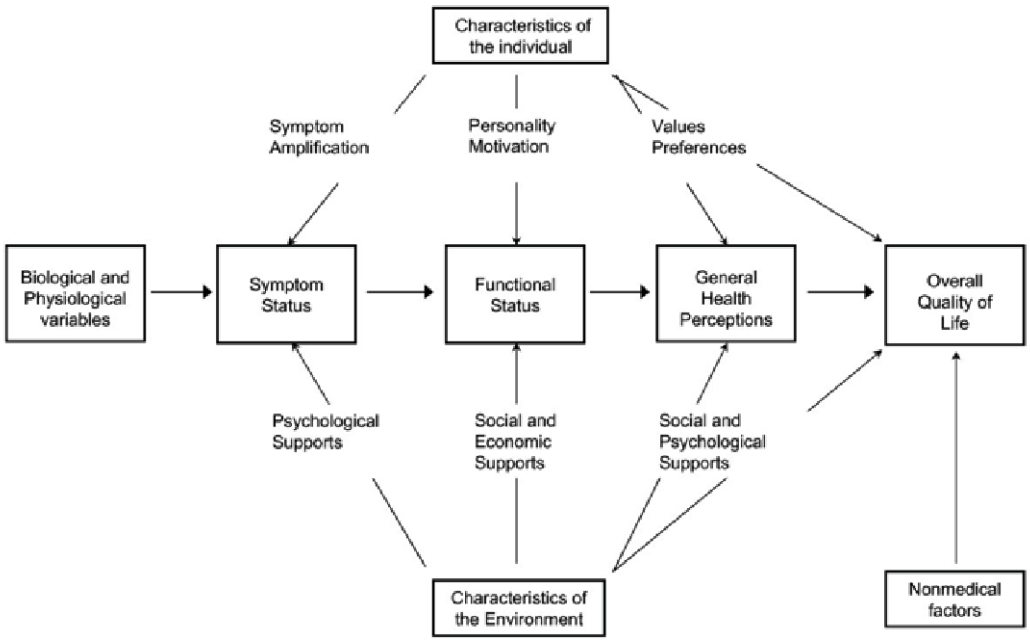


Figure 4. The Wilson and Cleary model (Wilson and Cleary, 1995)

The development of the Wilson and Cleary model (1995) will be discussed later in this chapter.

2.3.4 The importance of measuring HRQoL and OHRQoL

As described, HRQoL is important for measuring the impact of diseases with consideration of individual perspectives. For example, a patient-based assessment of health status measurement is required to understanding the outcomes of clinical intervention because there may be a difference between the clinician's opinion and the patient's own perception of his/her HRQoL and treatment need (Slevin et al., 1988). This will also allow insight into individuals' perceived oral health needs and motivations to seek health care. In health economics, HRQoL measures have become the standard means of assessing the effectiveness of health care intervention and the planning and prioritising of funding (Fitzpatrick et al., 1992). Similarly, it is crucial to measure the patient's perception of oral health and disease prevalence for accurate allocation of health funding and resources, for public health promotion and disease prevention programmes (Slade and Spencer, 1994).

2.3.5 Methods to measure HRQoL and OHRQoL

HRQoL and OHRQoL are subjective, and can therefore be measured only with questions. These range from single-item methods to multi-item summated rating scales. The latter provides information on the different domains of HRQoL and OHRQoL. A critique on the wide array of available measures is not the key focus of this research. However, an overview of the commonly used measures will provide background information on the effect of dry mouth and SS on QoL. Examples of the use of these measures will be given in the next section on the impact of dry mouth and SS on QoL.

2.3.5.1 Single-item approaches

Similar to measuring xerostomia, a single question (global item) is a statement which requires the respondent to integrate their experiences and perceptions of their health (or oral health) into an overall summary judgment.

The classic self-rated health status single question consists of asking respondents to rate their health as ‘excellent/very good/good/fair/poor’. This is because single-item measures measuring subjective health and well-being do not necessarily correlate with medical diagnoses. However, such measures were held to have greater validity in predicting help-seeking behaviours and health service use (Bowling, 2005). The visual analogue scale (VAS) is another frequently used single-item technique, in which the respondent places a mark on a horizontal line to indicate his/her QoL during a specified period. The line is anchored from the ‘lowest quality’ to the ‘highest quality’ at the ends. The simple VAS single-item approach has been shown to be reliable, valid, and sensitive, including its: (1) ability to discriminate between healthy and sick respondents; (2) sensitivity to the different stages of the disease progress, and (3) ability to predict mortality (Bowling, 2005).

Similarly, an example of a single-item used to measure OHRQoL is the global oral health status rating question: “How would you describe the health of your teeth or mouth?” The five possible responses to this item range from ‘poor’ to ‘excellent’ (Locker, 2001). A study on young middle-aged adult in populations in New Zealand and Australia investigated the clinical validity of Locker’s global self-reported oral

health item (Thomson et al., 2012). The proportion of disease in the population borne by those who responded with 'Fair' or 'Poor' ranged from 26% to 72%. These findings indicated that the Locker item provided an adequate summary of how individuals view their oral health. The measure is valid as a global self-reported oral health measure (unifying both subjective perceptions and objective observations) in young middle-aged adults. Such single-item questions can provide valuable information in a simple and economical way. However, this may be at the expense of detail and diversity of responses (Bowling, 2005; Dolan et al., 1998).

2.3.5.2 Multi-item approaches

On the other hand, multi-item measures aim to capture the complete profile of the different dimensions of HRQoL and OHRQoL. An example of a commonly used HRQoL multi-item measure is the 36-Item Short-Form Health Survey {SF-36; (Ware and Sherbourne, 1992)}. An example of a popular measure of OHRQoL in the general population is the Oral Health Impact Profile, or OHIP (Slade and Spencer, 1994). In consideration of the higher proportion of the older population affected by dry mouth and SS, a commonly used multi-item measure to assess OHRQoL in older adults, the Geriatric/General Oral Health Assessment Index will also be considered {GOHAI; (Atchison and Dolan, 1990)}.

2.3.5.2.1 The SF-36 (HRQoL)

The SF-36 is the most widely used multidimensional HRQoL measure (Bowling, 2005). This is a multipurpose, generic health survey instrument used to measure health (Ware and Sherbourne, 1992). The SF-36 consists of questions to measure 8 dimensions of HRQoL, including: (1) physical functioning; (2) social functioning; (3) physical role limitations; (4) emotional role limitations; (5) mental health; (6) energy; (7) pain; and (8) general health perceptions. It is easy to use, acceptable to patients, and fulfills the stringent criteria of reliability and validity (Brazier et al., 1992). It has been found to be useful in: (a) comparing general and specific populations; (b) comparing the relative burden of disease; (c) differentiating between the health benefits produced by a wide range of treatments; and (d) screening individual patients for the presence/absence of disease (Ware, 2004). Considering its convenience for practical applications, shortened versions of the SF-36 have been developed in order achieve reductions in respondent burden. These include the SF-12 (12 items) and the SF-8 (8

items). The shortened versions are inevitably less sensitive than the full versions, however; because they are based on the most powerful items from the parent instruments, a high degree of accuracy remains (Bowling, 2005).

2.3.5.2.2 The OHIP (OHRQoL)

Similarly, OHRQoL consists of the many dimensions that patients deem most relevant to their overall health and well-being. The OHIP (Slade and Spencer, 1994) was developed with the aim of providing a comprehensive measure of self-reported dysfunction, discomfort and disability attributed to general oral conditions (Slade, 1996). Based on Locker's conceptual model of measuring oral health (Locker, 1988), the OHIP is a 49-item measure consisting of statements divided into 7 domains including: (1) functional limitation; (2) pain; (3) physical disability; (4) psychological discomfort; (5) psychological disability; (6) social disability; and (7) handicap. An example of a statement is "Have you had to interrupt meals because of problems with your teeth, mouth, or dentures". An answer to each statement is in the Likert response format (0 = never, 1 = Hardly ever, 2 = occasionally, 3 = fairly often, 4 = very often). Each statement is weighted in order to facilitate the assessment of perceived severity of impacts. Overall OHIP scores can be counted in two different ways. The first method is to total the number of impacts reported at a threshold level (for example, "fairly often" or "very often") used as the dependent variable. This variable is used because it conveys a readily interpreted impression of frequent impacts on well-being (Slade et al., 1996). However, the distribution of this summary variable may be skewed, with many individuals reporting no impact at this threshold. The second method is to compute the sum of standardised subscale scores. The standardised OHIP variable captures a full range of responses to each of the 49 items (Slade et al., 1996). However, this method requires more computer programming, and has less intuitive appeal than the first method (Slade, 1996). The advantage of the OHIP is that its statements were derived from a representative group of dentate people, and not conceived by dental clinicians or researchers (Allen, 2003), thus allowing a more accurate measurement of self-perceived oral health status. It is considered to be the most sophisticated self-report measure of oral health (Locker, 1998). In order for the more convenient use of the OHIP in the clinical setting, the shorter Oral Health Impact Profile 14{OHIP-14; (Slade, 1997)} was derived. In theory, a shortened version of the OHIP may have compromised validity. However, the OHIP-14 has been shown to demonstrate good

levels of reliability, validity, and responsiveness in both the general and elderly population (Fernandes et al., 2006; Locker et al., 2001).

2.3.5.2.3 The GOHAI (OHRQoL)

Considering the aging global population, an index was developed to assess the impact of oral health problems on older adults – the GOHAI (Atchison and Dolan, 1990). The GOHAI assesses OHRQoL in three hypothesised dimensions: (1) physical function (including eating, speech, and swallowing); (2) psychological function (including concern about oral health, dissatisfaction with appearance, self-consciousness about oral health, and avoidance of social contact because of oral problems); and (3) pain or discomfort (including use of medication to relieve oral pain or discomfort) (Dolan et al., 1998). The GOHAI contains 12 statements (eg. "How often did you feel uncomfortable eating in front of people because of problems with your teeth or dentures") with a Likert response format (i.e. 0 = never, 1= seldom, 2 = sometimes, 3 = often, 4 = very often, 5 = always). Response codes are totaled to give a score from 0 to 60. The GOHAI assesses the oral health problems of older adults, and those with fewer teeth, wearing a removable denture, and perceiving a need for dental treatment were significantly related to a worse (lower) GOHAI score (Atchison and Dolan, 1990). Interestingly, higher scores on the GOHAI represent better OHRQoL; for the OHIP-14, it is the other way around.

2.3.5.2.4 The significance of using OHRQoL measures and HRQoL measures

Both the OHIP-14 and the GOHAI were found to discriminate between dentate individuals with and without dentures, with and without a chewing problem, and with and without dry mouth. Both measures also showed significant associations with self-rated oral health and satisfaction with oral health status. In addition, these measures were equally good at predicting overall psychological well-being and life satisfaction (Locker et al., 2001), thereby, further confirming the significance of oral health in relation to general health and well-being.

HRQoL and OHRQoL have been developed to understanding the patients' perspective of the impact of general and oral health problems on many aspects of their lives. As described, this understanding will help in the: (1) dissemination of public health funding; (2) understanding of treatment needs from patient perspective; (3) planning of

health promotion and disease prevention. When applied to specific conditions such as dry mouth and SS, the impact of these conditions on quality of life can be measured.

2.3.6 HRQoL and OHRQoL in dry mouth and SS

HRQoL and OHRQoL measures provide evidence that dry mouth and SS (both separately and together) negatively affect the QoL of patients. Dry mouth has been shown to negatively impact on the QoL of people {(Ikebe et al., 2007), (Thomson et al., 2006a), (Locker, 2003b)}. Similarly, SS has been shown to negatively impact on the QoL of its sufferers {(Segal et al., 2009), (Thomas et al., 1998)}. Oral distress has been found to be prevalent and significantly increased in patients with SS, with marked effect on their HRQoL (Enger et al., 2011).

Dry mouth affects the well-being of its sufferers, ranging from the young to older adults, as well as from healthy to medically compromised individuals. In the Dunedin study, the OHIP-14 was employed to measure the OHRQoL in 972 32-year-old participants, and dry mouth was found to have marked and consistent effects on its sufferers' day-to-day lives (Thomson et al., 2006c). In a Japanese study, the OHIP-14 was used to measure the OHRQoL in 287 independently living, relatively healthy Japanese older population. Subjective oral dryness and lower salivary flow rates were significantly associated with poorer OHIP-14 scores (Ikebe et al., 2007). Locker used two OHRQoL scales (OHIP-14 and the GOHAI) to measure OHRQoL in 225 medically compromised elderly residents in a Toronto geriatric residential facility, and found that dry mouth had an important influence on the well-being and QoL of that population (Locker, 2003b).

Research with HRQoL measures shows that SS patients are suffering with clinically significant impairment of their well-being. In a study using both the extensive Assessment of Symptoms and Experiences of SS (ASSESS) questionnaire and the SF-36, it was found that delayed diagnosis, sicca-related morbidity, fatigue, pain, and depression were more severe in 547 SS participants than in 606 controls. The findings suggested unmet health needs among the former, and the importance of earlier recognition of SS (Segal, Bowman et al. 2009). In a population-based survey of 1000 adults, those who were positively diagnosed with SS (3-4% of sample) were more

impaired in each of the 8 dimensions of the SF-36, than those without such a diagnosis. Those diagnosed with SS also suffered from higher levels of depression and fatigue (Thomas et al., 1998). A Spanish study of 110 SS patients by Belenguer et al. (2004) found them to have significantly lower SF-36 scores than the control population. The poorer HRQoL observed in these patients was associated with systemic expression of SS, such as pulmonary involvement. The findings revealed the importance of earlier diagnostic and therapeutic management of these patients (Belenguer et al., 2004).

Dry mouth has shown to negatively impact on the health of SS patients. In a Dutch study comparing 235 SS patients to age- and sex-matched controls from the general population, the former scored lower on the SF-36 than the latter. The use of artificial saliva (for dry mouth) by the patients acted as a marker for SGH. The findings revealed that the use of artificial saliva was one of the predictors of poorer HRQoL, together with fatigue, tendon myalgia, comorbidity, male sex and eligibility for disability compensation. In addition, employment rates were lower and disability compensation rates were higher in SS patients than in the general Dutch population (Meijer et al., 2009). The OHIP-14 and SF-36 were used to measure QoL in 177 Norwegian SS patients by Enger et al. (2011), and the impact of oral diseases on HRQoL in SS patients was found to be severe. Such distress may affect patients' confidence and self-esteem in social-role-dependent and emotional relationships (Enger et al., 2011). In an American study of 39 SS patients (using the OHIP-14 and the SF-36) to investigate the effect dry mouth on QoL, the former was found to have an independent influence on the latter (Stewart et al., 2008). The clinical implications of the study were that dentists and physicians should collaborate to maintain QoL for SS patents, and that dry mouth requires aggressive management. The findings of these studies verify that dry mouth and SS can be detrimental to the physical, psychological, and social aspects of people's welfare.

2.3.7 The development of the Wilson and Cleary model

So far, it has been shown that the patient perception of health status is not merely influenced by the clinical lack of disease. Attempts to understand a person's perception of how a condition affects his or her subjective well-being and functionality has led to the development of HRQoL and OHRQoL measures. In general, the literature shows that dry mouth and SS adversely impact on multiple aspects of an individual's life. To

make sense of how all these domains are linked together to reflect clinically meaningful health outcomes, conceptual models of HRQoL and OHRQoL have been developed. The components of a well-utilised model, the Wilson and Cleary model, together with its OHRQoL and revised version, will be explored. The utility, advantages, and disadvantages of such models will be expounded on in the next section.

2.3.7.1 Research on the Wilson and Cleary model

So far, research using the Wilson and Cleary model has been based on using QoL measures to quantify each level (box), drawing conclusions on (patients' perspectives on) HRQoL. The Wilson and Cleary model has been widely applied to help understand HRQoL in a range of populations, including patients with opioid dependency (Heslin et al., 2011), cardiac diseases (Heo et al., 2005; Janz et al., 2001), Hodgkin's Lymphoma (Wettergren et al., 2004), AIDS (Sousa and Kwok, 2006), and Parkinson's disease (Chrischilles et al., 2002), as well as an edentulous geriatric population (Baker et al., 2008), and, closer to home, dry mouth (Baker et al., 2007). The latter study will be described in order to consider the application of the Wilson and Cleary model to dry mouth.

In the study by Baker et al (2007) (similar to other research using the Wilson and Cleary model), components in the model were quantified (Baker et al., 2007). The study tested the pathways among the levels in the model, in relation to the OHRQoL of patients suffering from dry mouth. Using the levels of the Wilson and Cleary model the following measurements were made: (1) biological and physiological aspects using salivary flow and clinical signs; (2) symptom status using the XI (Thomson et al., 1999b); (3) functional status using the OHIP-14 (Slade, 1997); (4) general health perceptions were measured by a single-item global oral health rating (as described in section 4.5.1); and (5) overall quality of life, using the Hospital Anxiety and Depression Scale (HADS) (Zigmond and Snaith, 1983). Such quantification allows systematic modeling of the direct and indirect pathways hypothesised within the Wilson and Cleary model of patient outcome. The findings of Baker et al (2007) supported the construct validity of Wilson and Cleary's model for oral health, and highlighted the complex relationships in dry mouth. For example, the pathways

between the levels in the Wilson and Cleary model are not fully accounted for by the measured variables, suggesting an important role for individual and environmental characteristics. In addition, an understanding of the impact of dry mouth on individuals' daily lives is needed in order to gain a full understanding of the key underlying processes. To address these gaps in knowledge, Baker et al (2007) suggested that multi-method (qualitative methods or diary designs) approaches are necessary to understand the complexities of individual experiences of OHRQoL. The importance of this would be in developing effective intervention strategies to target what is important to an individual patient (versus interventions solely targeting biological status). Further recommendation was given to consider individual and environmental characteristics, and target 'high-risk' individuals with a focus on stress management and developing effective coping strategies (Baker et al., 2007).

2.3.7.2 The Wilson and Cleary model in relation to OHRQoL

Similarly, Locker and Gibson adapted Wilson and Cleary's model to portray OHRQoL, presenting a modified and simplified version that is adapted to oral health (Locker and Gibson, 2005). This adapted model (Figure 5) recognises the different dimensions of human experience, because it relates to clinical conditions and their outcomes, and specifies the probably main causal relationships between them (Locker and Gibson, 2005).

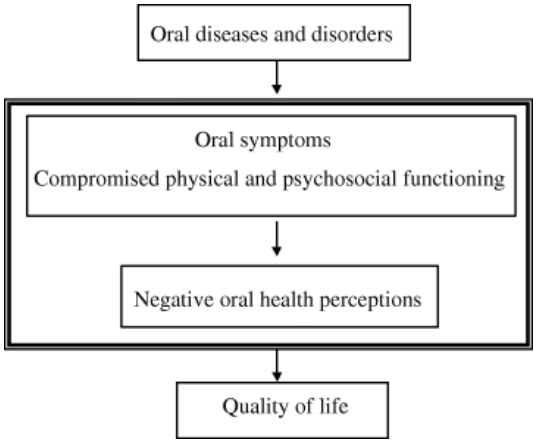


Figure 5. The Wilson and Cleary Model: adapted and applied to oral health (Locker and Gibson, 2005).

Similar to the concept behind the Wilson and Cleary model, OHRQoL is explained to have a sequence beginning from: (1) oral diseases that can result in (2) symptoms, that may affect an (3) individual's perception of oral health, subsequently affecting his or her (4) subjective well-being. This adapted model by Locker and Gibson provides a good general concept of OHRQoL. However, there seems to be less of a focus on the characterisctics of the individual and the environmental. Another revised version of the Wilson and Cleary model looks into these two areas.

2.3.7.3 The revised Wilson and Cleary model

Ferrans et al made revisions to the popular original Wilson and Cleary model to improve its application for health care research and clinical use (Ferrans et al., 2005). In addition, they took a further step to present the theoretical grounding on the levels: characteristics of (1) the individual; and (2) the environment, including their effects on other levels of the model. Previously, the measures that can be used to quantify each level of the Wilson and Cleary model in relation to dry mouth have been explained. Because the research findings will be based upon the revised Wilson and Cleary (Figure 6) as a framework, each level will be explained in relation to dry mouth and SS.

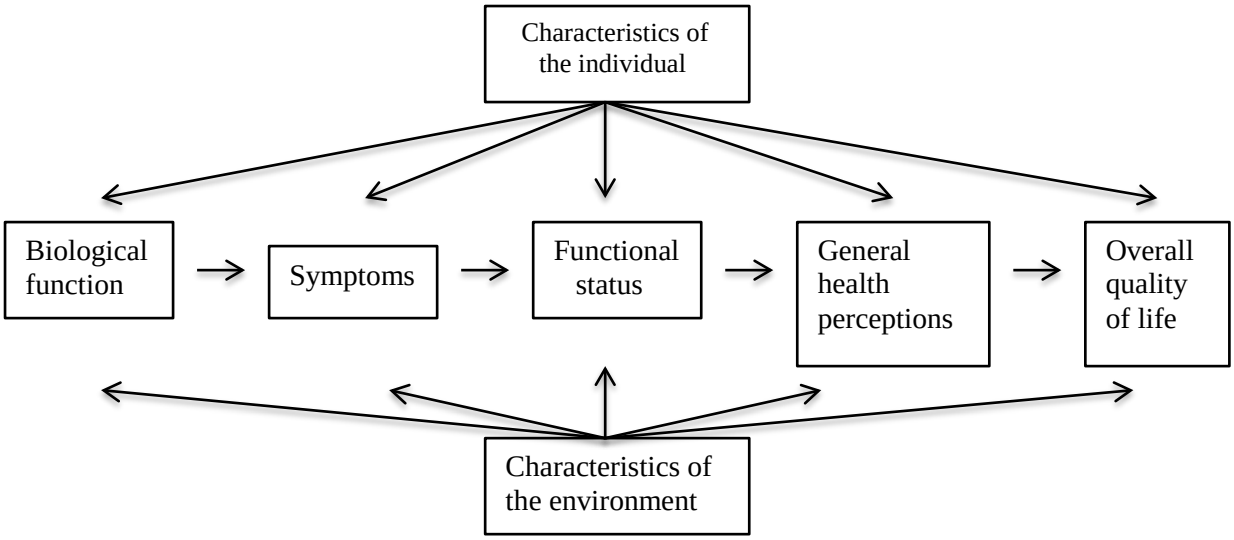


Figure 6. Revised Wilson and Cleary model for HRQoL, adapted from Ferrans et al. (2005)

The original model was revised in three substantive ways. First, arrows were added to indicate that biological function is influenced by both characteristics of the individuals

and the environment. Second, the non-medical factors from the original model were excluded, as they can be categorised into characteristics of the individual and the environment (that is already included in the model). Third, the examples labeling the arrows in the original model were omitted because it impossible for comprehensive description of the relationships between levels due to the diversity in HRQoL (Ferrans et al., 2005).

2.3.7.3.1 Characteristics of individual and environment

Health is mediated by characteristics of the individual and the environment. Characteristics of the individual are categorised as demographic, developmental, psychological, and biological factors that influence health. Characteristics of the environment are categorised as either social or physical (Eyler et al., 2002). Examples of social environmental characteristics are the influences of family, friends, and healthcare providers (McLeroy et al., 1988) and cultural heritage. Examples of the physical environmental characteristics are home, neighbourhood, and work settings. These will be further illustrated by empirical evidence from this research in the chapter on results.

2.3.7.3.2 Biological function

Biological function includes the dynamic processes required for life support such as molecular, cellular, and organ processes. In SS and dry mouth, these are the commonly measured parameters of anti-Ro/anti-La autoantibodies count and salivary flow rate. Biological function can be affected by characteristics of the individual and the environment. For instance, an individual's knowledge and attitude on a disease can influence lifestyle choices that may affect biological function. In addition, living in a hot and dry climate may result in one experiencing dry mouth to a larger extent than someone living in a less dehydrating environment. Changes in biological function can directly/indirectly influence other levels of health in the model, including symptoms, functional status, general perceptions of health, and overall quality of life. It is therefore important to optimise biological function in the management of dry mouth.

2.3.7.3.3 Symptoms

This is a shift from the cellular and organism level to a personal level, with symptoms being defined as "a patient's perception of an abnormal physical, emotional, or

cognitive state" (Wilson and Cleary, 1995). Instruments to measure dry mouth in SS include the XI (Thomson et al., 1999b). The interpretation of symptoms is influenced by complex interactions with individual (such as personality) and environmental factors. A longitudinal study of 366 community-dwelling middle-aged and older adults demonstrated how individual characteristics (psychological stress) can influence the decision to seek medical care for symptoms (Cameron et al., 1995). The study demonstrated that the presence and timing of other stressful events in life affected whether an individual seeks medical help for symptoms. In addition, those who experienced symptoms concurrently with other stressful events reported more negative mood states, rating their symptoms as more distressful than those who experienced the symptoms in the absence of stressful events. The interactions between an individual and his/her symptoms may be a complex bi-directional relationship.

2.3.7.3.4 Functional status

Wilson and Cleary defined functional status as the ability to perform tasks in the physical, role, social, and psychological domains. Functional status can be viewed as loss of function and its effect on daily life (Stineman et al., 2005), or optimisation of remaining function (Leidy, 1994). HRQoL and OHRQoL measures aim to understand the impact of disease on the multiple domains of functional status. In HRQoL, functional status is commonly measured by the physical and social functioning scales in the SF-36 Health Survey (Ware and Sherbourne, 1992). For OHRQoL, measures such as the GOHAI (Atchison and Dolan, 1990) and the OHIP (Slade and Spencer, 1994) have components that document the functional impacts of oral disorders on an individual. Some of the possible effects of oral problems on functional status are pain, problems with eating and sleeping, and difficulties in social situations (Locker and Quinonez, 2009). Similarly, the shortened XI (Thomson et al., 2011) consists of two functional impact items " I have difficulty in eating dry foods" and "I have difficulties in swallowing certain foods". These items document the negative influence of dry mouth on functional status. The characteristics of the individual and the environment can affect the negative impact of dry mouth and SS on functional status. For example, the extent of the decline in daily activities that can be influenced by the personality and lifestyle of an individual. A sporty, motivated person may attempt more activities than a more docile, sedentary individual. Environmental characteristics such as community

support for physical activities may also influence the impact of dry mouth and SS on a patient's functional status.

2.3.7.3.5 General Health Perceptions

Wilson and Cleary pointed out that general health perceptions are subjective in nature, integrating all the health concepts (levels) that appear in sequence in the model (biological function, symptoms, and functional status) (Wilson and Cleary, 1995). General health perceptions can be measured using single-item measures or items within the SF-36 (Ware and Sherbourne, 1992). An example of such an item in the SF-36 would be: "In general, would you say your health is excellent/very good/good/fair/poor". In addition, the change in general health perception with time is considered by the next item "Compared to one year ago how would you rate your health in general now?" The possible responses include: 'much better now than a year ago', 'somewhat better now than a year ago', 'about the same as one year ago', 'somewhat worse now than a year ago', 'much worse now than one year ago'. The characteristics of an individual that can affect health perceptions include values, expectations, and beliefs (Stewart and Ware, 1992). These will be illustrated using empirical findings from the research.

2.3.7.3.6 Overall QoL

The final level of the model was described by Wilson and Cleary as a subjective well-being with general measures of how happy and/or satisfied someone is with life as a whole (Wilson and Cleary, 1995). Overall QoL is commonly measured by a single global question asking how satisfied an individual is with life in general, or through a series of questions exploring satisfaction in various domains of life, such as those in the QoL Inventory (Frisch et al., 1992). An individual's general subjective well-being can be made up of multiple components including pleasant and unpleasant affect, life satisfaction, and domain satisfactions, as illustrated in Table 5. (Diener et al., 1999)

Table 5: Components of subjective well-being (after Diener et al., 1999)

Pleasant affect	**Unpleasant affect**	**Life satisfaction**	**Domain satisfactions**
Joy	Guilt and shame	Desire to change life	Work
Elation	Sadness	Satisfaction with current life	Family
Contentment, Pride	Anxiety and worry	Satisfaction with past	Leisure
	Anger		Health
Affection	Stress	Satisfaction with future	Finances
Happiness	Depression	Significant others' views of one's life	Self
Ecstasy	Envy		One's group

The range of components in the Table highlights the complexity of subjective well-being. This illustrated how overall QoL can be affected by characteristics of the individual (positive and negative emotions, and life satisfaction) and the environment (family, friends, and work).

2.3.7.3.7 Strengths of the revised Wilson and Cleary model by Ferans et al (2005)

The revised Wilson and Cleary model by Ferrans et al (2005) conceptualises HRQoL or OHRQoL (based on research testing the Wilson and Cleary model) (Ferrans et al., 2005). Research using such a model improves the understanding of multidimensional components and pathways of HRQoL and OHRQoL, which has clinical and health policy applications.

In healthcare research, it is essential to align research aims with the needs and values of patients (and their families) to provide a realistic overview of real-world situations. Owing to the multiple levels of HRQoL and OHRQoL, conceptual models act as schematic representations to provide understanding, by depicting the linkages between each level. In a recent systematic review of HRQoL models, the Wilson and Cleary

model and its revised version by Ferrans et al (2005) were found to be among the top three most frequently cited in the literature. In addition, the latter was recommended to be used in future research because the added individual and environmental characteristics better depict HRQoL (Bakas et al., 2012). Research on conceptual health models can advance the state of the science of HRQoL by contributing new information about the applicability of the model to research and practice, leading to model refinement (Bakas et al., 2012). Also, research testing of the model for theory development assists in the understanding in the causal mechanisms for health and identify points for clinical intervention (MacKinnon et al., 2002).

Research on the revised Wilson and Cleary model by Ferrans et al has the robust potential for design of interventions that can be tested and applied in clinical settings (Bakas et al., 2012). These interventions will not be solely based on biological function; they will also consider the patient's experience of the disease. This can include the understanding of clinical therapy outcomes from a patient perspective, as well as the motivation for patients to seek treatment.

From the individual to community level, an accurate insight into patients' experiences can play an important role in economical and effective public health resource allocation, health promotion policies, and disease prevention policies.

2.3.7.3.8 Limitations to the revised Wilson and Cleary model (2005)

As described earlier, HRQoL conceptual models such as the Wilson and Cleary model revised by Ferrans et al (2005) have been tested only using HRQoL measures to describe its clinical components. The concept of HRQoL and OHRQoL is based on the significance of patients' subjective experiences of diseases. Though meaningful, there are limitations as to how much these conceptual models (using HRQoL/OHRQoL measures) can provide insight into patient perceptions in terms of: (1) difficulty in quantifying experience; (2) conceptualising disease experience affected by individual and environmental characteristics; (3) conceptualising changes in patient experience over time; and (4) conceptualising the effect of treatment. These will be discussed in the context of OHRQoL.

2.3.7.3.8.1 Limitations in quantifying an experience with an index

While QoL indices cover a wide range of domains, their pre-determined questions, criteria and scores may not specifically elicit information on patient experience. For example, both improvement and deterioration in OHRQoL can occur simultaneously, as illustrated by Slade in a longitudinal observational study using the OHIP (Slade and Spencer, 1994) to measure OHRQoL at baseline and at a two-year follow-up visit (Slade, 1998). The results reported that tooth loss could alleviate pain, but decrease chewing efficiency, therefore resulting in OHRQoL improvements for some, and decrease for others. In addition, the importance of an item in a HRQoL/OHRQoL measure differs for each individual, and is difficult to generalise and measure. The importance of an impact may be indicated by a weighted response that divides the summed responses of a particular domain over the total possible score. However, the actual contribution of weights to the performance of OHRQoL measures has been questioned, and weights are rarely used these days (Allen and Locker, 1997; Locker et al., 2007; McGrath and Bedi, 2004). The severity of an impact to the patient will be best described by the patient in his or her own words in the context of social and environmental situations.

2.3.7.3.8.2 Limitations in conceptualising disease experience affected by individual and environmental characteristics

As described earlier, there have been numerous studies using indices to measure the negative impact of SS patients' oral health on their QoL. Attempts to measure characteristics of the individual include collecting data such as age, race, and sex (Jenkins, 2010). While attempts to consider characteristics of the environment include collecting data on social support, the characteristics of the individual and the environment are difficult to measure. This is because each individual is has a personality that may be affected by various life experiences or stages. In addition, the impact of characteristics of the individual and the environment on each level of the model (such as symptoms or overall health perception) cannot be measured in existing measures. For example, individual ratings of severity of various domains of disease impact are based on the assumption that every participant interprets the meaning of each statement in the questionnaires uniformly. However, due to the different characteristics of the individual and the environment, this may not be the case. Baker et

al (2007) considered the Wilson and Cleary model to be helpful in identifying general rules for a particular patient population, but less effective in exploring the characteristics of the individual and the environment because these two levels are not quantifiable using QoL measures. Therefore, employing multi-method approaches incorporating qualitative methods or diary designs has been advised to be necessary to understand the complexities of individuals' experiences within their environment (Baker et al., 2007).

2.3.7.3.8.3 Limitations in conceptualising changes in patient experiences with time

In chronic diseases, perceived health status changes with time (Dolan et al., 1998). This variable is not considered in a single-visit QoL questionnaire. Positive and negative changes may cancel each other out, giving the impression of no change. For example, even though the XI (Thomson et al., 1999b) seeks to understand patient experience over time with the heading "The following statements refer to your experiences of mouth dryness during the last 4 weeks", a patient may circle 'occasionally' for the item: 'My mouth feels dry' if he or she experienced 'always' half the time, and 'never' the other half of the time. This averaging out of the overall experience may result in the lost of accurate capturing of changes with time. To address this issue, there have been attempts to compare "before" and "after" QoL measures (Locker, 1998). However, the practicality of comparing such scores has been criticised, suggesting that a change in score intrinsically has no clinical meaning (Allen, 2003). HRQoL and OHRQoL measures capture patient perspectives in a snapshot, loosing the diversity that occurs daily depending on changes that can vary with time and experiences. These changes can be modified by psychological phenomena (characteristics of the individual) such as adaptation, coping, and optimism (Allison et al., 1997).

2.3.7.3.8.4 Limitations in conceptualising the effect of treatment

There are gaps in commonly used HRQoL conceptual models in considering the influence of management of therapeutic regimens and self-management on QoL (Bakas et al., 2012). These models focus on the effect of symptoms, and not on the clinical or self-management of the disease. In the absence of curative treatment for dry mouth, long-term coping and the management of the condition is required. Often, such coping mechanisms differ with each individual, depending on various mediators.

Qualitative methods used in adjunct to the conceptual model will provide insight into some of these limitations.

2.3.8 Conclusion

The literature clearly demonstrates measurable negative impact of dry mouth and SS on the QoL of patients. However, there is more to patient perspectives than HRQoL and OHRQoL measures can capture. Despite being a good conceptualisation of the various levels that can affect HRQoL, the Wilson and Cleary model revised by Ferrans et al (2005) is still unable to fully describe the vastness and complexity of the patient experience of dry mouth and SS. This suggests looking at a different paradigm of research, bringing us to the next chapter on qualitative research.

2.4 Qualitative research in dry mouth and SS

2.4.1 Introduction

This chapter will discuss qualitative research, with a focus on dry mouth and SS. To begin, the paradigms of research (positivist and interpretivist) will be established to set the background that informs the two main research methods: quantitative and qualitative. Next, there will be a comparison between the two methods. This will be followed by a discussion of the (limited) existing research on dry mouth and SS that is based on the interpretivist paradigm. The gaps in the current knowledge of the experience of dry mouth in SS patients will be identified, and this will lead to the research question.

2.4.2 The paradigms of research

A paradigm (or research philosophy) shapes how the researcher views the world. The two main research methods in health research (quantitative and qualitative) are based on the positivist and interpretive paradigms respectively (Broom and Willis, 2007). These two paradigms are on two ends of the same continuum that share the goal of understanding the world in which we live (Haase and Myers, 1988). Each paradigm has a patterned set of assumptions concerning: *ontology*, or the reality that the researcher investigates; *epistemology*, or the relationship between reality and the researcher; and *methodology*, or the techniques used by the researcher to investigate that reality (Guba, 1990; Lincoln et al., 2011).

The research paradigm will guide the type of research question being asked, leading to the selection of research methods that will determine the nature of knowledge produced. This process is illustrated in Figure 7 (Maxwell, 2012).

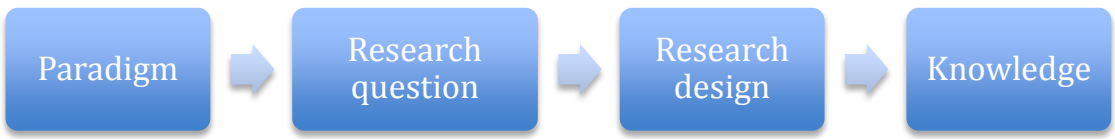


Figure 7. Research process based on paradigm (based on Maxwell 2012)

2.4.2.1 The positivist paradigm behind quantitative research

Quantitative research is based on the positivist paradigm. Its ontological standpoint is that there is a single, objective reality set in stone that exists independent of human perception. This reality is thought to be measurable without bias using standard instruments. The epistemological standpoint is that knowledge is objective and the investigator and the investigated are independent entities. The researcher is seen as a neutral recorder, and different researchers using the same instruments should reach the same conclusions. Hence, the researcher is capable of studying a phenomenon without being influenced or influencing it; "inquiry takes place as through a one way mirror" (Guba and Lincoln, 1994). Quantitative tools such as randomised control trials discover a universal truth that holds true so long as specific conditions hold. Large sample sizes (relative to those used in qualitative research) allow statistical methods to ensure that they are representative (Carey, 1993). The nature of knowledge studied involves the researcher testing a hypothesis with aims to prove or disprove it.

2.4.2.2 The interpretivist paradigm behind qualitative research

By contrast, qualitative research is based on the interpretivist paradigm. The ontological standpoint is that reality is interpreted and actively constructed. It cannot be measured directly; rather, it can only be perceived by people, each of whom views it through the lens of his or her prior experience, knowledge, and expectations. That lens affects what people see and how they interpret what they find. What we know, then, is not objective; it is always filtered through people, and it is always subjective (Rubin and Rubin, 2011). The epistemological standpoint is that knowledge is subjective and cannot be accessed independent of our minds. Common qualitative tools include diaries and interviews. Their use aims to discover and understand a phenomenon that is embedded in a complex and changing reality. The nature of knowledge deduced involves the researcher gathering data to understand in depth and establish patterns drawing on theory. The interpretivist and positivist paradigms are summarised in Table 6 (Carson et al., 2001).

Table 6. Summary of the positivist and interpretivist paradigms (after Carson et al., 2001)

	Positivist	**Interpretivist**
Research Goal	Test theories, discover general principles	Describe and understand complex situations
Nature of knowledge (epistemology)	Objective, universal truth	Subjective
Nature of reality (ontology)	Set in stone	Interpreted and actively constructed
Research style	Deductive Broad theories and systematically test implications	Inductive Build explanations from ground up, based on what is discovered
Research methods	Quantitative Measure physical, biological, or chemical phenomena in replicable ways Eg. Questionnaires Randomised controlled trials	Qualitative Understand phenomena Eg. Interviews Diaries
Researcher role	Neutral and does affect what is measured	Data-gathering instrument whose skills in listening, observing, and understanding are crucial

2.4.3 Quantitative and qualitative methods

Owing to the differing paradigms, there are some differences between quantitative and qualitative methods. These two methods are ends of a spectrum, with many intermediate points within. A detailed discussion of the range of research methods (which would constitute a significantly different discussion) is not the intent of this research. However, by establishing the diametric ends of the spectrum, background knowledge is provided to justify the interpretivist approach (behind qualitative methods) chosen for the purposes of answering the current research question. The

contrasts between the quantitative and qualitative approaches are summarised in Table 7 (Bullock, 2010).

Table 7: Summary of differences between quantitative versus qualitative research methods (after Bullock, 2010)

Quantitative	**Qualitative**
Theory testing	Theory emergent
Researcher's point of view	Participant's point of view
Researcher distant	Researcher close
Structured	Unstructured
Numbers-based data	Words-based data
Generalisation	Conceptual understanding
Hard, reliable data	Rich, deep data
Macro-focused	Micro-focused
Concerned with behaviour	Concerned with meaning
Artificial settings	Natural settings

Table 7 sets out to contrast the dimensions of the quantitative and qualitative approaches. However, it should provide only an outline, because the descriptors are endpoints, and the processes related to these need to be further explored (in the next chapter on methods) for complete understanding.

Despite their differences, qualitative and quantitative research should be seen as complementary rather than oppositional approaches to research (Edmunds and Brown, 2012). Quantitative and qualitative methods are part of the spectrum of research approaches, with specific tools that are selected based on the research objective (Casebeer and Verhoef, 1997). Knowledge has multiple dimensions, and quantitative and qualitative methodologies are different strategies used to explore its different aspects. Considering the impact of dry mouth on SS patients, quantitative methods can

be used to measure HRQoL and OHRQoL (as described in the previous chapter), or qualitative methods can be applied to understand the subjective patient experience. Some argue that quantification using a positivist paradigm is essential for an objective and rigorous investigation. Others argue that no description could be complete without a qualitative understanding of the subjective meanings of the human beings involved (Glassner and Moreno, 1989). Purists would argue that a pluralist approach adopting both methods is not viable, given the competing epistemological underpinnings. However, others argue that there is value in recognising the complementarity between approaches and accepting the strengths and limitations of each (Bullock, 2010). Neither paradigm or method is superior; the research philosophy which is adopted is related to the research question and will guide the appropriate methodology for the problem under investigation.

2.4.4 Qualitative research in dry mouth and SS

This section will consider the existing qualitative research in dry mouth and SS. The two conditions have been examined separately.

There have been few qualitative studies on the impact of dry mouth on individuals. In the dental literature, there have been studies published by Rydholm and Strang (2002), Folke (2009), and Owens et al (2014). In a Swedish study by Rydholm and Strang (2002), semi-structured interviews conducted with 16 patients with advanced malignancies and associated xerostomia identified four main categories: (1) subjective discomfort; (2) loss of function; (3) increased infection; and (4) psychosocial effects, including shame, increased feelings of being a patient rather than a person, and a tendency to avoid social contact, resulting in loneliness (Rydholm and Strang, 2002). Xerostomia was found to have a negative impact even in the palliative stages of the illness course, emphasising its relative significance. Folke (2009) interviewed 15 people with dry mouth, and found that their main concern of dry mouth was its devastating and debilitating impact on multiple domains of well-being. They concluded that dry mouth is not a trivial condition for those suffering it, and there is a need to improve the understanding (and therefore compassion) of health care professionals providing treatment for people afflicted by it. These findings revealed the complexities of xerostomia and broadened the focus from the oral cavity to the individual as a whole (Folke, 2009). These insights into the physical and psychosocial consequences of

xerostomia complement the quantitative findings that dry mouth negatively affects the QoL of its sufferers (as discussed in the previous chapter on QoL).

In a more recent UK study, Owens et al (2014) explored people's subjective experiences of dry mouth in the context of viewing the condition using the social model of disability and the concept of impairment. The findings determined that dry mouth has private and public dimensions, whereby impairment ends, and disability begins (Owens et al., 2014). Coincidentally, 6 of the 18 participants had SS, while the rest suffered from dry mouth with no additional co-morbid medical conditions. This is noteworthy because dry mouth is usually related to systemic conditions or the polypharmacy secondary to treating them.

There have been few qualitative studies on the general effects of SS on sufferers. Mengshoal et al (2013) in their Norwegian study interviewed 5 women and 4 men with SS and found that extreme fatigue resulted in patients having to scale down their lives, or even put them on hold. The findings showed the debilitating effect that fatigue from SS can have on its sufferers (Mengshoel et al., 2013). A Swedish study by Andersson et al (2001) examined the situational patterns in coping with SS in 69 patients using a questionnaire with written descriptions. The findings indicated that symptoms of dry mouth, fatigue, eye discomfort, and sleep disturbances were particularly stressful for those patients (Andersson et al., 2001). Schoofs (2001) used telephone interviews to explore the lived experiences of 10 women with SS, and found that they lived in hope of a future cure, are reliant on help from others, are hurting from physical and emotional suffering, and their social lives are hindered by the condition (Schoofs, 2001). These studies provide insight into the adverse effects that SS has on the QoL of its sufferers.

2.4.5 The gaps in knowledge

Gaps in the knowledge will set the argument for the selection of the appropriate methodology for the problem under investigation. So far, little qualitative research has been done on dry mouth and SS, and none purported to study the impact of the two conditions together. The primary manifestation of SS is dry mouth, and so there is a definite need to examine dry mouth in the context of SS because this is the daily reality for people with both conditions. The qualitative research thus far has established

(separately) that dry mouth and SS both have profound impact on the personal and social lives of sufferers. Continuation of research from the interpretivist approach should provide a rich insight into the disease experience.

Traditionally, the dominant paradigm in health sciences research tends towards positivist methods. The majority of the existing literature on the impact of dry mouth and SS on QoL is based on quantitative research (as described in the chapter on QoL). Using HRQoL and OHRQoL measures, it has been established that dry mouth negatively affects the well-being of people of all ages, from different countries, whether medically fit or unwell (Ikebe et al., 2007; Locker, 2003a; Thomson et al., 2006c). Similarly, SS has been shown to adversely affect QoL in a wide range of physical, emotional, and psychological domains in different patient groups (Belenguer et al., 2004; Segal et al., 2009; Thomas et al., 1998). Dry mouth has also been shown to be detrimental to the QoL of SS patients (Enger et al., 2011; Meijer et al., 2009; Stewart et al., 2008). Because neither dry mouth nor SS can be treated, the collective conclusion from these studies is to improve the understanding of the (psychosocial, physical, and emotional) impact of dry mouth and SS on people in order to aid management and long-term coping of the conditions.

Conceptual models are useful in understanding clinical variables that affect patient perspectives of HRQoL. The Wilson and Cleary model, when applied to dry mouth, reveals complex relationships between key clinical and non-clinical variables. However, quantitative measures are unable to describe the influence of characteristics of the individual and the environment on health (Baker et al., 2007). No HRQoL conceptual model has been applied to reveal the complex relationships behind the adverse impact of SS on QoL. There has not been any qualitative work undertaken to examine dry mouth amid the range of SS systemic manifestations. No research has yet provided a qualitative perspective to the revised Wilson and Cleary model by Ferrans et al (2005). There are gaps in the knowledge that have yet to be approached from an interpretivist paradigm.

2.4.6 Aim and objectives, and research question

The current research aims to understand the lived experience of dry mouth for SS patients with consideration of the characteristics of the individuals and the environment that they are in. The view adopted is depicted in Figure 8 below.

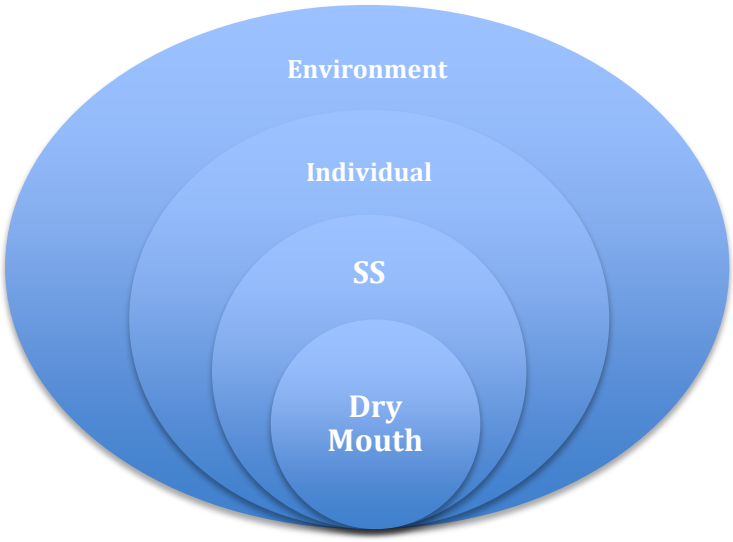


Figure 8. Research view based on the revised Wilson and Cleary model by Ferrans et al (2005)

Traditionally, the positivist paradigm has been adopted for healthcare research. By investigating the illness from an interpretivist point of view, the knowledge gained can complement (and perhaps contrast with) the revised Wilson and Cleary model by Ferrans et al (2005), especially in the levels 'characteristics of the individual' and 'characteristics of the environment'. The employment of qualitative methods based on the interpretive paradigm seeks to address the above gaps in knowledge.

The objective of this qualitative research project is to specifically explore how patients perceive SS to affect their daily lives, with a focus on dry mouth. This is to understand the dry mouth experience amid SS (a chronic systemic autoimmune disease) and to examine the individuals as a whole within their environment, thereby providing a valuable reference for health care professionals to promote better understanding of, and a greater empathy for individuals afflicted by dry mouth and SS. This research purports to address the limitations of the revised Wilson and Cleary model by Ferrans et al (2005) in conceptualising disease experience affected by: (1) individual and

environmental characteristics; (2) changes in time; and (3) treatment (as discussed in the chapter on QoL).

The research question is therefore what is the ongoing lived experience of dry mouth for SS patients, and how does each individual cope with dry mouth amid a spectrum of symptoms (with consideration to individual and environmental characteristics)? The next chapter will describe the methods employed to answer the research question.

Chapter 3 Methodology

3.1 Introduction

This chapter will discuss the methodology adopted for the current research. First, there will be an overview of the foundation work, including the: (1) Maori consultation; (2) ethical approval; and (3) grant application. Next, there will be a description of the participant recruitment process (for both the pilot and main studies). There will then be an explanation of the data collection process (using diaries, interviews and the SXI). Finally, there will be consideration of the data analysis, that has a main qualitative component (thematic content analysis) and a minor quantitative component (statistical analysis of the SXI scores).

3.2 Maori consultation

In May 2012, the Ngāi Tahu Research Consultation Committee endorsed the proposed research project (Appendix 3), considering the research to be of importance to Maori health. The Committee strongly urged that ethnicity data be collected as part of the research project and recommended the dissemination of research findings to relevant Maori health organisations, such as the National Maori Organisation for Dental Health (Te Ao Marama) and to Professor John Broughton, who is involved in Maori Dental Health research at the University of Otago.

3.3 Ethical approval

In May 2012, the University of Otago Human Ethics Committee (UOHEC) granted Category A approval for the research project (Appendix 4).

In December 2012, the UOHEC approved the following amendments in research procedure: (1) interviews with participants were to be held in the Dental School, rather than in their homes; (2) the estimated length of the interview was reduced from 1.5-2 hours to 1 hour; (3) contact with participants was to be made by phone call or email rather than home visits; and (4) a research email address was created (Appendix 5). These amendments were requested based on participants' preferences and observations that were made during the pilot study.

In April 2013, the UOHEC approved amendments to the recruitment methodology

(extending the sampling location from Dunedin alone to include Auckland), and allowed for reimbursement for participation (Appendix 6). These amendments were requested because (1) a potential participant living in Auckland voluntarily approached primary supervisor (Professor Anita Nolan) to express interest in the research project, and (2) the research grant application had been successful and allowed for participant reimbursement (in the form of supermarket vouchers).

3.4 NZDA/MOH grant

In August 2012, the New Zealand Dental Association/Ministry of Health (NZDA MOH) Oral Health Research Fund Assessment Panel awarded $6,256 for the research project, subject to ethics approval being obtained, and on the condition that the Assessment Panel received a satisfactory annual progress report by June 2013 (Appendix 7).

3.5 Data collection

This section will cover the: (1) participant recruitment process; (2) diary methods; (3) interview methods; (4) sample size; and (5) the SXI.

3.5.1 The recruitment of participants

The sampling purpose was to identify a group of SS patients with dry mouth who had the knowledge to inform the research question. The sample of interested, suitable, and articulate participants were selected on a volunteer basis from a pool of patients who had consulted primary supervisor Professor Anita Nolan (AN), an oral medicine specialist who was then at the University of Otago. The inclusion criterion was a positive diagnosis of SS (primary/secondary) according to the revised international classification criteria (AECG) (Baldini et al., 2012a). The AECG was chosen for this study because it is well-cited in the literature, and has high specificity and sensitivity. The exclusion criterion was the inability to commit to the data collection (that consisted of a 1-month diary entry and an interview). Primary researcher Joanna Ngo (JN) explained details of the research to interested participants face to face whenever possible, at their convenience, either in AN's consultation room, or in a Dunedin café. For interested participants living out of Dunedin and without upcoming appointments, contact was initiated through phone and post. Accordingly, information sheets

(Appendix 8) were given and consents (Appendix 9) were obtained either face to face or by post.

3.5.2 The diary

Qualitative diary research methods aim to capture the ongoing daily experiences of participants in their natural habitat, while reducing the bias of information from retrospection, or the loss of information through time lapse (Patterson, 2005). Participants were asked to maintain an introspective, flexible time-based diary to record daily events and reflections that they perceived to be related to dry mouth and SS. This captured prospectively the participants' thought processes in a detailed and individual level, along with any fluctuations that occurred with time (Bolger et al., 2003).

A pilot study with 2 participants was carried out to test the diary (and interview) processes. Two different diary methods were tested: (1) a guided with parameters approach; and (2) an unguided, minimal instruction approach. Each participant was asked to maintain a month-long diary (and undergo an interview afterwards). The pilot study identified that the guided diary entry method with open response format (Appendix 10) allowed participants to express thoughts, reflections, reports, intimate details, or explorations on what was important to them, and to structure entries as they felt appropriate (Elliott, 1997). Moreover, 1 month proved to be an appropriate length of time. The comparison of both pilot study diary methods is summarised in Table 8.

Table 8: Comparison of the diary methods tested in the pilot study

	Pilot 1 (guided)	**Pilot 2 (unguided)**
Entry design	Written/email	Free style
Length of time	1 month	Up to the participant
Number of entries	Once a day	Any time an experience associated with dry mouth/SS strikes the participant
Amount of guidance on content	Some parameters - Sleep pattern - Social (meals) - Relationship with others - Occupation - Dental problems - Positive and negative emotions about dry mouth	Free structure

During that month, the primary researcher (JN) contacted each participant weekly by the participants' preferred mode of contact (phone/email/directly) to discuss the diary entries, and to initiate (and maintain) good rapport. The level of rapport improved the quality of the interviews that were held later. Diary entries were in a written/email format, and were later transcribed into a Microsoft Word document by JN (to be analysed). A topic guide (list of core questions) for each interview was derived from the preceding diary entry (as discussed in the next section).

3.5.3 Interviews

Interviews encouraged conversational interaction, with open-ended and non-leading questions allowing participants to freely narrate their thoughts (on the effects of dry mouth and SS on their daily lives) (Durham et al., 2011). Semi-structured interviews were conducted with a set of flexible open-ended questions that defined the area to be explored, from which the interviewer or participant then diverged to pursue an idea in more depth (Britten, 1995). This allowed new perspectives that were not anticipated at the outset of research, and covered a range of reflection of events that occurred out of the diary period. The in-depth, semi-structured interviews were conducted in a neutral environment, usually a meeting room in the dental school. The interview time ranged from 25 minutes to 1 hour, depending on the amount of information each participant had to share. Interview technique training was provided by supervisor Dr Shelagh Ferguson (SF), who has long-standing expertise in interpretive and ethnographic research (including diary and interview methods).

3.5.3.1 Pilot interviews

The two pilot study interviews were conducted by SF and observed by JN. Prior to each interview, SF and JN discussed the transcribed diary records and produced a list of interview topics. The process of the pilot interviews is illustrated in Figure 9.

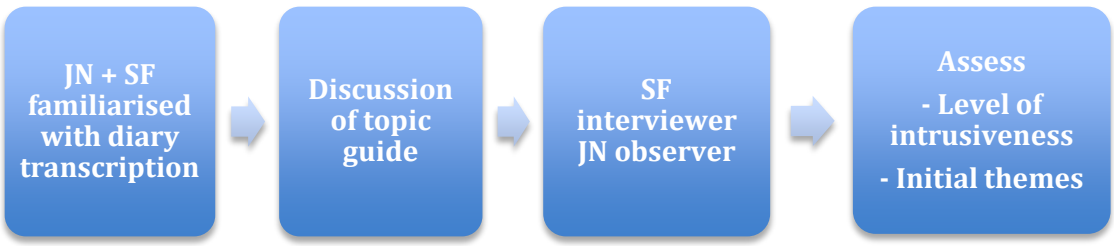


Figure 9. The pilot interviews process

3.5.3.2 Main study interviews process

Qualitative research is about subjectivity and complexity (depth of analysis); it does not seek to count or reduce, but to represent the rich experiences that reflect consistent and parallel patterns (Broom and Willis, 2007). The topic guide evolved with each interview to acquire more structure, narrowing down into four main themes as data collection proceeded. This is illustrated in Table 9.

Table 9: Evolution of topic guides

Topic Guide 1	**Topic Guide 10**
Introspection of life before and after SS Trauma of diagnosis? Impact of lack of sleep from dry mouth Proactive/reactive to diagnosis Is the general disease pattern better/worse than that reflected in the one-month diary? Social impacts • Social interaction • Embarrassment Dietary impacts • Alcohol consumption • Coffee consumption Family • Support/interaction • Response to diagnosis • Openess of discussions Healthcare professionals • How was the interaction? Coping strategies	Journey to diagnosis • How was the route? • Initial feelings and symptoms? Interaction with healthcare professionals • Positive or negative? • Active/passive patient role? Disease impact spectrum • Dry mouth and communication, sleep, work? • Relative to other symptoms (dry eyes, fatigue, sore joints)? Coping mechanisms • Support from friends/family? • Awareness and adjustments • Personalised remedies • Lifestyle • Mindset

The types of questions asked were based on: (1) behaviour or experience; (2) opinion or value; (3) feeling; (4) knowledge; (5) sensory experience; and (6) background details (Patton, 1987). Table 10 illustrates these questions types with examples.

Table 10: Interview question types with examples (after Patton 1987)

Question type	**Example**
Behaviour or experience	Are there some foods you have to avoid?
Opinion or belief	Would you say that's (dry mouth at night) the worst aspect of it impacting upon your life?
Feelings	How did you feel when they first told you that you had SS?
Knowledge	Did you know what it (SS) was?
Sensory	You mentioned food is sticking to your mouth… do you adjust to that?
Background or demographic	Do you think being a nurse affected how you got to your diagnosis…

The interviews were audio-taped with a high quality Olympus digital voice recorder that produced recordings with minimal background noise. Each digital audio file was transferred into the computer and played back with the VLC 2.0.6. open source cross-platform multimedia player.

3.5.4 Sample size

The sample size was fluid, and depended on data saturation, whereby there was redundancy of themes (no new themes discovered) (Lincoln, 1985). The next 8 interviews were conducted independently by JN, with SF available for feedback on interviewing methods. At the 7th participant, data saturation was observed. After this, an 8th participant was recruited to confirm that the redundancy of themes had been reached. The data from the pilot study were found to be rich, and so they were included into the complete study data collection, bringing the total sample size to 10. All of the participants were middle-aged or older (corresponding to the common age group in which SS is diagnosed) with an age range from 46 to 86, and a mean age of 64. All of

the participants were community-dwelling; 6 held regular jobs, and 4 were retired people. The proportion of females was 9:1, coincidentally, reflecting the usual female-to-male ratio in SS prevalence (Fox, 2005).

3.5.5 Transcription

Transcription was very time consuming, with each hour's worth of interview taking up to 6-7 hours to transcribe. A total of 160 hours were spent on transcribing all 10 diaries and interviews. Each audio file was played back and manually transcribed by JN using segmentation and verification as soon as possible after the interview, in order to optimise impressions of non-verbal cues, and the benefits of data familiarisation (Braun and Clarke, 2006). Transcribed data included punctuation, expressions, laughter, and consistent speaker identification to reduce discrepancies in data transfer, allowing, therefore, for accurate recording of non-speech interactions during the interviews. Transcripts of all the diaries and interviews are contained in the CD at the end of this thesis (Appendix 12).

3.5.6 The Shortened Xerostomia Inventory (SXI)

After each interview, each participant was asked to complete an SXI (Appendix 1). This has 5 items aimed to capture the experience of xerostomia during the past 4 weeks. The five possible responses for each item range from 'never' to 'very often', and aim to capture the severity of the condition. Supplementing the SXI, there is a global item, "How often does your mouth feel dry?" with four possible responses ('never', 'occasionally', 'frequently', and 'always'). This complemented the diary and interview data, and provided corroborative quantitative data. This quantitative measure of the xerostomia experience allowed for comparison with existing data.

3.6 Data analysis

This section describes the thematic content analysis (that was applied to the data collected from the diaries and interview) and the statistical analysis that was performed on the SXI scores. The amount of data that was gathered was extensive, as summarised in Table 11.

Table 11: Summary of transcribed data

	Participant	Number of pages		Number of words		Interview time in
		Diary	Interview	Diary	Interview	minutes.seconds
1	Margaret	6	21	2113	8028	50.42
2	Tina	8	12	3034	6915	42.50
3	Eleanor	6	10	1789	4145	25.17
4	Frieda	2	13	1263	4785	35.01
5	Mike	7	11	2366	6102	33.24
6	Anne	3	25	859	10750	58.52
7	Joy	5	11	1707	3884	26.12
8	PM	4	16	976	7682	52.58
9	Jo	6	26	2476	10531	60.38
10	Tracy	3	20	504	7065	42.00
	Total	215		86,974		427.9

3.6.1 Thematic content analysis

Owing to the exploratory nature of the research, thematic content analysis was applied (Braun and Clarke, 2006). The method of emergent coding allowed the themes to arise naturalistically from the data without bias or elicitation. The themes were produced inductively and not restricted to pre-determined codes (Creswell, 2003), hence providing a more accurate interpretation of the data. The process also involved the following steps: (1) familiarisation with the data: (transcription, reading, noting initial ideas); (2) generating initial codes by systematic coding of data (inter-researcher peer coding determined the reliability); (3) searching for themes, and collating data into potential themes; (4) reviewing themes by checking them in relation to coded extracts; (5) defining and naming themes, thus generating clear definitions of each theme; and (6) reporting on findings, by relating the analysis to the research question and literature, thus producing a scholarly report of the analysis. Analysis of the data provided an evidence-related, personal-experience-based pattern of themes that were significant to the dry mouth experience of these SS sufferers. When applicable, some of the themes were considered in relation to the Wilson and Cleary model revised by

Ferrans et al (2005). This model was chosen for the current research because it is applicable to any healthcare discipline in the real-world settings, and can be used in future as a common reference for comparison across studies. Moreover, it and suitably depicts a holistic view of HRQoL (Bakas et al., 2012). There will be an emphasis on the levels 'characteristics of the individual' and 'characteristics of the environment' in the model. Therefore, each participant's individual and environmental characteristics were summarised in a diagram to get a sense of who they were (Appendix 11).

3.6.2 SXI analysis

SPSS version 22 was used to compute the SXI scores and then to examine the gradient of mean SXI scores across the response categories of the standard dry mouth question. Analysis of variance was used to determine the statistical significance of the observed differences.

3.7 Conclusion

The methodological processes for the current research have been planned to answer the research question. Data collected from diaries, interviews, and SXI scores were used to understand the lived experience of dry mouth in these SS patients. The next chapter will discuss the findings from the SXI scores. This will be followed by the presentation of the results from the thematic content analysis.

Chapter 4 The SXI scores

4.1 Introduction

The SXI has been tested (and validated) in a number of diverse samples from Australia, The Netherlands, Japan, and New Zealand (Thomson et al., 2011). This chapter will compare the SXI scores from the above older populations and those of the current research participants. First, there will be a brief description of the older population samples that the SXI has previously been tested on, and then of the current research sample. Next, there will be presentation of SXI scores (mean and range) and associated data from the current sample, and these will be compared with existing data to demonstrate the more severe experience of dry mouth among SS sufferers.

4.2 Description of the samples

The sample sizes used in the quantitative study to validate the SXI ranged from 50 (The Netherlands) to 637 (South Australia), with broadly similar age ranges (older adults). There were two institutionalised samples (Melbourne and The Netherlands) and four community-dwelling samples. The proportion of females ranged from about one-third (in the NZ community sample) to almost three-quarters (in the Melbourne sample) (Thomson et al., 2011).

Table 12 is a summary of the description of the samples used to validate the SXI and the current research (Thomson et al., 2011).

Table 12: Summary of the descriptions of samples used to validate the SXI and the current research (after Thomson et al., 2011)

	South Australia	The Netherlands	Melbourne Australia	Osaka, Japan	NZ geriatric sample	NZ community sample	NZ SS Sample
Number of participants	637	50	245	401	167	86	10
Number of females (%)	291 (46)	30 (60)	169 (70)	206 (51)	97 (58)	28 (33)	9 (90)
Mean age	70	78	83	66	82	72	64
Age range	60-95	53-98	51-103	60-84	65-98	50-90	46-86
Nature of sample	Community	Institution	Institution	Community	Institution	Community	Community

4.3 Findings

SXI scores ranged from 17 to 24, with a mean score of 20.9. There were no apparent differences by age, with no significant difference in mean SXI score between those aged younger than 63 and those older (20.4 and 21.4 respectively; P = 0.56; ANOVA). The scatter plot (Figure 10) of SXI scores by age shows no apparent association.

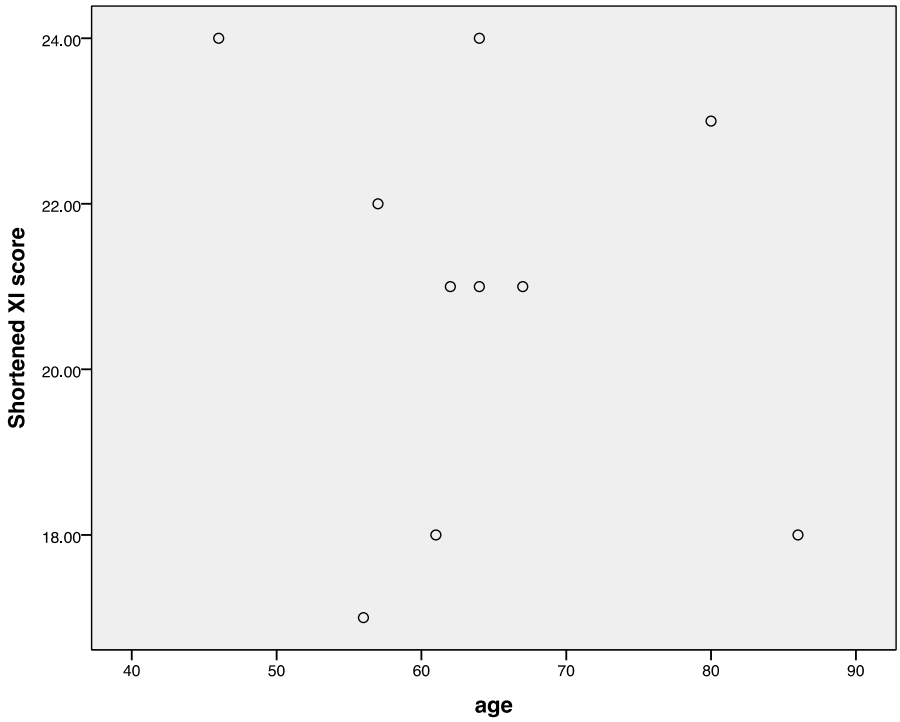


Figure 10: Scatter plot of participants' SXI score by age

4.4 Comparison of SXI scores of samples (older population) used to validate the SXI with the current research sample

In the SXI, the higher the score indicates a worse experiential and behavioural aspects of dry mouth. Comparison data are presented in Table 13.

Table 13: Summary data on the SXI scores for the samples used to validate the SXI and the current research sample (after Thomson et al,. 2001)

Sample	Mean SXI score
The Netherlands	7.8 (7.1-8.5)
South Australia	7.6 (7.4-7.8)
Melbourne, Australia	8.1 (7.8-8.4)
Osaka, Japan	8.7 (8.5-8.9)
NZ community sample	9.8 (9.1-0.5)
NZ geriatric sample	8.6 (8.2-9.0)
NZ SS sample (current study)	20.9 (17.0-24.0)

Table 13 shows a remarkable contrast; the mean SXI score for the participants of this research was more than twice that of any of the other samples used in that validation study. This is significant because xerostomia is already known to be more severe and of higher prevalence estimates in older people. In the study of 1205 dentate older South Australians to describe the incidence of xerostomia over a 6-year period, both xerostomia prevalence and severity were found to increase with age (as measured with the XI) (Thomson et al., 2006a) This illustrates the severity of dry mouth experienced by these SS patients. The analysis of the SXI scores has provided a quantifiable description of the dry mouth experiences of these participants, complementing the descriptions made in the themes.

4.5 Conclusion

The SXI was used in this research as a description of symptom manifestation (xerostomia) for each participant. Compared to a range of estimates from samples of older people, the current research participants had more than twice their mean score. This enabled a comparative, tangible understanding of the extent of dry mouth experienced by the research participants, further emphasising the finding that dry mouth is not at all a trivial symptom in the daily lives of this sample of SS sufferers.

Chapter 5 Results and commentary

5.1 Introduction

This chapter is a description of the results from the thematic content analysis of the rich data collected from the diaries and interviews. The four main themes derived were: (1) the journey to diagnosis (JTD); (2) interactions with healthcare professionals (IHCPs); (3) the disease impact spectrum (DIS); and (4) the positive (SS) coping process (PCP). These were important aspects of the participants' dry mouth and SS experiences as illustrated in Figure 11.

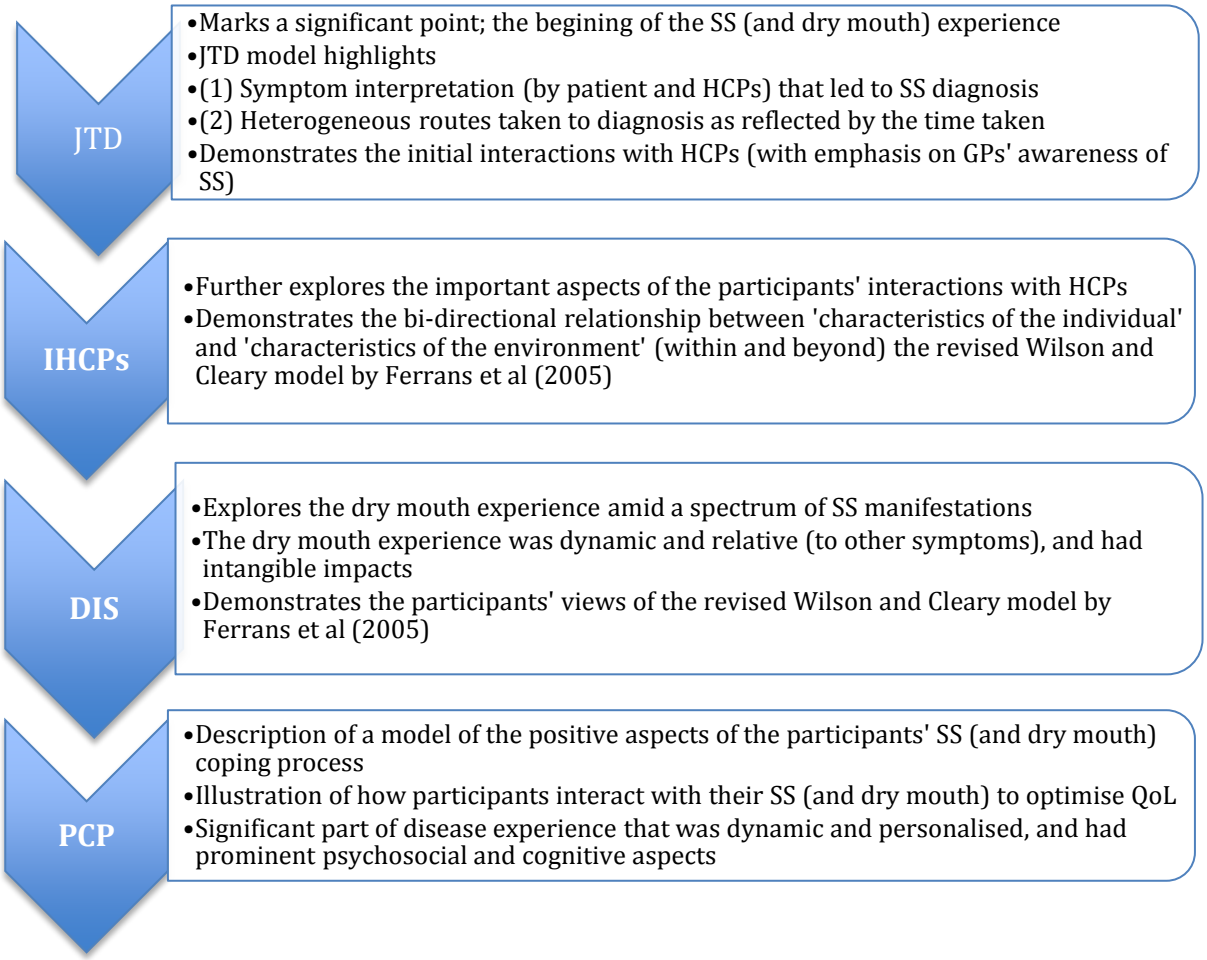


Figure 11. The mapping of the themes onto the participants' dry mouth (and SS) experience

Owing to the qualitative nature of the current research, each of these themes will be explained using empirical evidence from the transcribed data in a discursive approach, with a commentary that will include references to existing literature.

5.2 The journey to diagnosis

This theme was initiated by one of the pilot study participants, Tina, who began her diary by describing her SS diagnosis experience. Similarly, 5 other participants included such discussions at the beginning of their diary entries or interviews. These introspections described a significant part of their disease process. Thereby, the rest of the participants' diagnoses experiences were explored with an interview question such as, "Can you tell me when you got diagnosed with SS?" The heterogeneity of the journeys among the 10 participants was remarkable. The pattern observed across the data set included: (1) symptom interpretation by the patient; (2) symptom interpretation by the HCP; and (3) heterogeneous medical routes that led to diagnosis. The participants' JTD was an important process that did overlap with some of the levels in the revised Wilson and Cleary model by Ferrans et al (2005), such as the 'characteristics of the individual' to interpret 'symptoms' that were linked with their effect on 'functional status', and the HCPs' (who are part of the 'characteristics of the environment') interpretation of 'symptoms'. However, it would be a compromise to map these patients' experiences onto the model because of the difference in what is being conceptualised. The model conceptualises QoL in respect of the clinical variables, while the JTD is the process of getting to SS diagnosis, which the nature itself had major implications on QoL. Therefore, Figure 12 was derived from the empirical findings to represent the richness of the participants' experiences.

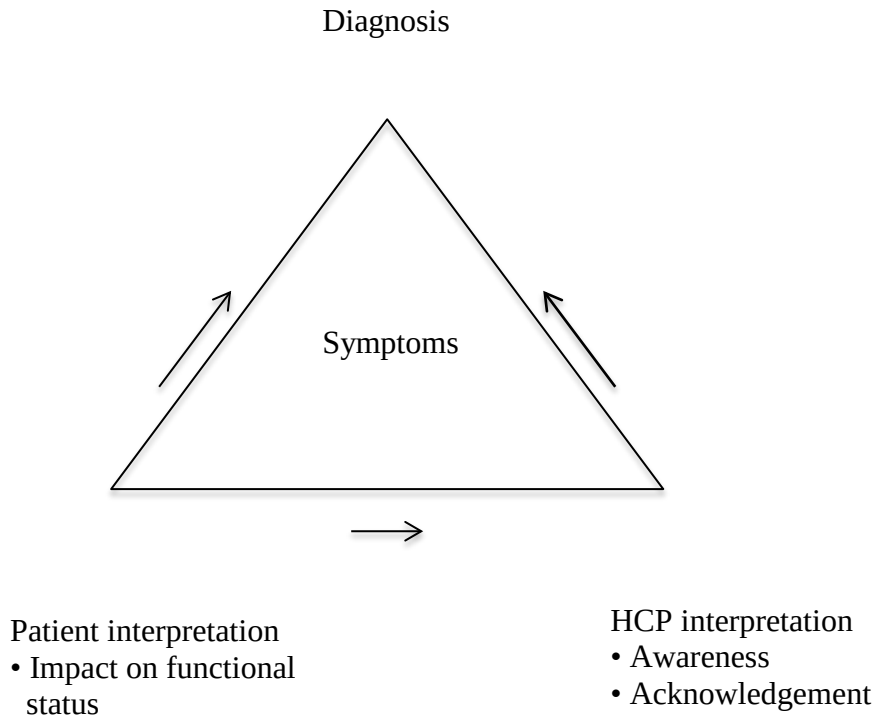


Figure 12: The journey to diagnosis

The triangle symbolises the relationship among symptom, patient, and HCP in the journey to diagnosis. In the centre of the triangle are the different symptoms that indicate an abnormal state (Wilson and Cleary, 1995). The journey is based on the symptom interpretation by the patient that leads to seeking HCP advice. This allows the symptoms to be interpreted by HCPs. The journey can take different routes (direct/indirect, as indicated by time taken), but ultimately leads to SS diagnosis (the apex of the triangle).

5.2.1 Symptom interpretation

Wilson and Cleary (1995) defined symptom as the patient's perception of an abnormal state (Wilson and Cleary, 1995). The definition focused on the patient's interpretation of a symptom. However, the empirical findings from the current research demonstrated the importance of symptom interpretation by the HCPs because it was shown to play an integral role in the journey to diagnosis.

From the participants' perspectives, symptom interpretation (making sense of it) was intertwined with impact on functional status (such as work or eating). This then led participants to seek medical care, thereby opening the interpretation of the symptoms to HCPs. HCPs' symptom interpretation depended on their awareness and acknowledgement (of the symptoms). When these symptoms were interpreted (by HCPs) to be SS-linked, diagnosis was made possible.

5.2.1.1 Symptom interpretation by patients (participants)

The initial physical symptoms from the sample of informants were hugely variable, with a different combination for each participant, ranging from none (with an incidental finding of SGH observed by a dental hygienist), intolerance to different foods, dry mouth, malar rash, bowel problems, swollen glands, pneumonia, sore joints, to general fatigue. These symptoms were experienced in different combinations by each participant. They were then interpreted differently, in close relation to their effect on participants' functional status. This section will provide findings to support this.

Jo is a 61–year-old European lady who is fit and strong. She works on the farm and is responsible for physically demanding chores such as irrigation. The process of how she interpreted her initial symptom of joint pain is outlined below.

Jo: *"During last winter I had trouble getting out of bed and getting clothes on and had no strength in my hands in the mornings, so I went back (to GP) as I was getting really concerned about being able to cope with irrigating as it is quite physical and I was in pain getting on the bike let alone doing anything. I also said my mouth was dry and had mentioned dry eyes to my optician but they were the least of my worries."*

The symptom experience includes an individual's perception, evaluation, and response to a symptom. People evaluate their symptoms by making judgments about the severity and effect on their lives (Dodd et al., 2001). Jo interpreted her joint pain to be more significant than her dry mouth and dry eyes. This was because Jo predicted that joint pain could possibly limit her functional status and in turn, can adversely affect her financial stability (which was imperative in her personal situation).

Jo: *"My biggest concerns were not being able to do the things I enjoy and I need to work to survive. I am a widow with a partner but we are financially independent and I don't qualify for the widows benefit."*

Following that, Jo had multiple tests, from allergy to blood tests, and went via the GP to the haematologist, who referred her to the rheumatologist. Her journey to diagnosis took over 3 years; Jo took partial responsibility for the time taken, saying that: "*it took way too long because I didn't really follow it through*". Jo recognised her responsibility as a patient, indicating that patient reciprocation is required to work with HCPs to reach diagnosis. In contrast, Mike, who is a HCP (district nurse), demonstrated initiative as a patient in working with HCPs to reach diagnosis. He experienced similar initial symptoms to Jo, but his interpretation of them and the resultant action taken demonstrated a relatively short time taken to reach diagnosis despite multiple tests and departments that were visited. Initially, Mike attributed his joint pain to normal ageing: *"I started getting niggles, nags, and pains, and ignored most of them of course, because that's age."*

This was also common to a few other participants (by virtue of the sample's age range) to initially rationalise their symptoms of aches and pains or poor sleep to be part of the normal ageing process. Mike is an active man who cycles to work; in retrospect, even though he noticed the dry eyes and dry mouth, he did not make the link, instead attributing them to cycling. This is an example of how an individual's lifestyle can affect the interpretation of initial symptoms.

Mike: *"I just put that down to that I bike and I'm dehydrated, didn't drink enough water or, I just didn't make the connection really."*

As Mike's mobility decreased, his visits to his GP increased. In the beginning, Mike suspected cardiac issues, and requested a referral to the Cardiology Department for a cardiology work-up; that proved to be fine. This was motivated by his previous family experience with major illnesses because he feared something similarly sinister.

Mike: *"At the same time, my mother as she was 89, she had a brain aneurysm… my sister… she had one, and this was all in the middle of the Sjögren's diagnosis. So I*

was thinking, wow, there's something going on here, normally its not hereditary, but I said to them look, and I was getting these headaches and all as well, that was part of it and I think I had high blood pressure, so that was possibly the headaches, so I went, and I pushed for a CT scan…"

Tina had a similar symptom (dry mouth and agitation) interpretation, where prior to SS diagnosis, she had anxieties about the possibility of a malignancy because her mother had passed away young from cancer.

As the pain persisted, Mike returned to the GP.

Mike: *"it started to work its way up by slowly affecting my joints, and that's when I hobbled in after about 3 months, I was hobbling into the GP, stooped over feeling like, I said to him, 'I feel like an 80 year old man." I said, 'Look, I'm not gonna be able to keep going to work if this keeps up.'*

As with Jo, Mike's symptom interpretation was connected to the concern about being unable to function at work. This in turn resulted in increased visits to the GP. These important interactions with HCPs will be discussed in a later section.

Mike *"My stress levels were high and rising - so after several (more than a yearly quota) of visits to the GP, he watching my obvious discomfort and increasing immobility - got on the phone straight away and organised a semi-urgent appointment at the rheumatology outpatients department the following day."*

Mike got his diagnosis within a year, after multiple GP and specialist visits, an ECG, and a CT scan. He attributed the short time taken to his nursing background acting as an advantage to understanding the healthcare referral system and the need to be persistent. Mike: *"I know my way around the department, I worked in the hospital for years and I kinda, definitely think that helped, because I was persistent."* Here, the patient's perspective of the symptoms led to him seeking medical help, and this enabled the ball to be shared in the HCPs' side of the court. Mike's GP's interpretation of the urgency of his symptoms was crucial because it led to a referral

to the suitable department for SS diagnosis. The next section will discuss other findings regarding HCPs' symptom interpretation.

5.2.1.2 Symptom interpretation by HCPs

For the current research participants, the symptom interpretation by HCPs can direct or delay the route to diagnosis. The HCPs involved were a combination of the dental and medical team, including: (1) dental hygienists; (2) dentists; (3) oral medicine specialists; (4) GPs; and (5) haematologists; (6) opticians; and (7) rheumatologists. Their awareness and acknowledgement of symptoms (including dry mouth) have been shown to play a significant role. For example, Margaret had a GP who picked up the relationship between her vague systemic conditions and her persistent swollen salivary glands.

Things took a twist in 1994 for Margaret (who also suffered from depression): *"I started getting sick then, getting tired and then I come home and then I just went downhill and downhill, I had different like, irritable bowel problems, lot of problems coming up, and then my glands swelled and, umm, I went to the doctor with it, so he got me in…"*

SS is known to cause extreme debilitating fatigue that may even be linked to depression in sufferers (Bax et al., 2002). As a disease of exocrine glands, SS also affects many parts of the gastrointestinal system (Ebert, 2012), and commonly cause swollen or tender parotid glands (Kruszka and O'Brian, 2009). Margaret interpreted her symptoms to be problematic, and sought her doctor. Fortunately, Margaret's GP was aware of SS and linked them altogether, and referred her to an oral medicine specialist for a minor labial salivary gland biopsy.

Margaret: "He (GP) thought oh, (it was quite lucky that the doctor (GP) thought of Sjögren's) he said, 'oh you might have Sjögren's.' So he sent me down to the specialist (Oral medicine) down at Kew hospital and he took a piece of my lower lip there, and it come back I had Sjögren's."

These findings demonstrate the influence of HCPs' awareness in interpreting Margaret's symptoms to be SS-related, resulting in diagnosis. On the other hand, Joyce experienced "*a long time (5-6 years) with the glands up and the butterfly face (malar rash associated with SLE) and then just getting sicker. When I say sicker and sicker I mean for a start I might take just half a day off work.*"

Once again, symptom interpretation was associated with its adverse effect on work. Joyce's symptom of swollen salivary glands (a similar symptom to that experienced by Margaret) was not interpreted by her GP to be SS-linked. Eventually, Joyce experienced more symptoms (pneumonia) and her GP interpreted these to justify a referral to the rheumatologist (leading to SS diagnosis). After a journey to diagnosis that felt lengthy to Joyce, she questioned the level of awareness that GPs have of initial SS symptoms.

Joyce: *"Well the thing is do the general practitioners know about it (SS)? Do they just think that somebody just gets flu and you know pneumonia or do they sort of think that it might be something, do they know what to look for because I was so long."*

Other SS sufferers shared similar difficulties. An American qualitative study using telephone interviews of women suffering from SS found that, because of the multiple symptoms of SS, there is a struggle to find a doctor who is able to recognise and diagnose the condition, and then to manage it adequately (Schoofs, 2001). The range of initial SS symptoms is broad and can be vague, causing HCPs difficulty in identifying them as a manifestation of the condition. As suggested by Joyce and demonstrated in Margaret's experience, the length of diagnosis may be improved if there could be an increase in basic awareness by GPs, who are often the first port of call in the patients' journey to SS diagnosis. In addition, this can also improve the situation for those who are undiagnosed (currently postulated to be more than half of sufferers). Early diagnosis and appropriate management are essential for optimal health outcomes in SS (Kassan and Moutsopoulos, 2004).

Other than the level of awareness, symptom interpretation by HCPs can only begin if they acknowledge the symptoms. Frieda, an 80-year-old Dutch lady, recalled that her early symptom of dry mouth was belittled and ignored.

Frieda: *"Sjögrens has been with me for quite a number of years now. Leading up to the awareness was when I had been to the beach one day and thought my tongue felt "sunburnt". This was really when I became aware of my dry mouth. My doctor (GP) would have no bar of it and sent me home with 'you must be snoring at night'. I struggled with this and my dry eyes for a few years also with my tiredness. It took quite a battle to finally have it checked out at the Dunedin Hospital and got the SS diagnosis."*

The term "battle" used by Frieda highlighted the perceived difficulty of her journey. This stemmed from the lack of acknowledgement of her initial symptom by her GP. Dry mouth is an important symptom of SS that can often be ignored because of a lack of awareness of its significance. This is an important issue because GPs were the first point of contact for most of the current participants. Even dental practitioners have been shown to require more training to be confident and competent in managing dry mouth and its complications (Abdelghany et al., 2011).

Frieda: *"I think it took about 3 years to finally, erm, to finally get him to acknowledge that it was something. And that was the time that I was getting so so, very very tired. I just didn't know to drag myself. So I went to him and I just cried, I cried and cried and that was the first time he took notice, and I think that was the time when I went to the hospital"*

In a study reporting the accounts of women being diagnosed with a long-term illness, doctors have been described as questioning the very reality of the women's symptoms, sometimes misdiagnosing them because the symptoms were interpreted by medical practitioners as not being serious (Kralik et al., 2001). Frieda's journey to diagnosis took more than three years. Even though it may not seem too long relative to other participants, the difficulty of the route was clearly related to struggling to have her initial symptom acknowledged by her GP.

There are other complexities to the diagnosis of SS. Even when the HCP is aware of and acknowledges the symptom to be significant to SS, the diagnostic methods can limit this interpretation. For Tina, it began with an oral symptom that eventually led

her to the oral medicine specialist in the Dental School. However, it took 6 years of testing to find out that she had SS.

Tina: *"Several years ago I started having real problems with different foods. My mouth became very agitated and at the time the dentist and doctor wondered if I had developed allergies to certain foods and drink. It seemed worse if I ate certain things like tomatoes and fruits with a higher acid content. I do not drink much alcohol at all but if ever I tried to have a glass of wine I felt like my mouth was swelling up."*

This demonstrated the oral manifestations of Tina's initial symptoms and her interpretation of their effects on her functional status (in terms of eating and choice of foods). These led Tina to seek advice from HCPs, who interpreted her symptoms to be associated with SS. However, multiple tests (including a biopsy) did not reflect SS during the initial stages. Diagnosis for SS is now based on classification criteria (such as the AECG criteria). These are not perfect for diagnosis, and a proportion of patients may be misclassified, particularly in the early stages of the disease. Thus, classification cannot be considered the medical standard for a diagnosis, and medical expertise is needed to establish a definite diagnosis (Vitali et al., 2002). This barrier is significant because it caused a significant delay in diagnosis: 6 years down the road, dry mouth became the presenting symptom, and, this time round, another visit to the GP resulted in a second referral to the oral medicine specialist and "*finally after a biopsy of my lip*", Tina "*got the positive diagnosis of SS*".

In this case, the lack of sensitivity of diagnostic measures was a barrier to diagnosis, even though patient and HCP symptom interpretations were directed towards SS diagnosis. The next section will examine the two spectrums of the heterogeneous routes taken to SS diagnosis in this current research.

5.2.2 Heterogeneous routes to diagnosis

The time range taken to diagnosis by the current participants reflects the heterogeneous routes taken. In an American study investigating the health experience of 277 SS patients, the mean time to diagnosis was reported to be 7 years. This was not unusual and was attributed to the non-specific nature of the presenting symptoms,

or to poor physician awareness of SS (Segal et al., 2009). For the participants in the current study, the length of time from initial symptom to SS diagnosis ranged from 1 year to 28 years. Joy had a rather straightforward incidental diagnosis because her dental hygienist picked up SGH, the objective sign of dry mouth.

Joy: *"So how did I first learn that I had SS? During a routine visit to the hygienist, she became concerned about the dryness of my mouth and suggested that an appointment be made at the dental school. This then started regular contact with the School. When Dr. Anita Nolan (oral medicine specialist) arrived, she arranged a biopsy for Sjögren's, blood tests and a referral to the rheumatology clinic."*

HCPs are trained to observe signs and link them to diseases. Joy's experience was different from the rest of the participants because she did not complain about any symptom (of dry mouth) leading to diagnosis, even though she does describe how she coped with it (dry mouth and food choices). Perhaps Joy's well-adapted attitude of acceptance ("*I've got that I will just deal with it as it comes along.. that's all you can do*!") had influenced how she coped with her symptom without seeking HCP advice.

In contrast, for other participants, it was a journey beginning with vague ailments that are not understood, resulting in visits to the GP. The initial symptoms varied a great deal, and the ensuing medical route took a few different turns as well. Ellie travelled the longest journey to diagnosis, some 28 years. She is a 61-year-old European woman who lived on a farm, bringing up four children. Her journey to diagnosis was the longest one among all the participants, beginning with sore joints and swollen feet at the age of 30.

Ellie: *"At the age of 30 I became quite ill with very sore joints and swollen hands and feet. The doctors (GP) thought I had sero-negative rheumatoid arthritis. I had all the symptoms but blood tests couldn't confirm. My life was absolutely miserable for 12 years, bringing up 4 small children and keeping up with the chores."*

Ellie's symptom interpretation was associated with her stage of life. Even though her diagnosis took 28 years from the start of her symptoms, she noted that the toughest

period of her life was during the stage of raising children and managing farm chores. Things did not go downhill in a linear manner for Ellie.

Ellie: *"Gradually it subsided and I became quite well again. I continued to keep quite well until 2009. Throughout 2009 I kept getting chest infections and I think by then I was quite conscious of a dry mouth and eyes. I had quite a bout of pneumonia which left my lungs with bronchiectasis. I kept getting these bouts frequently especially during the winter months, and many courses of antibiotics. My doctor (GP) referred me to Professor Highton (rheumatologist), who has done lots of blood tests, x-rays etc. and sent me to Anita Nolan (oral medicine specialist), and the eye clinic."*

This is a classic example of the insidious onset of symptoms that occurs in SS. The initial symptoms were vague and gradual, appearing to improve, and then coming in a serious bout of symptoms that led to diagnosis when she was further questioned about the route to diagnosis.

Ellie: *"I was speaking at something and, and I just had this dry mouth all the time and I got a drink and it didn't help… I have had quite a heavy cold and I thought oh maybe its just got to do with the cold. And err, but then when the cold left me the dryness continued. Yeah and I just was at the doctor's one day and with these eyes you see, they're probably more so the problem that draws attention to it. And he the doctor my local doctor says I think I know what you've got, 'you've got Sjögren's, Sjögrens.' And I said, 'I beg your pardon?' cause I've never heard of it you see. So he then sent me to Professor Highton who referred me to Anita you see. So it was Anita that had actually confirmed. But it was just yeah just very gradual onset really."*

Once again, there is a demonstration of Ellie's interpretation of her initial symptoms and how she tried to make sense of it by linking her dry mouth to her cold. Moreover, her HCP's interpretation of her dry eye to be SS-linked directed her towards diagnosis. The vague and gradual onset of the initial symptoms made the journey to SS diagnosis difficult. The symptoms can portend many other differential diagnoses. For example, the joint pain experienced by Ellie is a common initial symptom for SS. However, it can also be caused by many different types of injuries or conditions such

as viral infections or common age-related osteoarthritis. Her initial reaction of not knowing what SS was reflected (and highlighted) the limited public awareness of the condition. This factor will be considered in the following chapters on disease impact spectrum and coping mechanisms. Most of the research participants experienced a sense of relief when informed of their diagnosis. Joy and Tracy, however, were exceptions. Joy was surprised that neither her doctor nor dentist ever mentioned the possibility of SS. This lack of relief could be related to the fact that she had an incidental diagnosis of SS, and was not looking for an explanation of symptoms (like the rest of the participants). For Tracy, it was just a subset of the long-standing diagnosis of SLE, and the SS diagnosis was "*just another box ticked I suppose (laughs).. it just went into the umbrella of umm it's a, it's a autoimmune disease kinda thing and umm that was just part of it*."

For the other 8 participants, there was a unanimous sense of relief to finally "*have a name for the disease*". They were grateful to know the reason for their symptoms and that it was not all just in their minds. However, this was "*followed by shock and other emotions about its (SS) permanency and the connotations of an autoimmune disease*." (Mike) There were questions such as, "*what does it mean for me*?" or "*can it be treated*?" (Ellie). After being told that SS (and dry mouth) is a chronic condition with no cure, some found it "*hard to come to terms with the fact that there is no cure*" (Tina). Moreover, the reality of the disease consequences had to be accepted. Margaret (like others) found the greater risk of "*lymphoma, which is cancer*" to be "*pretty scary stuff*."

5.2.3 Conclusion

The journey to diagnosis was mostly based on symptoms. There was a wide breadth of initial symptoms (in different combinations) to be interpreted by each research participant. This depended on factors such as each participant's: (1) life stage; (2) disease experience; and (3) functional status affected. These led them to seek medical help, opening the symptom interpretation to HCPs. The findings show HCPs' symptom interpretation to be crucial to SS diagnosis. This interpretation depended on their awareness of, and acknowledgement of, the SS symptoms. There were uncontrollable factors, such as diagnostic barriers. The heterogeneity of the routes to diagnosis was demonstrated by the range in the time taken (1-28 years). Most of the

participants demonstrated a common relief in finding a name for their disease, followed by a reality check of SS consequences. The next section will continue to explore the participants' interactions with HCPs and how it affected QoL.

5.3 Interactions with Healthcare Professionals

5.3.1 Introduction

The theme ‘Interaction with healthcare professionals’ describes the relationship between the current research participants and their healthcare professionals (HCPs). First, there will be a discussion of these interactions in relation to the revised Wilson and Cleary model by Ferrans et al (2005). Next, there will a brief examination of the dichotomised roles played by the research participants, either active or passive, influencing the interactions with HCPs. Based on the empirical findings from the current research, there will be a description of the interactions with HCPs that affected QoL, in the context of: (1) referrals to specialists; (2) drug prescription; (3) patient education; (4) HCP accessibility; (5) financial aid; and (6) empathy.

5.3.2 The interactions with HCPs in relation to the revised Wilson and Cleary model

As discussed earlier, in respect of the revised Wilson and Cleary model by Ferrans et al (2005), the characteristics of the individual that influence health include demographic, developmental, and psychological factors (Ferrans et al., 2005), while HCPs are part of the social environment that can influence health (McLeroy et al., 1988). Empirical findings from the current research will be used to examine the bi-directional link between the two levels ‘characteristics of the individual’ and ‘characteristics of the environment’ in that model (Figure 13).

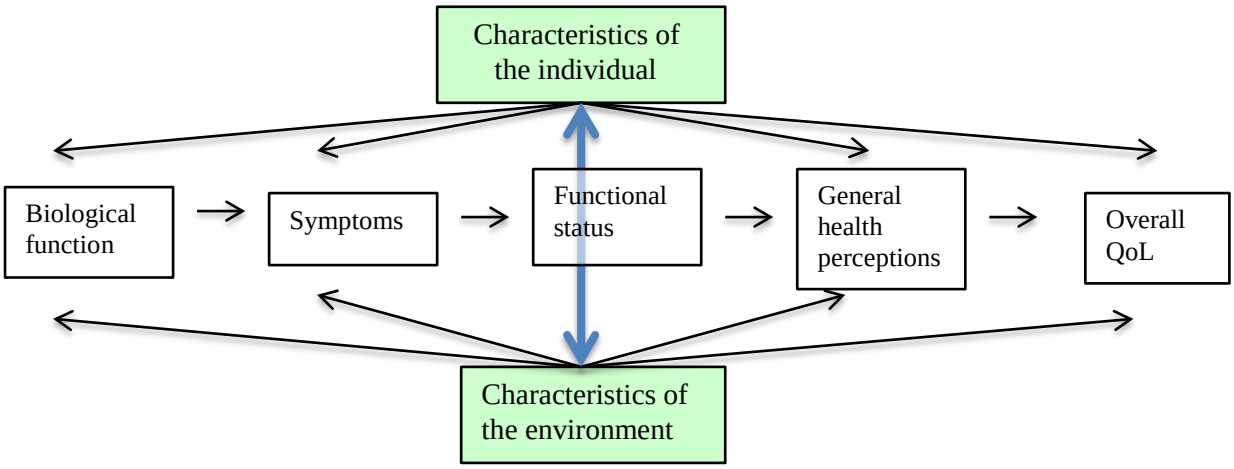


Figure 13. The revised Wilson and Cleary model for HRQoL (Ferrans et al., 2005) (highlighting in green the relationship between 'Characteristics of the individual' and 'Characteristics of the environment')

The characteristics of the individual related to these interactions included the participants' active or passive patient role, their personality, health perceptions and preferences. Owing to the nature of SS, the participants interacted with a wide variety of HCPs. These included: (1) dental hygienists; (2) dentists; (3) oral medicine specialists; (4) general practitioners (GPs); (5) rheumatologists; (6) haematologists; (7) emergency department staff; (8) optometrists; (9) ophthalmologists; (10) pharmacists; and (11) nutritionists. While the model conceptualised the existence of the relationship between patients and HCPs in influencing QoL, the clinical variables (biological function, symptoms, functional status, general health perceptions, and overall QoL) were unable to fully represent the richness of the interactions that were experienced by the research participants. The pertinent factors that were observed to significantly influence these interactions included participants' patient role, personality, preferences, lifestyle and perceptions, in the context of certain important aspects. These will be explored in the next sections.

5.3.3 Roles of the individual

Bodenheimer et al (2002) described two types of patient-HCP relationships (in the management of chronic disease): the traditional relationship, and the patient-professional partnership. These are two ends of a spectrum rather than wholly distinct

concepts. In the traditional relationship, the patient is more passive, and agrees to the HCP's treatment prescription. In the patient-professional partnership, the patient plays an active role in care collaboration and self-management education (Bodenheimer et al., 2002). The latter is becoming more dominant and tenable now, through a more humanistic-orientated emphasis on patient care. For the current research participants, there was a general dichotomy into passive and active patient roles (Brown et al., 2002), affecting their interaction with HCPs. These roles will briefly be discussed in the following sections.

5.3.3.1 Passive roles

The participants who played a more passive role tended to be less controlling, and more accepting of the prescribed treatment. One of them was Tina, who was very healthy prior to SS, and therefore had had limited interactions with HCPs. She described herself as not a "*pro-active*" patient who did as she was told: "*goes to the appointments*" when necessary, and "*just put up with the other things*." She described her compliance with the multiple medical appointments (such as with the ophthalmologist and the rheumatologist) that were "*just one thing after the other*", despite their being against her personal preferences.

Tina: "*Sometimes I just, I get to a point where I've had a whole lot of appointments and that's it, just leave me alone.. just wanna be normal.. I've got this thing that I have to deal with on an hour-by-hour, day-by-day basis… the implications you know.. it affects my work… I can still do my job but its quite difficult to deal with and then, if I'm having a lot of appointments as well.. how do I fit that in?*"

The findings depicted how Tina attended numerous appointments because of referrals between HCPs. These interactions with HCPs had negatively impacted on her QoL; they were interpreted as a transformation in 'normality', and had an adverse effect on her work. Tina found that difficult to manage because work was part of her coping strategy; work can have a deep connotation for individuals because it is an area for them to express their identities (Christiansen, 1999). Tina did not reveal these impacts on QoL to those HCPs, and perhaps she never will, because of the passive role she played. In the current research, half of the participants were observed to play a similar

role. Understanding this passive patient role is important for implementing interventions to increase patient involvement, because active patient participation in medical decision-making has been shown to improve health outcomes (Greenfield et al., 1988). The challenge lies in facilitating patient-HCP relationships that can balance advocacy for active participation with individual patient preferences (Arora and McHorney, 2000).

5.3.3.2 Active roles

The participants who played more active patient roles tended to chase after their own referrals, self-alter their prescription drugs, and participate in the medical decision-making. Such individuals tend to be their own principal caregivers and expect HCPs to play a supportive role (Bodenheimer et al., 2002). For example, in the Department of Rheumatology, patients often "*fall out of the (appointment) system*".

Tracy: "*They (Rheumatology department receptionist) will send you an appointment for 6 months. A year later I haven't got an appointment, and then and then you have to go back to your GP and then you start the (referral) process again*."

Tracy was more passive, and waited unsuccessfully for overdue appointments. This resulted in her having to go through the referral process again in order to get back into the specialist department appointment system. In contrast, Mike, who was a HCP himself (district nurse), directed his own appointments that led to his diagnosis (as described in 'the journey to diagnosis'). During maintenance appointments, the hospital system "*forgets*" and he was always "*following them up*". Moreover, Mike perceived that his "*clinicians were not answering*" what he thought "*were basic medical questions*." He took control and "*decided to halve*" his "*chloroquine medication (disease-modifying antirheumatic drug)*" as an experiment "*because they are not gonna give me (him) answers*." These findings reflected Mike's lack of faith in HCPs that led him to self-manage SS. A patient's confidence in his or her HCP's knowledge and skills is a component of trust (Arora and McHorney, 2000), and older and less educated patients seem to have greater trust in HCPs (Anderson and Dedrick, 1990). The active/passive roles played by the current research participants depended on their trust and openness with their HCPs, and they had an effect on QoL. The

patient-physician relationship can impact on health, it is therefore important to understand the nature of the relationship (Arora and McHorney, 2000). The next section will discuss the important aspects of this relationship from the research participants' perspectives.

5.3.4 Interactions (of the current research participants) with HCPs

The empirical evidence from the current research demonstrated the dynamics in the interactions between the participants and their HCPs. These interactions affected the participants' QoL in the context of: (1) referral to specialists; (2) prescription medications; (3) patient education; (4) HCP accessibility; (5) financial aid; and (6) empathy. Each will be discussed.

5.3.4.1 Referrals to specialists

The referrals to specialists had different connotations for the current research participants. Tina found that the multiple appointments affected her life balance, and the initial meetings with specialists were intimidating because she "*didn't know what to expect*"; this is an example of the historic "white coat" perception (Hochberg, 2007). In contrast, Margaret (who lived away from Dunedin) found it "*hard to get a specialist*", and perceived the referral to an oral medicine specialist in Dunedin to be a "*breakthrough*" for both her and her husband Ian. There was a sense of hope while they anticipated the specialist appointment, because they perceived him/her to own a specific set of skills "*to do the major thing*". Such specialist referral appointments had very different meanings (and hence impact on QoL) for the research participants. This may have been influenced by the participants' life stages; for Tina, the referrals were intimidating and difficult to manage (because of her hectic work schedule), by contrast, for Margaret (who was retired), the referrals were a source of hope and positively affected her (and her husband's) QoL. Although specialist referrals are an inherent part of managing SS, it would be useful for HCPs to understand what the appointments could mean for each patient, and perhaps make some adjustments (such as co-managed appointment time intervals for individuals like Tina). Moreover, both Tracy and Tina observed the effect that student involvement (in the Dental School) had had on them. Tracy felt that the biopsy and follow-ups had been more beneficial for student demonstration and teaching, rather than her welfare. Tina, who was rather

shy, was put on the spot because of students observing her appointment (with the oral medicine specialist).

Tina: "*I don't know whether they were trainee specialists or whatever.. because the times I've seen her (oral medicine specialist) before it had just been her and the nurse, and I walked in there was this wall of men standing there and it was this really small room and it was like, uggh God! You know, open up the floor and swallow me because I just don't wanna be here, and I find that quite, I'm quite a shy person I find that quite intimidating.*"

The findings demonstrated how Tina's interaction with her HCP had a negative impact on QoL, because she was self-conscious and felt pressurised by the unexpected presence of observing students during the consultation. This is a reminder for HCPs to take steps to ensure patient willingness and comfort during such teaching sessions. This could perhaps be in the form of seeking permission from patients prior to the appointment (under the New Zealand Health and Disability Commissioners Regulations (1996), Tina had the right to refuse to have students in the consultation room), understanding their level of comfort with student involvement, or clearly outlining the roles and responsibilities of participating (or observing) students (Hartz and Beal, 2000).

5.3.4.2 Prescription medications

As discussed earlier, owing to the lack of cure for dry mouth and SS, disease management is mainly by symptom management. Prescription medication was the outcome of the interaction with HCPs for 9 out of the 10 current research participants. The half (of the research participants) who tended towards passive patient roles received their prescriptions (that they perceived to have a positive impact on QoL) with compliance. Ellie did "*feel a little better*" with antibiotics that were prescribed for her cold. Joyce would bring along a prescription of antibiotics for her vacations (to take in the event that she felt unwell during the trips), and did not express concern when her Plaquenil (a disease-modifying antirheumatic drug) dose was increased. Moreover, she was glad because the dosage became "*easier to remember rather than every second day up*". In contrast, the other more "active" half of the participants

found that prescription medications had a negative impact on QoL. They expected more information on the side-effects of their medications and were interested in complementary and alternative medicine. Mike was concerned about being on long-term medication and wanted to know how long he would be on Plaquenil. His rheumatologist's response was, "*you are on a low dose, doesn't matter how long you're on it for*." Mike felt "*dismissed*" because he was "*hoping to get off the medication*." Following that, Mike sought alternative medicine, such as acupuncture and supplements. In chronic diseases, seeking alternative medicine can be a way of attempting to alter the illness trajectory by finding someone who might provide alternative pictures of it (Milliken & Northcott, 1996). Likewise, Tracy experienced longstanding joint pain and opposed the idea of long-term medication.

Tracy: "*You're chucking more pills down your throat.. I don't wanna carry on doing that because every time I've gone to the GP they just said, 'oh well you know you've got your four pain pathways we can block every one of those.' So you know, your anti-inflammatory, your paracetamol, your codeine, and so then you end up with more tablets.*"

Tracy disagreed with, and was frustrated with her HCP's approach. She had no confidence in prescription medications and did not want to "*go down that road*" that she perceived not to address the root of her problem. Subsequently, Tracy began to explore complementary and alternative medicine, and started to see a nutritionist.

Tracy: "*I want to try and get better but not by just going oh we will stick a needle, stick a cortisone in there.. they've never ever mentioned diet.. diet could help with your pain or with the Lupus (that was associated with secondary SS) and stuff like that which umm I think is a bit of a shame because I think it could help, so I'll just take that route myself and see what happens*."

These findings revealed that different participants had divergent beliefs about the effect of prescription medications on QoL. While HCPs prescribed medications in faith that it would positively influence the participants' QoL, the latter may perceive the opposite. Moreover, there was a communication barrier between Tracy and her HCPs; she did not express her views on conventional versus complementary

medicine, and her HCPs did not fulfil the expected role of providing information that was pertinent to her. Instead of collaborative care, whereby physicians and patients make healthcare decisions together, the lack of communication made Tracy decide to "*take that route myself*". Jo recounted her attempt to collaborate with her HCPs on homeopathy regarding the use of colloidal silver (a natural antibiotic); her rheumatologist told her to "*just forget it, its not going to do you any good*", and her optometrist dismissively "*rolled his eyes and said "oh whatever"!*"

Complementary and alternative medicine (CAM) is growing to be popular among patients with chronic diseases because of the long-term side-effects of conventional medication. Physicians' indifference (or opposition) toward CAM use because of their emphasis on scientific evidence (and patients' anticipation of a negative response from their physician) have been found to be barriers to patient-physician communication (Tasaki et al., 2002). HCPs need to be more aware of their patients' opinions on the use of CAM. They should be open to providing information on the risks and benefits of CAM, and refer patients to other services that may address unmet needs. Given a difference in epistemiologic beliefs (on the effect of conventional medicine on QoL), the challenge is to find common ground for an open discussion.

5.3.4.3 Patient education

HCPs can be a helpful source of knowledge for patients. There is a growing acceptance of the view that patients need knowledge and information in order to engage and participate in the management of their illness (Kennedy and Rogers, 2002). This patient education can be in the form of offering available resources, or providing direct information. Margaret was grateful to her GP who "*wrote away to Sjögren's society*" to connect her to a reliable source of information. However, because SS is relatively uncommon, some of the current research participants had pointed out HCPs' lack of knowledge of SS. Jo was unsuccessful in seeking information from her GP during her SS journey: "*he just.. shrug.. shoulders and oh don't know about that*". A willingness to disclose information to patients constitutes an integral component of shared management of chronic disease (Coulter, 1997). While Jo was looking for collaborative care with her GP, she perceived that he had transferred that responsibility to her specialists after referral. Ellie found her

pharmacist to be ignorant, and experienced a reversal of roles when she had to explain SS to him/her.

Ellie: "*There's not a lot of knowledge.. in the chemist and places like that, they're not really all that helpful I don't think, because they don't really know. I have to explain to the girl working behind the counter that I've got Sjögren's disease and do you know what that is*?"

Nonetheless, it may be worth taking into account that the 'girl working behind the counter' may not necessarily have been a pharmacist. Likewise, Frieda found that her dentist was: "*excellent but of course, he doesn't know the first thing about the Sjögren's*". The lack of patient education and information provided by HCPs had an adverse effect on the QoL of some participants (in respect of the detrimental effects dry mouth had on their dentition). Joyce was frustrated that her rheumatologist never mentioned the side-effects of dry mouth on her dentition (that cost her a lot physically and financially), and exclaimed that "*the teeth were never thought about*!" A similar dissatisfaction was expressed by PM: "*you guys (old generation dentists) knew I had a dry mouth and you never did anything about it*", and wondered in exasperation, "*why didn't they tell me I needed to see a specialist*?" Moreoever, PM viewed the new generation of HCPs to be "*far better than what they were ten years ago*" (in terms of providing knowledge). HCPs' provision of knowledge is important because it facilitates patient education to achieve adequate control and to prevent adverse health outcomes (Williams et al., 1998). This was a noteworthy aspect of the interactions with HCPs that can affect QoL.

Moreover, patient education became confusing when HCPs had differing opinions. Margaret described how one GP told her to "*get lemon drops*" while another GP told her "*don't touch them*" because of the acidity that can be detrimental to her teeth. As it turns out, Margaret was edentulous. Nonetheless, this led her to find "*a way to deal with it*" herself, using chewing gum and water. This was an interesting paradoxical outcome, because self-management education is important for patients to be taught problem-solving skills for the problems encountered in chronic illness (Bodenheimer et al., 2002).

5.3.4.4 HCP accessibility

Traditionally, consultations with HCPs have been associated with cost. Tracy found that, "*without paying money they (HCPs) are not really accessible*". She did not turn to HCPs when she had questions about her SS. This is a pity, because patient education (by HCPs) plays a critical role in facilitating patients' acceptance of their diagnosis and understanding behavioural changes required for active participation in treatment (Grueninger, 1995). Owing to the nature of the current research (that required building a rapport between interviewer and participants), email contact was maintained between the interviewer (who is a HCP) and the participants. The positive impact of accessible HCP contact was evident in Jo's experience, where she expressed gratitude for being able to access a HCP (the interviewer) easily when she developed lumps around her neck (that were possible signs for SS-associated lymphoma).

Jo: "*Its good to know that there is somebody out there that.. I can contact.. when I got those lumps I thought, 'oh yeah! (smiles).. instead of rushing off to the doctor and getting on antibiotics or whatever they shove you unto, it was good just being able to dial you up (referring to email).. I've got a lump what do I do because.. she (oral medicine specialist) had pointed out, got to watch out for those sorts of things and its good to know there's gonna be a bit of action at the other end (clear's throat) otherwise you'll be left sort of hanging there thinking, oh got a lump nobody cares"*.

During the course of SS, there will be new findings that patients can have uncertainties about. This finding depicted how easy access to medical advice had a positive impact on Jo's QoL. There was an element of being reassured and cared for (by HCPs), without having to take prescription medications (that Jo perceived to be unnecessary). There have been initiatives for electronic-consultations (Stoves et al., 2010) and chronic diseases are particularly suitable for remote management, especially when there is continuity between the patient and service provider (Holman and Lorig, 2000). Electronic contact may be a useful avenue for effective patient-physician interaction in collaborative SS care.

5.3.4.5 Financial aid

The current research participants appreciated indirect financial aid from HCPs because of the monetary burden that can come with chronic illness (Emanuel et al., 2000). Joy perceived that her "*dentist is very good*" because he had not charged her the full amount. Likewise, Joyce thought highly of her dentist because of the free preventive dental products provided. Moreover, she provided an example of the contrast between private and public healthcare costs.

Joyce: "*All those years I've paid out thousands of dollars and I could have been getting it done at the hospital for nothing, oh!*"

The findings demonstrate the economic burden for participants suffering from dry mouth, and therefore there was positive regard for financial support from HCPs because of its positive impact on QoL. Public health care policy makers have reason to explore funding allocation for these patients.

5.3.4.6 Source of empathy

Traditionally, HCPs have been focused on the treatment and cure of diseases. Mike expressed how his HCPs "*were quite pleased to have something unusual (male patient with SS)*" instead of addressing the uncertainty he had about his diagnosis. Even though there was no cure for SS, the participants valued HCPs as a source of empathy, because the sense of "*being understood*" through listening was important to them.

Frieda: "*It is so important that you are able to talk to them (HCPs), I know at the moment there's nothing that you can do for me! It doesn't matter, but you were listening, you were just not tossing it*."

Similarly, Jo was happy to travel for 3 hours to Dunedin for her appointment with her oral medicine specialist because she found it therapeutic. Jo felt "*a lot happier now*" because she could "*actually talk to somebody who actually knows something about it (SS) even if there is not a lot that you can do about it (SS).*" Tina felt that this understanding was important for SS patients because it was "*a kind of a weird thing to*

have", she had not "*come across anybody with it*" and none of her "*friends or colleagues have ever heard of it before*". The common pattern across the data emphasised the value of empathy from HCPs that was expressed through listening. Listening is important to patients because they perceive it to be: (1) an essential component of clinical data gathering and diagnosis; (2) a healing and therapeutic agent; and (3) a means of fostering and strengthening the doctor–patient relationship (Jagosh et al., 2011). PM perceived female HCPs to be more empathetic because they listened and understood "*how sensitive*" she was. By contrast, she found that male HCPs were: "*not listening to what you are actually saying*". Empathetic communication varies with physician and patient gender. Empathetic opportunities created by females have been found to exhibit more emotional intensity than those created by males. Moreover, female physicians have been found to communicate higher degrees of empathy (Bylund and Makoul, 2002). Owing to the female predilection in SS, the empathetic needs of its sufferers may be higher. Empathy was found to be an important component of the patient-HCP relationship for these participants, having a positive impact on overall QoL.

5.3.5 Conclusion

This is the first qualitative approach to examine the patient-physician relationship that is represented by the bi-directional link between the two levels 'characteristics of the individual' and 'characteristics of the environment' in the revised Wilson and Cleary model by Ferrans et al (2005). There were aspects of these interactions with HCPs for the current participants that were not represented by the model. The current research participants either tended towards active or passive patient roles, and, together with their personalities, lifestyles, preferences, and perceptions, these had influenced their interactions with HCPs. The important aspects of these interactions were described in the context of: (1) referral to specialists; (2) prescription medications; (3) patient education; (4) HCP accessibility; (5) financial aid; and (6) empathy. Through the perspectives of the current research participants, the theme 'Interactions with HCPs' have provided an opportunity for clinicians to understand the impact of their behavior on patients' QoL. The next section will discuss how the participants interact and perceive their disease, and its impact on QoL.

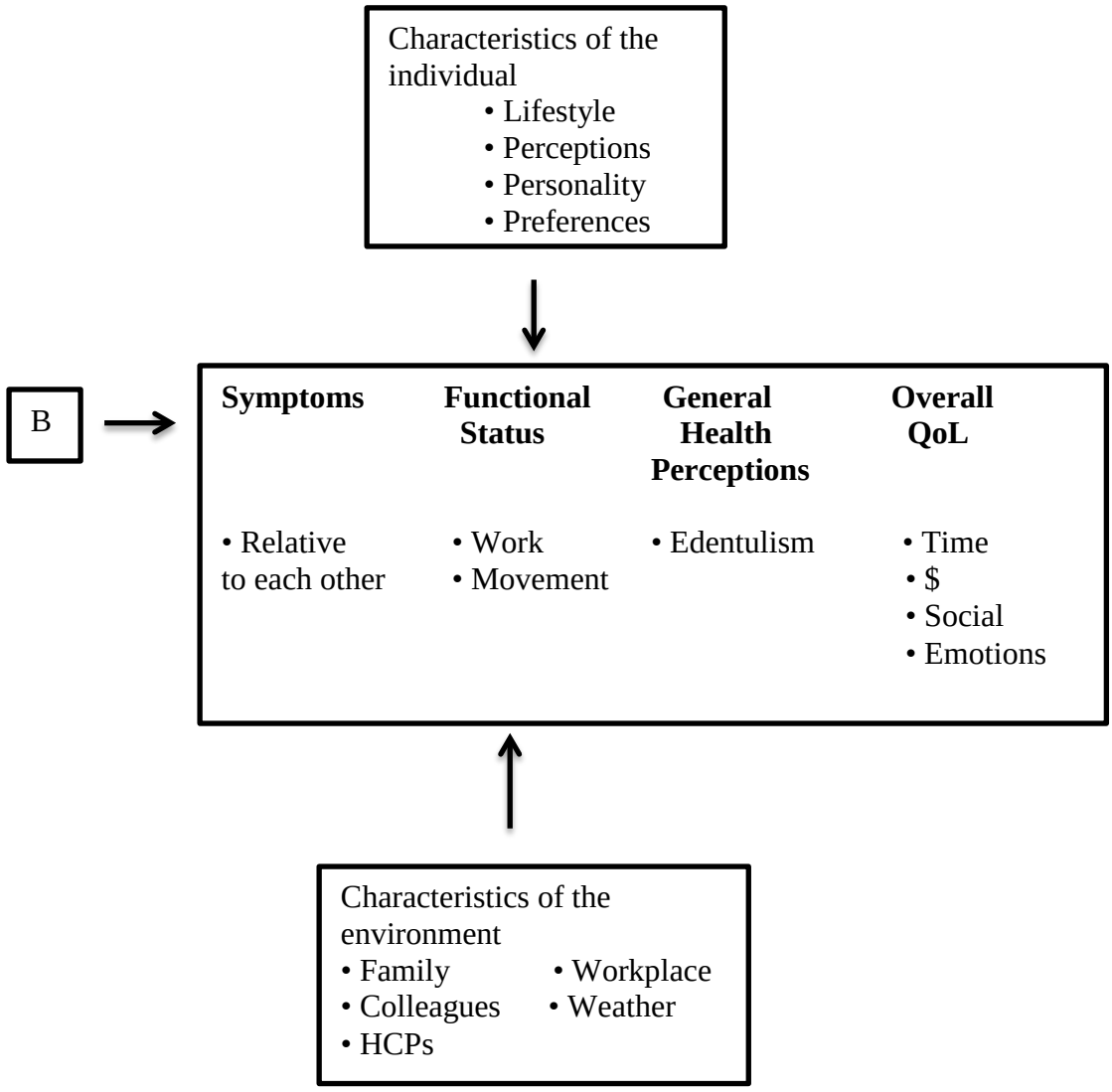
5.4 Disease impact spectrum

5.4.1 Introduction

This chapter will explore the theme 'Disease impact spectrum' in order to provide insight into the dry mouth experience for SS patients. First, the research participants' conceptualisation of the SS (and dry mouth) impact on their QoL will be discussed in relation to the revised Wilson and Cleary model by Ferrans et al (2005). The empirical findings demonstrated that the current research participants' dry mouth experiences were amid a spectrum of other consequences of SS. These included: (1) dry eyes; (2) fatigue and sore joints; (3) lowered immunity; (4) the effect of treatment; and (5) a background fear. Next, the dry mouth impact will be described in the areas of: (a) communication; (b) time; (c) finances; (d) emotional stress; and (e) lifestyle. Dry mouth was described as a 'hidden disability', and its impact on the daily lives of SS patients is best understood when considered as a subset of the disease impact spectrum of SS, the individual, and his/her environment.

5.4.2 The participants' conceptualisation of the disease impact on their QoL

The revised Wilson and Cleary model by Ferrans et al (2005) is a conceptual model for understanding the impact of diseases on patients' QoL. It was based on how HCPs conceived HRQoL (or OHRQoL) to be a network of relationships among various clinical variables, with input from patients' individual and environmental characteristics. The theme 'Disease impact spectrum' focuses on how the research participants viewed SS (and dry mouth) as impacting on their QoL in a dynamic manner. Their conceptualisation of disease impact was not completely represented by the model. The findings described aspects of the participants' perceptions such as: (1) how the multiple SS symptoms related to each other; (2) how they experienced the less-tangible impacts of dry mouth; (3) how treatment had negatively affected them; and (4) the fear generated from the nature and consequence of SS. The participants' conceptualisation of HRQoL/OHRQoL is represented in Figure 14.



[B: Biological function]

Figure 14: The current research participants' view of the revised Wilson and Cleary model by Ferrans et al (2005).

The size of the boxes indicates their relative importance. From the participants' perspectives, biological function was not important (never mentioned). Of course, this is something important to HCPs, who measure them for assessment and diagnosis. From the participants' perspectives, the levels of the model ('symptoms', 'functional status', 'general health perceptions', and 'overall QoL') were reconceptualised and merged into one level through their cognitive processes of how SS (and dry mouth) had impacted on them. This was different to how the levels were distinct and

measureable in the model. The next sections will go on to describe the range of symptoms experienced in SS, with a focus on dry mouth.

5.4.3 The range of physical symptoms in the SS disease impact spectrum

As described earlier, SS is an autoimmune disease that affects multiple organs and systems. The disease impact of SS has tended to be considered in terms of its effect on individuals' QoL in a segregated manner (divided into dry eyes, dry mouth, or employment status) (Meijer et al., 2009; Miljanović et al., 2007; Stewart et al., 2008). During the description of their dry mouth experience, every research participant brought up the other impacts of SS. These included: (1) dry eyes; (2) fatigue and sore joints; (3) lowered immunity; (4) the effect of treatment; and (5) a background fear. This next section will briefly discuss each of them, with empirical evidence from the data.

5.4.3.1 Dry eyes and dry skin

Dry eyes have an adverse impact on several common and important tasks of daily living, thus negatively affecting QoL in SS patients (Miljanović et al., 2007). The skin has been known to exhibit dryness, with patients frequently experiencing dermal stinging and itching (Tzioufas and Moutsopoulos, 2003). These two symptoms were common among the current research participants, being intermittent for most of the research participants, while having a constant effect on some.

Mike: "*Dry eyes are a constant annoyance*."

The use of the word 'annoyance' for Mike indicated the nuisance factor derived from dry eyes, which was described as being ongoing. Likewise, Joyce experienced a continuous dryness of her scalp.

Joyce: "*My scalp of course is perpetually dry*."

For Joyce, her dry skin manifested on her scalp, and she went on to describe how she managed it with sensitive skin products. Tina, who was often conscious about what

others thought of her at work, described the psychological effect having a dry skin had had on her.

Tina: "*This can be a real problem as often you are not always aware that you have blood or dry skin on your face, which is embarrassing*."

Understanding what dry eyes and dry skin meant to the research participants was important because these (symptoms) acted as a benchmark against which they could relate their dry mouth.

Ellie: "*My eyes are equally bad (compared to dry mouth), if not worse. I put many drops in them during the day and night*."

There will be a comparison and prioritising of symptoms as patients self-manage multiple chronic conditions (Lindsay, 2009). The concept that the individual's interpretations of symptoms is dynamic and relative (to each other)—depending on the 'nuisance factor' of each of them, the resultant psychological impacts, or whether one symptom is constant, while the other is intermittent—is not represented in the revised Wilson and Cleary model by Ferrans et al (2005).

5.4.3.2 Fatigue and joint pain

Fatigue and joint pain have been shown to negatively affect the QoL of SS patients, with somatic fatigue being the dominant predictor of physical function and health (Segal et al., 2009). These manifestations of SS had a significant effect on most of the current research participants. Tina made sense of the impact that fatigue had had on her by comparing herself pre- and post- SS.

Tina: "*I could keep going for four or five days with very little sleep.. but I can't do that now. And like last week I just got to the point where I just I could hardly walk a clear line, I was just exhausted and so its been a big wake up call for me to realise how tired it can make me feel*."

The cognitive process for Tina to understand the impact of fatigue was based on her pre-SS function. Moreover, Tina's interaction with her daughter revealed that the fatigue she experienced was more than a physical symptom.

Tina: "*My daughter thought I would ask questions you know, three or four times and I'd forget that I'd even asked that. So it was not just physical but it was mental tiredness as well*."

In QoL measures (such as the SF-36), the two domains (physical and psychological) of functional status have commonly been scored separately. However, the reality for Tina was that her mental fatigue was intertwined with her physical tiredness. Interestingly, this was identified through family interaction, providing insight into the influence from the level 'characteristics of the environment' (on health perceptions) in the revised Wilson and Cleary model by Ferrans et al (2005).

Mike experienced a "*constant pain almost 24/7"* that affected his daily function. Tasks that were formally simple had become challenging (such as reaching the top of a shelf), causing him to "*feel restricted*". Mike felt that "*getting rid of it (joint pain) was a priority"* to him. It was also dependent on the level of demand that work had on him. "*On the more stressful days (at work)"*, Mike tended to "*feel more tired than usual.*" Mike's experience showed that his perception of the disease impact on his QoL was based on the cognition of the effect of the symptom (pain) on his functional status (that was relative to pre-SS). As a result, eliminating the pain became important to him. The influence of his workload on QoL was evident, demonstrating that disease impact can be dynamic under environmental influences.

As with Tracy, her lifestyle and family influenced her interpretation of the impact joint pain had on her QoL.

Tracy: "*I used to play a lot of sports and I suppose that for me is the biggest thing that's kind of stopped just because of joint pain, and my body just feels a lot weaker than what is ever has done.*."

"*I still want to you know, to be able to run around and take them (her 2 children) out and do stuff with them I don't want to be.. mom-in-the-car watching from the sidelines..*"

Owing to her active lifestyle, Tracy interpreted the impact of joint pain on her QoL in terms of how it had affected her ability to do sports (alone and with her kids). Likewise, Tracy benchmarked her functional status to her previous experiences. Her perspective of disease impact on QoL was influenced by her sporty personality, family role (Mum), and interaction with her kids. It was important to understand what fatigue and joint pain meant to the research participants because dry mouth was perceived to be relatively manageable for some.

Jo: "*As soon as I got pain-free I feel like I am a new person, I still got a dry mouth and a cough but I still feel like a new person.*"

The effect of fatigue and pain was serious for the current research participants. These findings provided insight into how the participants interpreted the impact of joint pain and fatigue, together with their effect on functional status, with reference to their functional ability pre-SS. Moreover, the perception of disease impact was influenced by characteristics of the individual (lifestyle) and the environment (work and family). This understanding has provided a more accurate background to the dry mouth experience of these SS patients.

5.4.3.3 Decrease in immunity

SS is an autoimmune disease, and the medications used to treat it may suppress general immunity. The current research participants experienced a greater susceptibility to common colds and flus. Tina found that she became more "*prone to picking up these bugs which take ages to clear*". When Joyce experienced a cold or flu concurrently with SS, its impact on her QoL was worse.

Joyce: "*Sjögren's is bad enough, flu and cold on top of it, it's a real bitch! Because with the immune thing you get really sick.*"

Ellie noticed that her SS (and dry mouth) experience was dynamic because of the weather. The cold weather made her cough or suffer from a blocked nose, in contrast, in summer time, she had less chest problems and was "*more on top of*" herself. Moreover, her dry mouth worsened when she had to breathe through her mouth.

Ellie: "*My mouth has a tiny bit of moisture in it -- when its closed. The moment it opens it evaporates. I have to make a conscious effort to keep the mouth closed. At times when my nose is blocked or when I have a cold it is not possible to do this*."

The impact of a decreased immunity, together with weather changes on the SS (and dry mouth) experience is a new insight that reflects on the factors that can render the disease impact dynamic.

5.4.3.4 The effect on treatment

The effects of medications and post-biopsy site numbness (from the SS diagnosis) were described by the research participants. Plaquenil is a disease-modifying antirheumatic drug (hydroxychloroquine), commonly used to manage autoimmune diseases such as SLE and RA (that are associated with secondary SS). The common side-effects of the drug include nausea, diarrhoea, retinopathy, skin reactions, neuromyopathy, and the risk of accumulation in the presence of renal or hepatic insufficiency (Ochsendorf and Runne, 1991).

For Mike, his "*hair started thinning out*" and he had to have "*blood tests and urine tests to make sure it wasn't damaging kidneys and stuff*." Mike was frustrated when he was "*told about the potential side-effects of the medication*." For Joyce, it affected her lifestyle as she had to "*watch the sun with the Plaquenil, or (her) arms will be blotchy*."

Pilocarpine is a nonselective muscarinic receptor agonist drug which is commonly prescribed to manage dry mouth. Its adverse effects are dose-dependent and include sweating, rhinitis, headache, urinary frequency, diarrhoea, dyspepsia, nausea, and dizziness (Lovelace et al., 2014). Tracy was prescribed pilocarpine many years ago,

and when questioned about whether the drug worked for her, she could not remember, but her recollection of its side-effects was vivid.

Tracy: "*I am not taking that pilocarpine; that was horrible.. it was hideous, that was Prof Ferguson he gave that to me and umm I just was flushing hot flushes and feeling nauseous all the time.. I didn't like that at all so (clears throat)..*"

The effect of SS on QoL is not from the disease alone; the effect of treatment was significant to the current research participants, both physically and emotionally. These empirical findings provided a conceptualistation of the effect of treatment on the participants' QoL, addressing the limitation of the Wilson and Cleary model (1995) in capturing the influence of therapeutic regimens (Bakas et al., 2012). Moreover, this provided background understanding of why the current research participants were keen to try complementary and alternative medications (to be discussed later).

Minor labial gland biopsy is one of the diagnostic procedures for SS in the AECG criteria (Vitali et al., 2002). Post-biopsy numbness was a complication that adversely affected Ellie's functional status and self-esteem. The selection from the interview with Ellie who was never "*an over-confident person"* described how this complication "*dented it (her confidence) a wee bit more"*.

Ellie: "*Well I just don't like umm being the centre of attention at all, can't handle it! I found too like with the wee biopsy.. my lips are.. all still a bit numb and so if I'm drinking something I sometimes dribble a wee bit you know? (soft laughter) Just all those wee things that nobody else probably notices but I do.*"

Interviewer: "*And how does it make you feel*?"

Ellie: "*A bit stupid really (laughter). I don't know, I realise that I can't do anything about, I just gotta learn to cope with it, yeah.*"

This is an example of how the qualitative interviewing process probed into the disease experience with associated deeper impacts; in this case, it explored the side-effect of a diagnostic procedure. Post-biopsy numbness is known to improve with time;

unfortunately, it had not completely disappeared for Ellie. Her experience allowed insight into the SS impact on QoL in terms of the psychosocial consequences of an iatrogenic procedure.

5.4.3.5 The background fear

Owing to the 44 times greater risk for lymphoma (Kassan et al., 1978), and the genetic predisposition in its aetiology (Loiseau et al., 2001; Reveille et al., 1984), SS had an emotional (and psychological) impact on the current research participants. Some of them lived with a background fear of SS complications to themselves and their family. Mike, a district nurse, observed that: "*Lymphoma scares the hell outta you*!" Knowing that he had a greater chance of a cancer negatively affected Mike's QoL. Moreover, Mike's interpretation of the complications of SS was intensified by his background knowledge in cancer by virtue of his occupation.

The genetic predisposition caused Margaret's and Ellie's daughters to be "*worried that they might get it too*"; therefore, as mothers, the former had to "*try and reassure*" them. These underlying fears (because of the complications and nature of SS) affected the research participants and their family interactions; this was an important and less tangible impact that SS had had on their QoL.

The findings in this section demonstrated the breadth of SS's impact on the current research participants, who experienced different combinations of its multiple manifestations. This sets the background on how they viewed dry mouth as part of the SS disease impact spectrum. This relative perception of symptoms was summarised by Margaret: "*I can put up with that (dry mouth) but I can't up with some of the other things that are happening*". The next sections will focus on understanding the diverse impacts of dry mouth.

5.4.4 Dry mouth

Dry mouth in SS patients has been related to: (1) problems with mastication; (2) alterations in taste sensations; (3) pain in the salivary glands; (4) speech difficulties; (5) nocturnal discomfort; and (6) an increase in fluid intake (Soto-Rojas and Kraus, 2002). In this section, based on empirical findings, there will be a description of the research participants' dry mouth experience. There will also be consideration of the

physical impacts (on eating, sleeping and the dentition) and the less tangible adverse impacts (on communication, time, finances, emotional stress, and lifestyle) that dry mouth has had on them.

5.4.4.1 Dry mouth and eating

The effect of dry mouth on eating is important. Three of the five items in the SXI measure the association between dry mouth and eating. These include: 'I have difficulty in eating dry foods'; 'My mouth feels dry when eating a meal'; and 'I have difficulties swallowing certain foods'. The empirical findings supported the impact of dry mouth, as described by these items and further demonstrated aspects of (1) pain, (2) taste disturbances, and (3) alteration in food choices.

For Tina, pain was inherent to eating (and brushing) because of the dryness associated with the skin around her mouth and her lips.

Tina: "*It brings a lot of pain at times when you try and eat, clean your teeth and deal with the pain around my mouth when my skins flare up and my lips get so dry, they are constantly cracking. It is a constant battle trying to get everything lubricated. My water bottle is my best friend.*"

The use of the term "*constant battle*" emphasised the ongoing difficulties that Tina had to face. This added personal meaning to the SS impact on her QoL. In relation to the revised Wilson and Cleary model by Ferrans et al (2005), its components were merged from Tina's perspective; the dry mouth (symptom) effect on her simple daily activities (functional status) had negatively affected Tina's overall QoL. Moreover, some light was shed on the coping strategies adopted by Tina.

Taste disturbances (dysgeusia) associated with dry mouth in SS patients have been described in terms of an altered, bitter, and metallic taste in the mouth, or the lack of (or diminished) taste perception (Soto-Rojas and Kraus, 2002). This taste disturbance had changed with time for Jo and Ellie. Initially, there was a period when Jo "*couldn't taste anything*". With time, things improved and she instead developed taste changes to a specific food; coconut began to taste "*quite bitter"* to her. In contrast, Ellie described her experience as a gradual decrease in taste where her "*taste buds seem to*

have diminished" and she "*certainly can't taste food as well*" as before. There can also be a "*metallic sort of taste*" as a side-effect of amitriptyline (a tricyclic antidepressant) that Tracy took for her joint pain.

These details captured in qualitative interviews added depth to the understanding of how dry mouth affected the participants' taste; providing insight into how the term 'taste disturbances' was dynamic and different to each of them.

Tina articulated how her choice of foods had been affected in relation to the cyclical nature of her dry mouth experience.

Tina: "*When my condition is really bad and my mouth is inflamed (I have had numerous bouts of thrush) the only foods that I can easily tolerate without stinging are dairy based projects like cheese, yoghurt, milk. Although not the best thing to have - milkshakes are great at times when I can't eat much else. More bland type food like mashed potato, roast meats etc and vegetables are easier to eat. I cannot tolerate foods with a lot of texture i.e. crusty bread, toast etc during bad times*."

Tina explained the complex process of how dry mouth had negatively impacted on her QoL: first the periodic 'flare-ups' involved *Candida* infections, and were in relation to the episodic nature of SS; next, these resulted in intolerance to certain foods she therefore had to choose foods that were of suitable textures and acidity because "*anything with too much acid stings*"; this resulted in dietary nutrition concerns. This was an interesting observation of the dynamicity and cognition of her dry mouth experience, and the resulting coping process.

Ellie expressed her decreased tolerance to foods of higher temperatures. She could not "*drink or eat hot foods*", and they had "*to be lukewarm or cold*". This affected her QoL considerably, having to have lukewarm cups of coffee/tea in a perennially cold city. The 'spiciness' of food had an effect on some participants, but not on others. For Tina, "*curries etc have become a thing of the past as well as spicy sauces, relishes and dressings..*". On the other hand, Mike "*had medium-hot curry and beer at the Indian café*" with no trouble at all.

These findings richly demonstrated the variety of QoL impacts that resulted from dry mouth-related food choices, because of different individual preferences and tolerances to foods in terms of texture, acidity, temperature, and level of spice. The association of dry mouth and self-reported modification and avoidance of foods has been previously reported in older adults (Quandt et al., 2011). Even though it did not result in poorer dietary quality for the older people in that North Carolina study, such a possibility should not be discounted. There may be room for consideration of conjoint management of SS with a dietitian as part of the healthcare team. The participants' perspectives also provided insight into the type of dietary advice clinicians can provide for newly-diagnosed SS patients.

5.4.4.2 Dry mouth and sleep

Dry mouth has been known to cause individuals' nocturnal discomfort and to cause them to wake up at night to drink water (Soto-Rojas and Kraus, 2002; Thomson et al., 1999b). The empirical findings from the current research provided more detail about how dry mouth affected the participants' sleep. Some identified dry mouth as the sole cause of their sleep disturbance. Margaret explained that she was "*up most of the night*" because of her "*raspy throat and burning nose*". The effect of dry mouth on sleep was intermittent for Tina; one of her diary entries recorded that she "*didn't sleep well last night*", and "*kept waking up with a dry mouth*".

Others identified it to be part of the cause of their sleep interruptions. For Ellie, dry mouth affected her sleep more frequently and she "*often will wake with*" her "*tongue stuck to the roof*" of her mouth. Moreover, she found that her mouth breathing at night aggravated the situation. Mike occasionally woke up with his "*tongue stuck to teeth*" and felt like he was "*breathing in a desert*". However, he felt as if it could be coincidental because he was not "*a regular sleeper anyway, so can't just blame Sjögrens.*"

On the other hand, Jo did not perceive her sleep to be affected by her dry mouth at all even though on some nights she would "*wake up and it's as dry as (a New Zealand colloquial term to emphasise the severity of an event)*"; she would "*just get back to sleep again*."

Sleep deprivation can potentially have a profound effect on function, and it can be a work hazard (Orzeł-Gryglewska, 2010). The findings suggested that dry mouth affected the sleep of the participants, but the resultant effect on QoL was different for each of them, depending on their concurrent habits and sleep patterns. The diary approach appropriately captured these variations in time as they lived it (Bolger et al., 2003).

5.4.4.3 Dry mouth and communication

Dry mouth in SS patients is reported to be associated with voice disturbances or speech difficulties (Soto-Rojas and Kraus, 2002). Xerostomia in older adults has been associated with communication and social interaction problems (Locker, 1995; Rydholm and Strang, 2002). The current research participants found dry mouth to affect their communication because of speech and coughing. This impact appeared to be personality-dependent.

The process of dry mouth affecting speech was described by Margaret: "*if I have no water or chewing gum my words don't come out properly, no one can pick up what I am saying as my mouth and tongue are so dry they can't pronounce words properly. I couldn't communicate without water or chewing gum.*"

Dry mouth affected Margaret's articulation, preventing others from comprehending her. She therefore used water and chewing gum to improve her QoL. Tracy's description demonstrated that this impact was influenced by her introverted personality: "*I am not very good when there is a lot more people.. maybe I wouldn't join in so much, I just sit there and talk less rather than actually talk.*" Tracy revealed that she withdrew even more in social situations because of her dry mouth. On the other hand, Jo expressed her worry that she was "*annoying other people by coughing all the time*", and that it would be "*great to be able to talk to someone without coughing and spluttering*!" However, when questioned about whether it affected her communication, she replied: "*no, I just blabber on anyway*".

The negative impact of dry mouth on verbal communication, in extreme cases, may cause avoidance of social contact, resulting in loneliness (Rydholm and Strang, 2002).

An obstacle in communication can therefore have a potentially severe effect on QoL. The empirical findings suggested that personality had an influence on the impact of dry mouth on communication. This may be something for HCPs to take into consideration when assessing patients with dry mouth.

5.4.4.4 Dry mouth and the dentition

The negative effects of dry mouth on the dentition have been well-established. It may manifest as dental caries and erosion (Soto-Rojas and Kraus, 2002), resulting in poor QoL through those conditions (Turner and Ship, 2007). For the current research participants, this had additional resultant non-physical impacts, including time, finances, and emotional stress.

As discussed earlier, dry mouth has adverse effects on the dentition and the oral mucosa. Because the condition is chronic and untreatable, visiting the dentist and daily preventive measures become a time-consuming long-term commitment. Joyce observed that dry mouth management was part of her SS regime.

Joyce: "*Half an hour to wash your eyes, do your teeth properly umm put the moisturiser on, and at night it's the same. I don't feel like doing it.. I'll have to do it, high maintenance thing and tiredness sets in.*"

The findings revealed a non-tangible side of the dry mouth experience. Time is needed in order to perform necessary daily preventive routines to manage dry mouth and other complications of SS (such as dry skin and dry eyes). This became less manageable for Joyce because of the concurrent SS-related fatigue. Moreover, time was required for attending multiple dental appointments, with "*a lot needing to be done*". Mike raised that it was "*difficult getting regular time off for clinic visits*", and he had to take time off in lieu for "*that last appointment that was 3 hours long*".

These findings revealed the negative impact on QoL that time strain (related to dry mouth) had on the research participants; it was intertwined with fatigue (possibly physically and mentally) and having to take time off work. Such time off (because of chronic conditions) can affect work productivity, demonstrating the humanistic and economic burden (DiBonaventura et al., 2011) of dry mouth. Individuals have been

found to give up their work because of the time taken off for appointments because of dry mouth (Owens et al., 2014). Mike summarised the chronicity of this impact: "*I think dry mouth is, and will continue to be an ongoing issue with my dental health in general.*"

The expense of dental care has been found to escalate for those suffering from xerostomia (Folke, 2009). For many of the current research participants, this was because of multiple dental appointments (with many procedures). Tina expressed her concerns about ongoing dental costs as a result of dry mouth's detrimental effects on her teeth.

Tina: "*The condition is also costing us a great deal in dentist's bills. My dentists describes my condition as pouring acid on concrete, which is doing a lot of damage to my teeth..*"

Tina described how the adverse effects on her teeth and her finances were beyond her control because of the very nature of dry mouth (as affirmed by her dentist). Moreover, there was concern regarding the high cost of dental products (for dry mouth). Joy had to "*spend extra on dental products as well as time on using them*". Despite diligent and good oral home care, Joy found her teeth to be her "*biggest concern*" because every time she visited the dentist, there was "*always a filling*" to be done. As a result, two of the research participants (Margaret and Jo) perceived having natural teeth to be a disadvantage and wished for false teeth instead. In contrast, Joyce did not welcome the idea of a prosthesis, and perceived "*false teeth*" to be "*a bugger*"; keeping her natural teeth was her main priority. Subsequently, Joyce was "*paying out thousands every year to a dentist*". Finally, her private dentist suggested, "*Restorative dentistry at Dunedin Dental School.*" Joyce's response was: "*I haven't got 30-40 thousand for this. Now what happens? Am I a casualty of hospital cuts? Perhaps removal of all teeth*?"

Individuals have been shown to perceive the loss of teeth to be associated with compromised oral function, loss of social status and diminished self-esteem (Nordenram et al., 2013). Edentulism as a result of dry mouth can adversely affect individuals' QoL; these findings showed how this is variable because of the

participants' different health perceptions with regards to having natural teeth. This highlighted the value for clinicians to be discerning of patients' beliefs during treatment planning (Street Jr and Haidet, 2011). Moreover, Joyce brought up the influence that Government funding policies had had on her situation. Her questions revealed a sense of desperation. For some of the participants, a Government subsidy was considered to be a possible source of alleviation of their financial strain: "*biotene (saliva substitute) is very expensive – is there any subsidy?*" (Ellie) These findings validate the efforts of those lobbying for an oral health care subsidy for these SS patients with dry mouth.

Worsening dental conditions have been shown to cause worry in individuals with dry mouth (Folke, 2009). Emotional stress can also affect one's overall QoL, as measured by the SF-36 (which includes domains such as emotional role limitations and mental health). Joyce expressed her concern about the future of her teeth: "*there's twenty more years of eating which is wearing them as it is.. and more fillings*". This resulted in her feeling helpless: "*teeth to me have actually become a bit of a problem; I don't know what to do*." Joyce described the priority that the impact of dry mouth had above her other symptoms.

Joyce: "*to me it's the teeth it's a big thing with the Sjögren's because of the dry mouth, the teeth are the first thing that seems to go. Eyes and skin you can manage with drops and moisturisers, but the teeth will get holes and be pulled out.*"

Joyce's interpretation of dry mouth's impact on her was intertwined with its adverse effect on her teeth, and her inability to manage it relative to other symptoms, with a resultant sense of powerlessness. This aspect of patient cognition of disease impact is not fully conceptualized by the levels in the Wilson and Cleary model revised by Ferrans et al (2005). HCPs need to look beyond individual symptoms that are segregated by body parts, and understand the disease impact spectrum holistically.

5.4.4.5 Dry mouth and lifestyle

Dry mouth has been shown to negatively impact on the lifestyle (social and work life) of its sufferers (Folke, 2009; Owens et al., 2014). Drawing from the empirical

evidence, dry mouth impacted on the current research participants' social lives, daily cooking, exercise, and work lives.

Dry mouth has been found to be an impairment with social effects because of problems faced when eating out, which is a social event (Owens et al., 2014). For the current research participants, the challenge posed by dry mouth was in choosing foods to eat, and how this affected social situations. Tina was someone who enjoyed meeting friends for lunch; she found it a "*challenge to pick foods"* that she could easily have. On the other hand, Jo explained that "*eating out is fine*" because her friends knew (and therefore understood) her. Margaret felt judged by others, and thought that people might "*look down upon*" her because she could not eat certain foods. Her background "*depressive problem*" made her perceive dry mouth as holding a negative connotation; she thought: "*when someone doesn't have a self-esteem, their mouth dries up doesn't it*?" Margaret did not want others to "*think ahh there she is getting awfully depressed and down*." The consumption of food has a symbolic meaning in social practices; a shared meal has cultural importance in terms of intimacy and group identity (Corbett et al., 2009). Therefore, the impact of dry mouth on social eating affected the QoL of the participants in a deep manner, depending on their lifestyle, perceptions (that were related to their background medical history), and support from friends.

Cooking can be viewed by individuals primarily as a household chore, but preparing food can also be a way to please others and themselves (Daniels et al., 2012). For Tina, food was a priority and dry mouth had affected her ability to enjoy cooking, thus taking away a simple pleasure in her life.

Tina: "*I've always liked to cook and umm, you know, I love going out for meals and like most people do, like going to cafés and restaurants and things and now its, its a lot more difficult, and even just day-to-day cooking, I've got to think of you know, cooking for the family but you know, what can I have? You know, some days because I just can't have the same things I used to be able to have so its kind of taken the fun out of a lot of things*."

Tina's personality and lifestyle had a strong influence on how she perceived dry mouth (and its effects on eating out and cooking for her family) to affect her QoL. Likewise, there was a range of opinions of the impact that dry mouth had had on the exercise routines of the participants because of their different personalities and lifestyles. For Tina and PM, it was adverse, but it was not for Joy and Mike.

Tina "*I used to go to the gym five times a week and now even just walking from here to the car you know, down the corridor you know, I would need water or, its just really difficult to manage*"

Tina's dry mouth and her fatigue made it more challenging (than before her SS) for her to go to the gym. As with PM, who enjoyed tramping, she found it demanding to carry enough water and food that was moist enough to cope with her dry mouth when out in the countryside.

Meanwhile, Joy was an 86-year-old regular gym attender (3 times a week) who coped well despite having to breathe through her mouth while exercising, "*making it more dry*". The effect dry mouth had on exercise was significant because being physically active has been related to feelings of well-being and improved QoL (Spirduso and Cronin, 2001). Exercise was also adopted as a coping strategy for SS by some of the current research participants (to be discussed later).

Dry mouth has been considered to be a disability that can have a profound impact on work because of the lack of sleep, resulting in the loss of jobs for some individuals (Owens et al., 2014). The negative effect dry mouth had on the current research participants was dependent on the nature of their jobs.

Tina: "*managing at work really difficult especially if I have to do a lot of talking like today where I had three meetings. I either have to take water with me or try and manage the best I can which is difficult when you whole mouth and lips dry up and you feel like your tongue is going to get stuck on the roof of your mouth*!"

With a job that required constant talking, dry mouth was burdensome for Tina; she used water to improve her QoL. Likewise, Frieda met people because she owned a

Bed and Breakfast (B&B), and found that dry mouth made it "*embarrassing to receive B&B people*" because she often had phlegm stuck in her throat. Even though Jo did not need to talk much while working in the farm, she had to do "*a lot of digging or exertion*" and her mouth could "*get really dry*". Moreover she worked with pesticides and had her "*mask on for spraying*"; that caused her to have difficulty in breathing. Interestingly, weather-related influences were noted by Jo, who felt that her dry mouth was "*worse when the southerly (wind from the south) gets up*." Nonetheless, Jo coped well with the impact of dry mouth on her life and said "*it (dry mouth) doesn't actually make any difference, its just irritating.*"

Work can have different kinds of meaning such as the contribution to economic maintenance of one's family and self-expression of identity (Astin, 1984). These findings demonstrated the different impacts that dry mouth had had on the current research participants, depending on the nature of their occupation. The resultant impact on QoL was influenced by participants' perceptions.

5.4.5 Conclusion

Owing to the broad disease impact spectrum that impaired the current research participants in so many ways, SS was described as a '*hidden disability*'. These empirical findings reflected the fact that dry mouth was not experienced in isolation, but relative to the other SS manifestations. Understanding of how the participants perceived disease impact in relation to their personality and lifestyle generated insight into their "symptom burden". This is a concept that encompasses both the severity of the symptoms and the patient's perception of the impact of the symptoms (Cleeland, 2007). As discussed earlier, the participants had a slightly different view on what was significant to their perception of the disease impact on QoL than the concept represented in the revised Wilson and Cleary model by Ferrans et al (2005). Nonetheless, there was a strong sense of how characteristics of the individual and the environment played a part in their interpretation of how SS affected their QoL. The next section will explain how the participants have positively coped with their disease.

5.5 The positive SS (and dry mouth) coping process

5.5.1 Introduction

This chapter will examine the positive coping process demonstrated by the research participants. First, there will be a brief discussion on the importance of understanding this coping process in relation to clinical care. Next, the process of coping with SS (and dry mouth) will be explained using empirical evidence from the data set. It was a significant part of the disease experience (for the research participants) that was dynamic and personalised, and it had prominent psychosocial and cognitive components. This is illustrated in Figure 15.

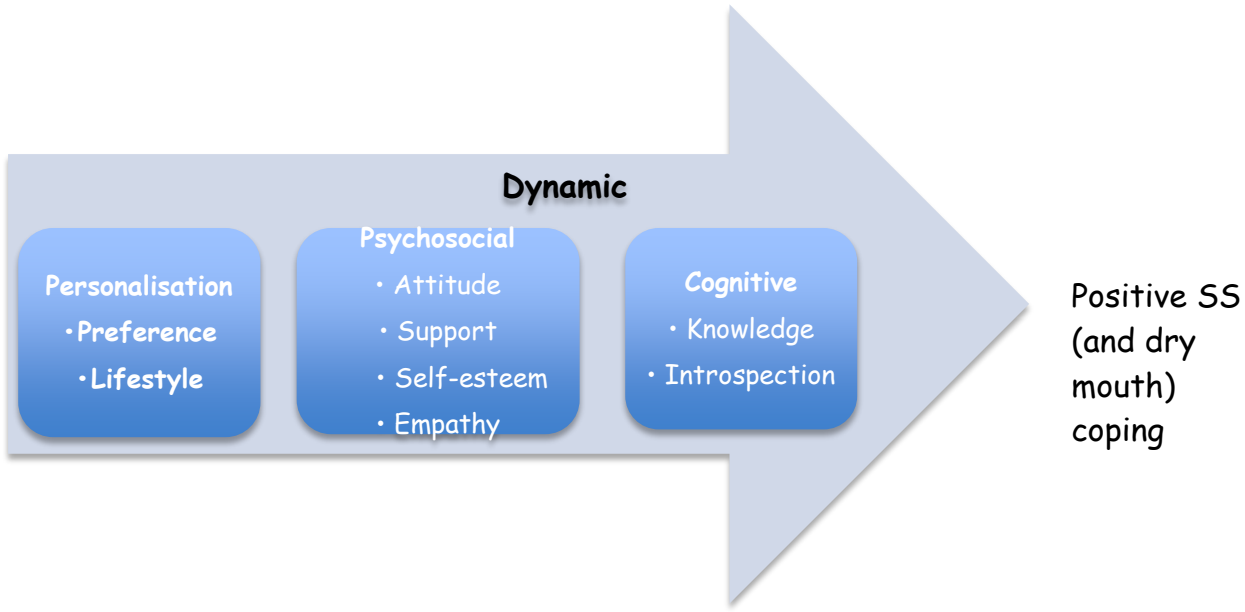


Figure 15: The positive SS (and dry mouth) coping process

The arrow represents the ongoing process that is dynamic because of the changes that occured with time and the cyclical nature of SS. The three areas observed (not in any sequence) to be significant in the participants' coping process were: (1) personalisation (depending on individual preference and lifestyle); (2) psychosocial (integrating the concepts of attitude, support, self-esteem, and empathy); and (3) cognitive (consisting of knowledge and introspection).

5.5.2 The importance of understanding the participants' coping process

Ferrans et al (2005) highlighted the importance of understanding the level 'characteristics of the individual' in their revised Wilson and Cleary model. This level interacts with other levels of the model (such as 'symptom', 'functional status', and 'overall QoL'). In the coping process for the research participants, there have been observed interactions with their symptoms and maintaining normal functional status, and also the psychosocial and emotional areas of QoL. However, it goes deeper to provide additional insight into the psychosocial and cognitive factors associated with the SS (and dry mouth) experience. Understanding the participants' positive coping process should provide insight into the degree that 'characteristics of the individual' inflence how the individual interacts with his/her SS to optimise QoL. As described earlier, patients with chronic diseases play a central role in managing their illness (Northern, 2001). This contributes to the concept of self-management education, where HCPs impart problem-solving skills to patients. These skills allow patients to identify their problems, and they provide techniques to help make decisions, take appropriate actions, and alter these actions as they encounter changes in circumstances or disease (D'Zurilla and Nezu, 1986). Self-management education supports patients to experience the best possible QoL with their chronic condition (Bodenheimer et al., 2002). When successful, it can optimise healthcare utilisation (Northern, 2001). There is no cure for SS and dry mouth; there is only long-term management, and it is therefore essential to understand patients' coping processes in order for HCPs to be a partner in the collaborative management (of the conditions). The next sections will discuss the significant components observed in the research participants' positive SS (and dry mouth) coping processes.

5.5.3 The dynamic process of coping

Coping with SS (and dry mouth) was a dynamic process for the research participants because of the changes that occurred with time and the cyclical nature of SS. In general, long illness duration has been associated with poorer outcomes in respect of the functional status of those suffering from chronic diseases; such as RA, chronic obstructive pulmonary disease or psoriasis (Scharloo et al., 1998).

This was experienced by Tracy: "*I am getting older, it is getting worse, pain; you know the joints.. is definitely getting worse and the dryness has gotten worse*."

Despite worsening symptoms, the chronicity of SS had a positive effect on illness representation for some of the participants (such as Margaret, Joy, and Jo). Initially upon diagnosis, Jo thought: "*Oh God, I'm going to have to live with a dry mouth for the rest of my life*." With hindsight, she expressed an altered illness representation: "*I've forgotten what it's like not to have it (SS) now*." There was a distinct change that occurred during the coping process, and Margaret offered insight into and explanation for this observation.

Margaret: " *I don't think about the dry mouth because I've had it for so long.. it takes time to accept what you've got wrong and why its going like that.. when I first got it, it was pretty scary.. now I'm used to all these things, its not so bad because I know what it is*."

Margaret described how time had changed her illness perception as part of her coping process. It began with an uncertainty that evoked fear, and, with gradual understanding (of her disease construct), there was acceptance. Joy brought up another aspect of coping: adjustment (to her dry mouth) that occurred with time. This is a phenomenon called "response shift", whereby people adapt and reappraise their situation (Sprangers and Schwartz, 1999).

Joy: "*You just have to adjust.. I don't even think about it now.. know what you can eat and what you can't..*"

Moreover, the cyclical nature of SS was demonstrated to affect the coping process for the research participants. Mike aptly described this aspect of his SS experience.

Mike: "*SS can behave like surf.. some quiet peaceful days and some active annoying days*."

The cyclical disease experience helped the participants to cope because they knew what to expect. Jo described how SS was "*a roller-coaster that you have to balance*",

and how she felt "*crappy*" but knew from experience that she would "*come right*" in her own time. Similarly, Tracy expressed how she knew that, if she were "*having a flare-up*", she could also "*have a remission (Tracy laughs) so it won't be so bad (Tracy laughs).*"

These findings describe the periodic nature of SS that the participants have become familiar with, and the need to optimise these ups and downs to as part of their coping strategy. Owing to changing contextual factors, adaptation to chronic illness is neither linear nor lockstep (Stanton et al., 2007). It is important for HCPs to appreciate the significance of dynamicity in the coping process (for patients). Ideally, there should be a focus on "*more education about it at the beginning when you are diagnosed with it*" (Joy). Later into the coping process, patients should be allowed to have more autonomy in the appointment scheduling (such as to have an appointment during a 'flare-up' instead of a remission). Electronic communication for disease-monitoring and advice-seeking during 'flare-ups' can also optimise healthcare utilisation. Moreover, these processes may offer explanation for the changes with time that can be observed when using quantitative HRQoL or OHRQoL measures during the trajectory of a chronic disease. The next section will go on to discuss the personalised process of coping for the current research participants.

5.5.4 The personalised coping process

This section will describe how the coping process was personalised for each participant, depending on his/her preferences and lifestyle. Different remedies were used by the research participants to manage their dry mouth. These included: (1) sipping or increasing their uptake of water; (2) using saliva-inducing products (such as pilocarpine and chewing gum) and saliva-substitutes (such as dry mouth gel); and (3) using preventive products (such as sensitive/high-fluoride toothpaste). Each participant adopted a trial-and-error approach to discover what worked best for him/her. Ellie described how she would "*just buy it (new product) and take it home and try it*", because she believed that "*everyone's different*", depending on his/her progression. During the interview, Ellie expressed her eagerness to find out more about a new product and asked: "*There's a new biotene chewing gum do you know anything about that? Is it okay? What is it like?.. I'll do whatever*!" She was an informed patient, and displayed an open and positive attitude towards possible coping

strategies. Patient empowerment through knowledge was also part of the cognitive process of positive coping (to be discussed later).

This curiosity extended to complementary and alternative products, such as supplements, acupuncture, herbal tea, and "sensitive" soap powders. Joyce said: "*I don't know what it (krill oil) will be like but it says better than flax, omega.. okay right, on I'll try it*." In the absence of a cure, the participants were hopeful of finding a remedy that would improve their functional well-being. Their preference for natural products and supplements was evident. Some particpants were creative; Frieda for example, found homemade blackcurrant juice to be effective for the dry mouth that was affecting her sleep at night. For Margaret, "*chewing gum is number one*," and she had even mastered how to keep it at the side of her denture in order to chew only when necessary (to relieve her dry mouth). At the end of the one-hour interview, she produced a hidden piece of gum from the buccal flange of her upper denture and began to chew it skillfully. Ellie described some of her preferred products for tackling different dry-mouth-associated effects (on her sleep and her lips).

Ellie: "*I sleep with water on my bedside.. and oral balance or biotene (both saliva substitute products), which I find very helpful.. lasts longer than water and is very soothing. I sometimes put it on the top of my mouth to cushion my tongue when it touches the roof of my mouth. My lips are constantly dry despite using lip balm, Vaseline is the best one I have found as it lasts longer*."

This was a vivid description of how Ellie had pinpointed her exact dry-mouth-associated issues and related remedies. Moreover, the importance of personalising remedies was shown from how the same product could produce opposite reactions in different participants. Tina did not enjoy dry mouth gel (a salivary substitute) and found that, "*five minutes down the track*" her mouth was dry again. She was "*sick of gel*" and "*sick of the taste*". This reflected (and highlighted) the contrast between short-term remedies and her long-term condition. On the other hand, Joyce expressed how the same product worked well for her.

Joyce: "*I quite like the gel.. I must admit if I don't use it.. mouth is very dry in the morning..*"

Empirical evidence showed that the curious, creative, and personalising approach adopted by the research participants produced positive coping outcomes. HCPs should take into account this insight into diverse preferences in order for individualised management planning (together with patients). This understanding should facilitate patient education for self-management in order to enhance a personalised problem-solving approach (Stanton et al., 2007). Moreover, in view of the reported potential dangers associated with unconventional remedies for chronic diseases (Southwood et al., 1990), HCPs need to be aware of the complementary and alternative medicine coping strategies adopted by their patients, and be prepared to provide safe advice when necessary.

Lifestyle considerations were also important to the research participants when personalising their coping processes. In general, they were unanimous about avoiding spicy, hard, dry or sharp foods, alcohol, caffeine, and "*sharp*" toothpaste. This was the result of their reduced tolerance to such triggers as a consequence of dry mouth. Joy enjoyed socialising with friends over a cup of tea and made it manageable by "*dunking*" her biscuits (in tea) to moisten them. Pauline had done a nutrition course in the polytechnic and adopted a health-conscious way of diet; she would therefore avoid monosodium glutamate (MSG), processed foods, coffee, and tea. Tracy led an active lifestyle and enjoyed going for daily walks; she found chewing gum to be effective in managing her dry mouth (while walking). Frieda owned a 'B&B' and spent a good amount of time maintaining the garden grounds. She coped by avoiding the sun, using a hat and scarf, and taking half-hourly breaks. Jo had a physically-demanding job on the farm and was careful "*not to bite off too much in one day*." Joyce summarised her ongoing adaptation of SS to her lifestyle in the statement: "*its a balancing thing you've just got to be aware all the time*."

Individuals with chronic conditions self-manage their illness by making decisions that are related to their daily activities (Bodenheimer et al., 2002). With experience, the participants had adapted their disease to their lifestyle in order to cope positively. HCPs need to be aware of their patients' lifestyles for the collaborative management of SS and dry mouth. The next section will go on to elaborate on the psychosocial well-being component of the participants' positive SS (and dry mouth) coping process.

5.5.5 The psychosocial aspects of the coping process

Chronic diseases carry important psychological and social consequences that demand significant adjustment (Stanton et al., 2007). The psychosocial aspects of the coping process for the research participants were evident in these areas: (1) attitude; (2) support; (3) self-esteem; and (4) empathy.

5.5.5.1 Attitude

The research participants displayed positive attitudes towards their illness, and also 'benchmarked' SS with other worse conditions. Tracy did not "*tend to sit there*" and "*dwell on it*". Instead, she chose to "*get on with it*" because she had two young school-aged children and could not afford to be unwell. This was especially so because she had no immediate family support (they were back in the UK). Moreover, a previous back injury placed the discomfort from joint pain at a relatively manageable level. The interplay of the factors (1) family support, (2) previous health experience and (3) life stage was evident in Tracy's strong and positive coping attitude. Jo added that positive thinking was important because "*if you start thinking down that's where you will head quickly*." In reality, this was not always possible, as demonstrated by Tina who "*had a couple of days of feeling sorry*" for herself. In relation to a positive attitude towards the greater risk for lymphoma, Frieda acknowledged it but claimed that one "*mustn't think about it*." A sense of power and control over the disease was expressed by Mike who "*decided early on*" that SS "*wasn't going to dictate*" how he lived his life; he was "*determined to be positive and continue with usual routines*." Joy was an 86-year-old who went to the gym three times a week, and she wisely summarised her positive coping attitude in the statement below.

Joy: "*You've got two options, you either accept it and get on with it and change, make changes or you can give up..*"

These were examples of positive mindsets as part of a thriving management style (Knafl et al., 1996). Another way of positive thinking was to take a step back and view SS relative to other worse conditions that were experienced by other members of the family or the public. Joyce expressed that, "*it (SS) is something you live with, there are people with a hundred times worse than I am*". Jo, who cared for her

daughter with multiple sclerosis (a debilitating motor neurone disease), felt that "*other people got more complaints*" because she was "*not crippled.. haven't got a motor neurone (disease), haven't got cancer*." This form of comparing one's current circumstances with a hypothetical worse situation is a way of using positive illusions to cope with adversities (Taylor and Armor, 1996). These positive attitudes adopted during the coping process for the research participants may explain why they all coped so well with SS and dry mouth. HCPs may need to recognise such positive attributes and identify those patients who do not exhibit them, because they may need more help.

5.5.5.2 Support

The support received by the research participants during their coping process helped in maintaining their psychosocial well-being. This came from family, friends, people met during work, and HCPs (as discussed earlier). Family members were often their core support unit; different family dynamics were expressed in the variety of interactions observed. Mike had a supportive relationship with his wife.

Mike: "*Couldn't wait to get home and tell my wife (SS diagnosis) and having her support helped lots*."

He found confiding in his wife to be comforting right from the beginning of his SS journey. On the other hand, Mike preferred not to share (his SS experience) with his extended family and acquaintances because he did not want to be reminded of the downside of things. This support (from the spouse) had different levels and depended on the nature of the participants' relationships with their spouses. For example, Margaret described a dependency on her husband Ian for reassurance. This was also evident during the interview, when she requested that he sat in, and she occasionally cross-checked her responses with him. Ellie, on the other hand, was unable to confide in her husband, whom she described as someone who "*does not know what I am thinking half the time*."

Friends had a positive support effect on the coping process for some of the participants. Jo shared that she would: "*make sure I'm with people that make me*

laugh". From a different perspective, Ellie found having people around her to be a positive distraction from thinking about SS.

Ellie: "*I'm much better around people you know.. when you're on your own you think about yourself, its not healthy*."

Likewise, at the workplace, colleagues and clients provided a form of support. PM was a school teacher who found it helpful when the parents showed concern by asking her "*if there is anything they can do*." Similarly, Mike drew comfort from his colleagues, who kept an eye on him and remained supportive.

No individual can exist alone; the environment consisting of family, friends, and HCPs provides support for psychosocial well-being (Arora et al., 2007). Seeking social support is an active coping approach with positive outcomes (Stanton et al., 2007). Impaired HRQoL and emotional distress have been found to be prevalent among patients with chronic diseases; thus, a complete evaluation of psychosocial status was recommended to be included in the clinical treatment of patients (Larsson et al., 2008). The findings provided insight into types of support needed for the participants to cope positively. Therefore, understanding the social circle of patients can be important in assessing their coping processes.

The public awareness of SS was important to a few of the research participants. Interestingly, this was reflected in their delight at knowing that the celebrity tennis star Venus Williams also had SS. This allowed others to identify with their disease, and provided a language to discuss SS with friends (for a sense for support). Tina eloquently articulated what this meant for her.

Tina: "*Its different when you say I've got arthritis.. people got some understanding.. most people never heard of it (SS).. the only thing in my favour is that Venus Williams has got it.. often I use that (laughter from interviewer).. Venus Williams keeps pulling out of tennis tournaments, well she's got it..*"

The awareness of the disease helped others to identify with Tina's disease; she felt understood. Similarly, Frieda felt that "*Venus helped terrifically to make it (SS)*

known". Moreover, Mike could identify with the fatigue that Venus experienced, and Joyce "*couldn't believe it*" when she watched SS being mentioned on a TV programme. A sense of loneliness and lack of support has been found in those suffering from a less-known disease (Dellve et al., 2006). Increased public knowledge and awareness of SS therefore allowed it to be less of a "*hidden disability*". These findings reflected an interesting form of support that facilitated positive SS coping by the participants. Providing such an information source with newly-diagnosed SS patients may aid in their acceptance and coping process.

5.5.5.3 Self-esteem

Self-esteem was shown to be an important component in the psychosocial coping of SS. The research participants used activities as part of their SS coping process in order to maintain their functional well-being. Joyce found it important to keep working because of her fear of becoming less mobile.

Joyce: "*I'm not going to stop working because I'm a bit frightened if I stop work my body will just stiffen up*.."

Concurrently, activities provided positive effects on the self-esteem of some participants, highlighting the psychosocial aspect of coping. For example, Mike started jogging regularly because he enjoyed the "*normality of daily exercise*", and it helped him to "*feel better*" and "*resist feelings of self-pity*". The types of activities participants used for coping (with SS) were dependent on their personalities and lifestyles. Margaret was a homekeeper and she found household chores to have an uplifting effect on her self-esteem.

Margaret: "*Luxed the house, this made me feel a lot better about myself.*"

Similarly, Jo (who worked in the farm) found fulfillment and motivation in mowing the lawn. It would give her a sense of accomplishment and a "*that bit of life up to keep going*". These findings emphasise the importance of the sense of self-worth and accomplishment that was derived from performing simple daily activities. Likewise, perceived self-efficacy has been found to play a role in mediating health outcomes for

people with chronic arthritis (Lorig et al., 1989). HCPs should recognise the psychosocial benefits that can be achieved by maintaining normality (Knafl et al., 1996).

Some participants found the research process to be an altruistic act with positive influences on their self-esteem. Joyce "*enjoyed it actually*" and was happy that the researchers were "*able to get something from it.*" Margaret revealed that the research process was therapeutic because she could confide in researchers who was interested in her experience, and that being recruited (as a research participant) had had a positive effect on her self-esteem.

Margaret: "*Got someone I can talk to.. there are people out there that are understanding and they want to know about it.. I feel good I was chosen to do this, I feel really good about that*!"

Potential therapeutic effects on emotions have been observed in participants who took part in UK research involving qualitative interviews (Haynes, 2006). The therapeutic effects associated with improving self-esteem from the research process were an interesting finding. Could the research process itself be therapeutic? Further research may be required on the effect of qualitative research on coping with chronic diseases.

5.5.5.4 Empathy

The research participants were able to empathise with others because of their individual disease experiences. A few of them expressed how they hoped that dry mouth and SS would not negatively affect others in the same way as themselves. Frieda had had to struggle to be acknowledged by her HCPs and stated: "*I want it to be known by women because they don't know, like I didn't know what it was.. struggle and struggle.*" Joyce perceived early diagnosis to be important to prevent dental problems "*because teeth are so important*". Joy was a self-sufficient 86-year-old who owned a farm and was aware of the financial burden of dry mouth.

Joy: "*I would like to see some form of compensation.. elderly people who are on benefit.. no way that would cover..*"

Empathy can be a coping mechanism because it is a form of helping others, and positively impacts on the physical and psychosocial well-being of the provider (Schwartz and Sendor, 1999). These finding demonstrated a positive coping approach with participants actively trying to identify benefits in their experience (Stanton et al., 2007). HCPs can perhaps guide patients towards such perspectives for positive coping.

5.5.6 The cognitive aspect of the coping process

Individuals can use various cognitive strategies to counteract the negative effect of illness on their well-being (de Ridder et al., 2008). Information-seeking has been found to have a salubrious effect on adjustment in the process of coping with chronic diseases (such as RA and cancer) (Felton and Revenson, 1984). In the current study, the acquiring of knowledge (about SS) helped the research participants in the cognition of their condition. Some of it was intimidating, and some was helpful. The Internet was a source of knowledge for 9 out of the 10 research participants (or their families). When the information was presented as 'worst case scenarios', it was found to be intimidating.

Ellie: "*Some of the things on the computer they are pretty horrific, you know you get a lot of the worst case scenario things and so I really thought, well I don't know that I actually really need to know all just at this point in time..*"

It was observed that the participants turned to the Internet for information in order to understand their condition around the time of diagnosis, and this was part of their coping process. HCPs should perhaps inform patients of suitable information sources such as reliable websites (including social media sites such as the SS foundation Facebook group), books, or societies.

Margaret: "*Look forward to them (SS society's pamphlet) coming.. I've got as much information now.. I know I can accept what I've got. I mean it takes time to accept what you've got wrong, and why its going like that.*"
Margaret appreciated the information provided in pamphlets because it allowed her to go through the coping process by accepting her SS identity and understanding her symptoms. The acceptance of a chronic disease has been shown to improve functional

status and overall QoL (McCracken and Eccleston, 2003). Moreover, PM mentioned that the knowledge she acquired from textbooks and literature (because of her health science education background) had helped her to know what she was "*up against*" and to "*cope better*". This sense of empowerment can aid in disease coping and improving health outcomes (Panja et al., 2005).

Some of the research participants found the diary process to be reflective and therapeutic. Interestingly, those who described not wanting to "*dwell on things*" actually found it "*quite good to think about what happened (through the diary entries)*" (Jo). Tina offered an explanation on how the introspection helped her to deal with some SS-related emotions.

Tina: "*Given me the opportunity.. tried to put it aside and its make me deal with it.. get some feelings out.. kinda packed it away*."

Avoidance and inhibition of emotions has been associated with maladaptive outcomes such as an increase in disease occurrence and risk of disease progression (de Ridder et al., 2008). Some of the research participants suggested keeping their diaries for future use. This suggested that ongoing introspection may be helpful throughout the coping process. Joy had "*saved them on the computer*" so she "*can read them over*". Ellie remarked that it was "*good to have it written down*" to "*follow a pattern*", and that she had kept "*a copy for daughters*" in hope that "*it will help*" them to understand SS. HCPs should perhaps encourage patients to use diaries and reflect on SS to enhance their coping processes.

5.5.7 Conclusion

In conclusion, the empirical findings from the current research demonstrated the aspects of a positive coping process. The process has been shown to be dynamic and personalised, with emphasis on the psychosocial and cognitive aspect of coping. This model of coping is original, and future research is required to further test it on SS or other chronic diseases. In relation to the revised Wilson and Cleary model by Ferrans et al (2005), these findings provide some insight into how the participants interact with SS and dry mouth.

HCPs are well trained in the physical and biological aspects of SS management. In view of the lack of treatment, this insight into the participants' coping processes may encourage more effective collaborative treatment and patient education for self-management of the disease (Bodenheimer et al., 2002).

Chapter 6 Discussion

6.1 Introduction

This chapter will discuss the findings and meaning of the current research. First, there will be an overview of the strengths and limitations of the study. This will then be followed by a brief summary of the existing literature, in order to describe the contributions of the findings in the context of: (1) understanding patients' perspectives of the lived experience of dry mouth in SS; (2) practical clinical applications in managing dry mouth in SS sufferers; and (3) theoretical contributions to understanding HRQoL and OHRQoL. There will then be consideration of the recommended future directions for the research findings.

6.2 Limitations

The limitations of the study lie in the areas of: (1) the "undiscovered" knowledge from the patients who did not participate; (2) the researcher's influence on the data; and (3) the possibility that participants may have tended to provide information that was biased towards what they perceived to be required of them (also known as the "Hawthorne" effect) (Adair, 1984).

6.2.1 The patients who did not participate in the study

The purposive sampling approach was used to identify a specific group of people, those who have dry mouth and SS, because they have been constructed to represent the subjective knowledge of the lived experience of dry mouth for SS patients. However, not all of the participants who had been approached agreed to participate in the research. Moreover, not all of the participants who consented followed through with the data collection, and there were five potential participants who did not consent. Those individuals were unable to commit because of reasons that included: (1) moving house; (2) a disinterest in doing diaries and interviews; (3) workload; (4) having a sister who had recently been diagnosed with cancer; and (5) not having any dry mouth symptoms despite having SS ('perfectly fine, and hopes things stay this way'). There were four participants who initially consented but did not follow through with the data collection. The reasons for this included: (a) having to go through a separation; (b) being unwell because of continuous SLE (an autoimmune condition associated with secondary SS) flare-ups; (c) travelling, followed by illness, and then

becoming uncontactable; and (d) becoming diagnosed with breast cancer. There was one participant, Tracy, who initially consented but had to delay starting on the diary entries for almost a year because she was busy with the relocation of her home. Tracy eventually became a cooperative participant after she had settled into her new home.

Nonetheless, the current research represented the fullest analysis of the dry mouth experience in SS patients that was possible to be captured, and the absence of all the invited participants may have reflected a limitation in the opinions expressed–'the missing voices'. The information that could have been provided by the potential participants and those who discontinued is just as valuable as those who did participate; however, the nature of that "undiscovered" information and knowledge is unknown. There is a possibility that those who coped more positively were more willing to talk about their disease, hence, creating a bias in terms of knowledge. However, this is just speculation, and there is no way to gain any confirmatory information from the individuals who did not wish to participate in the research. It might be possible to gain such information if another attempt to involve participation was made at a more appropriate time. For example, when Tracy was given sufficient time to settle into a normal lifestyle after a hectic period of moving homes, she was able to participate in the research. She was the last participant recruited (to check for redundancy of themes), and the data collected from her diaries and interviews proved to be in line with the rest of the themes. Other participants were offered, but have declined such flexibility in time frame for data collection.

In addition, the reasons proposed by those who did not participate in the research were important. They revealed the influence of personal preferences (not wanting to write diaries or participate in interviews), life stages, having another priority (such as a more severe illness), or a diseased family member. These reiterate the importance of the influence of the characteristics of the individual and the environment on disease experience (as discussed in the results and commentary chapters). It is noteworthy that the insight gained in interpretive work is subjective to the sample and situation of the study—if the work were repeated in a different time and context, it may generate different insight and have different emphases.

6.2.2 The influence of the researcher on the data

In qualitative research, the researcher is regarded as the research instrument. Therefore, it is inevitable that pure objective observation is impossible. There was the potential of presenting the data in a narrative that took advantage of the reader's trust in the integrity of the researcher, because data extraction in qualitative research can be used selectively to present an argument that is rhetorically convincing but scientifically incomplete (Mays and Pope, 1995). However, this is a double-edged sword, because the strength of qualitative research is in the same role which the researcher plays in data-gathering; that individual's skills in listening, observing, and understanding are crucial to understanding the knowledge to be gained (Carson et al., 2001). In the current research, the methods used to overcome the potentially biased influence of the researcher on the data included: (1) maintaining a purely researcher-participant relationship (instead of a clinician/researcher-participant relationship); (2) using the revised Wilson and Cleary model by Ferrans et al (2005) as a reference framework model; and (3) employing inter-researcher peer coding in ensuring the trustworthiness of the data. The methods used to prevent the presentation of the data as a mere 'convincing' narrative were to make available the full transcript of the raw data on a computer disk, and to present detailed commentaries in the Results and Commentary chapters. All of these strategies seek to create an account of method and data that can stand independently, so that another trained researcher could analyse the same data in the same way and come to essentially the same conclusions, and to produce a plausible and coherent explanation of the phenomenon under scrutiny (Mays and Pope, 1995). The primary researcher was a novice in this field, and this may have influenced the full richness or integrity of the data collection and analysis through her less experienced interviewing techniques. However, this limitation was foreseen, and the first two interviews were performed under the guidance of an experienced supervisor (SF). Every researcher has to start somewhere, and the researcher's technique has been honed from this study.

6.2.3 Participants providing data that they thought was required of them

There may be a possibility that the participants in the research tended to provide data that they perceived to be required of them. This was mentioned in some questions in the diaries or interviews such as *"I wasn't too sure I was giving you the right*

information..” (Jo), or when Ellie mentioned “*I don’t know if its (asthma history) relevant*..”. This has been described as the “Hawthorne effect”, referring to the tendency of participants to modify their behavior because of their awareness of being under study (Adair, 1984). More recently, this has also been described as the social desirability bias—referring to the tendency of research subjects to give socially desirable responses instead of choosing responses that are reflective of their true feelings (Grimm, 2010). This bias was unavoidable because of the expectations that were involved in human interaction, and it was, in fact, that same human interaction that elicited patients’ perspectives in the current research. This limitation was addressed by: (1) embracing the subjectivity of the participants; (2) reassuring the participants that there are no ‘correct’ answers and that any aspect of their experience is relevant (not privileging medical systemic analysis but the holistic experience of the patients); and (3) triangulation of techniques (to be discussed in the next section). During the follow-up interviews, the interviewer reassured the participants that “*everything is relevant*”. This was a technique-sensitive aspect of qualitative interviewing that involved the human emotional quotient. Despite being a novice, the primary researcher had addressed this aspect. Part of the researcher training (by SF) was in developing the skill in ‘how to create a safe environment for participants where they could disclose fully’. This was evident from a comment made by PM, which implied that she had perceived (and was grateful) that the interview was an area where she could “*share freely without being judged*”. Moreover, during the consent process, there was a clear discussion and emphasis on the purpose of the research, to understand the ‘lived experience’ of dry mouth from the personal perspectives (of the SS patients). An example of the statement used during the consent process (Appendix 9) was: “*we want to know what it means for you to have dry mouth in your daily life*.”

6.3 Strengths

The strengths of the current research were in its systematic research design, data collection and analysis (as described in the chapter on methods). These methods enhanced the trustworthiness and transferability (also known as rigour) of the research.

6.3.1 Data collection, transcription, and analysis by the primary researcher

The data collection was undertaken after an initial meet-up or phone call, in order to establish rapport between the primary researcher and each participant. This improved the researcher-participant level of comfort, and enhanced the participants' ability to share their experiences freely. After direct observation of behaviour and listening to the interviews, the transcription (of the collected data) was performed by the primary researcher. This allowed for the inclusion of any non-verbal communication for consideration during the interpretation of the meaning of the data. Moreover, transcription by the researcher facilitated her familiarisation with and immersion in the entire data set by repeated reading in an active way to search for meanings and patterns (Braun and Clarke, 2006). Transcription has been seen as a key phase of data analysis within interpretative qualitative methodology (Bird, 2005), whereby meanings are created, rather than simply being a mechanical process of putting spoken sounds on paper (Lapadat and Lindsay, 1999).

6.3.2 Trustworthiness

The trustworthiness of the study was ensured by the analysis being conducted by a group (also known as 'peer coding'), with the primary researcher and two supervisors meeting regularly (every 1-2 weeks) to review the initial codes from the transcribed data. This was followed by a detailed comparison of the different possible interpretations of the data. Subsequently, multiple mind-maps and discussions were employed in order to establish the different themes. The nature of the research also allowed for flexibility and reinforcement of the themes throughout the research (when necessary), based on the development of the empirical evidence. That process allowed for a more complete analysis of the data that was led by the source of knowledge (the experiences of the research participants). Moreover, triangulation was established by using two different data collection methods to answer the same research question; these were the diary entries and the interviews, hence improving the trustworthiness of this qualitative research (Mays and Pope, 1995).

6.3.3 Transferability of the research

The transferability of a qualitative research project is the generalisability of the study findings to other situations and contexts. Usually, each qualitative study has its own premises and participants, and the findings are generally not transferable (Folke,

2009). Although purposive sampling was used, the findings from this research were (to an extent) transferable to another two populations with dry mouth in the UK and Sweden. A recently published UK qualitative study by Owens et al (2014) in Sheffield aimed to explore people's subjective experiences of dry mouth. In that study, data were collected from interviews with 18 participants; of those, 6 were recruited from the British Sjögren's Syndrome Association (BSSA), and the remaining 12 participants had reported dry mouth with no additional co-morbid conditions. Dry mouth was found to have impairment effects on speech, sleeping, working, eating and self-care. The social dimensions of the impairment were explored, and dry mouth was viewed as a disability as well as an impairment (Owens et al., 2014). Although that study approach used social models to define the borders of dry mouth as a public disability or a private impairment, the participants' perspectives in the current research were in line with the experiences of those in that UK sample. The Swedish qualitative study by Folke (2009) aimed to promote a better understanding of (and a greater empathy for) individuals suffering from xerostomia. There were 15 participants who were interviewed, of whom none was mentioned to have SS, but 4 of them had had radiation to the head and neck for cancer therapy. The findings of that study identified xerostomia as an '*aggravating misery*' and revealed the participants' main concerns to be '*professional consultation*', '*search for affirmation*' and '*social withdrawal*'. Despite the different population, there were some overlaps in the content of the themes from that Swedish study and the present study. It is therefore appropriate and important to acknowledge an existing degree of transferability of the current research findings to other dry mouth sufferers, suggesting that insight can also be provided into the dry mouth experience of people with other systemic conditions.

6.4 Research contribution

The literature review has established using (both qualitative and quantitative methods) that dry mouth and SS both negatively impact upon the QoL of their sufferers. The revised Wilson and Cleary model (2005) is a conceptual model that aims to describe the relationship among clinical variables, with influence from the characteristics of the individual and the environment, in order to improve the conceptual understanding of QoL (Ferrans et al., 2005). Thus far, this model has been applied to many different conditions to understand their related HRQoL by employing different indices for

measuring the clinical variables. However, there have been limitations in the conceptualisation of: (1) disease experience affected by the characteristics of the individual and the environment; (2) the changes in patient experiences with time; and (3) the effect of treatment (as discussed in the chapter on QoL) (Bakas et al., 2012; Baker et al., 2007). The interpretivist paradigm behind the qualitative approach seeks rich insight into disease experience. There has not been any research undertaken that utilises this approach to: (a) examine the dry mouth experience amid the range of SS manifestations; (b) reveal the adverse impacts of these experiences on QoL; or (c) provide a patient's perspective to the revised Wilson and Cleary model (2005). The research question was: what is the ongoing lived experience of dry mouth for SS patients, and how does each individual cope with dry mouth amid a spectrum of symptoms (with consideration to individual and environmental characteristics)?

The themes from the research findings included: (1) the journey to diagnosis; (2) the disease impact spectrum; (3) interactions with HCPs; and (4) the positive SS (and dry mouth) coping process. Findings from the current research have contributed to knowledge by complementing and enriching the existing knowledge that is represented in the literature. They agreed with the current knowledge that dry mouth and SS adversely affects the QoL of individuals. Similar to the outcome when the Wilson and Cleary model (1995) was applied to dry mouth (Baker et al., 2007), the findings also highlighted the complexity of relationships among the different levels (of clinical and non-clinical variables) in the Wilson and Cleary model revised by Ferrans et al (2005). Confirming the Sheffield study's finding that dry mouth is both an impairment and a disability (Owens et al., 2014), dry mouth in the SS sufferers in the current research had both private (that represented impairment) and public effects (that represented disability). The findings supported the negative impact of dry mouth and SS on QoL, and added significant depth to the existing knowledge with detailed lived experiences to substantiate the existing quantitative data, and supported and extended the existing qualitative findings from the UK and Sweden. The contribution to knowledge lies in three areas: (1) understanding patients' perspectives of the lived experience of dry mouth in SS; (2) practical clinical applications in managing dry mouth in SS sufferers; and (3) theoretical contributions to understanding HRQoL and OHRQoL. These will each be discussed.

6.4.1 Contribution to understanding patients' experience

The current study largely addressed the how question—How are patients affected? How do they cope? How do they articulate their experience? The journey to diagnosis was a significant process for the research participants; it began with symptom interpretation. There were a multitude of different combinations of initial symptoms that affected the participants, and their interpretations of their initial symptoms were intertwined with their effects on functional status (such as work and daily activities). This led them to seek medical advice, making the symptom interpretation open to HCPs; this then led to heterogeneous routes towards diagnosis (depending on HCPs' level of awareness and acknowledgement). The hurdles (or lack thereof) experienced by the research participants also provided insight into how to shorten the length of time taken to diagnosis (and make the route more direct because this created a significantly more positive disease experience for the participants). The interactions with HCPs were significant to the participants because they had an effect on QoL. The "active" and "passive" patients had different views in respect of their HCP expectations and roles. The important aspects of these interactions included: (1) the referral to specialists; (2) prescription medications; (3) patient education; (4) HCP accessibility; (5) financial aid; and (6) empathy. Insight into patient perceptions of these interactions has important clinical applications to the management of dry mouth and SS (to be discussed in the next section). The theme 'Disease impact spectrum' explored the participants' perspectives of how dry mouth and SS impacted on QoL. Dry mouth had physical and intangible effects in many areas of the participants' lives (depending on their individual and environmental characteristics). Moreover, the dry mouth experience was a dynamic one that was relative to the other SS manifestations. The transferability of the research findings suggest that this knowledge is very likely to be applicable to other groups of individuals suffering from dry mouth as a result of other factors (such as polypharmacy or radiation therapy). The impact on QoL led to a positive coping process that was dynamic and personalised for patients, with a focus on the psychosocial and cognitive aspects of coping.

6.4.2 Contribution to practical clinical applications

Understanding the participants' perspectives of disease yielded insight into clinical applications for the management of dry mouth and SS. The mean SXI score of 21

elicited the severity of the participants' dry mouth experience. This was more than twice the mean SXI score found in several other older populations (as described in the SXI results score chapter). The patients' experiences provided clinicians with insight into what that high SXI score actually means in daily life. In 'the journey to diagnosis', GPs played a key role in directing the diagnosis of SS because they were the first port of call. It is therefore important to improve GPs' awareness of SS manifestations (such as dry mouth). This can be achieved through continuing professional development involving collaboration between dental and medical colleagues. Furthermore, it was important for HCPs to recognise that patients sought advice because of symptoms that had affected their functional status and QoL. HCPs' levels of awareness and acknowledgement of and toward patients' symptoms played a significant role in the route to diagnosis (the heterogeneity of which was reflected in the range of time taken from 1 to 28 years). Moreover, the importance of the time around diagnosis itself was not to be underestimated. It was a crucial and vulnerable period for participants, optimal for the incorporation of self-management education and collaborative care (Bodenheimer et al., 2002). The theme 'Interactions with HCPs' provided insight into the aspects that the participants' described to be pertinent in the patient-physician relationship. The participants tended toward two patient roles (the more "active" or "passive") and these affected their expectations of HCPs. For example, those who were more "active" required detailed information about the side-effects of their medications and their long-term disease management plan. By contrast, those who were more "passive" were compliant toward the prescribed treatment and management. It would be useful for HCPs to increase patient involvement, because active patient participation in medical decision-making has been shown to improve health outcomes (Greenfield et al., 1988). Moreover, it is important for HCPs to understand patients' preferences, personality, lifestyle, and perceptions, in order to begin a partnership and dialogue in managing dry mouth and SS. There has been some description of interaction with HCPs in a negative light as '*lack of empathy and professional commitment*' with regards to the dry mouth experience by the participants interviewed by Folke (Folke, 2009). However, this seems rather pessimistic. The findings of the current study showed a range of both positive and negative experiences with HCPs, represented in the relationship between 'characteristics of the individual' and 'characteristics of the environment' in the revised Wilson and Cleary model by Ferrans et al (2005). This therefore allowed a

more practical application of the model in the clinical settings, because HCPs can visualise their impact (as part of the characteristics of the environment) on patients' QoL. The theme 'Disease impact spectrum' reflected how the participants perceived the impact of dry mouth (including those on intangible areas of their lives) as a subset of the other consequences of SS. This patient perspective is opposite to the usual view adopted by specialists in various departments, who tend to focus on the 'area of the body' that has been referred to them. Other practical clinical implications provided were in terms of how patients can be adversely affected by the effect of treatment (such as having their self-esteem adversely affected by post-operative biopsy site numbness). Considering the participants' experience of the impact of dry mouth on food choices and nutrition, it would be prudent to provide dietitians' advice for newly-diagnosed SS patients, in order to facilitate the coping process. When considering the disease's impact on QoL, HCPs need to be aware of their patient as an individual, and also to understand the environment in which he/she lives. The coping process was dynamic for the participants, and this knowledge can assist HCPs in understanding how patients deal with their dry mouth and SS. All of the research participants adopted positive mindsets towards SS, but with some coping better than others. Identifying factors that improve the coping process will aid in achieving the collaborative care and self-management education that is part of the preferred patient-physician partnership in dealing with chronic diseases (Bodenheimer et al., 2002).

6.4.3 Contributions to theory

There were many aspects of the participants' dry mouth and SS experience that overlapped with some of the components of the revised Wilson and Cleary model (2005). There has been added knowledge to the conceptualisation of: (1) disease experience affected by the characteristics of the individual and the environment; (2) the changes in patient experiences with time (with emphasis on the time of diagnosis); and (3) the effect of treatment. The theme 'Interactions with HCPs' added insight into the bidirectional relationship between the components 'characteristics of the individual' and 'characteristics of the environment' in that model. Nonetheless, the pertinent patterns in the participants' 'the journey to diagnosis' and 'Positive SS (and dry mouth) coping process' were not completely described by the revised Wilson and Cleary model (2005), and original models were drawn from the empirical findings. The theoretical contribution from the theme 'Disease impact spectrum' was in the re-

conceptualisation of the revised Wilson and Cleary model (2005) from the patients' perspective. This is a significant contribution because no other study has ever provided a qualitative perspective to this model. Moreover, it is noteworthy because the very concept of HRQoL and OHRQoL is based on patients' perspectives of disease impact. This 'revised revised' Wilson and Cleary model (as shown in the chapter on disease impact spectrum) is a result of the underpinning interpretive perspective that provided insight into the patient's view of QoL.

6.5 Future directions

In this section, there will be a discussion of the possible future research directions using the empirical findings from the current research. These include: (1) further qualitative studies to test the revised Wilson and Cleary model (2005) and the original models derived from the data; (2) clinical trials; (3) adding to current HRQoL/OHRQoL measures; and (4) deriving an original SS-specific dry mouth inventory. Advocacy for funding for dry mouth sufferers has been validated by the research findings, and this should be provided to improve the QoL for these individuals.

This research has shown how an interpretivist approach to a conceptual model of HRQoL can be effective in illustrating some of the relationships between its levels, and hence addressing the 'how question'. More research in this direction in different samples, cultures, or geographic locations can further test the conceptual model and provide more insight into HRQoL and OHRQoL, which are dynamic and complex concepts. The relevance of the 'biological function' level in the revised Wilson and Cleary model in understanding patients' perspectives of HRQoL and OHRQoL may need to be further tested with more qualitative research. The models on 'the journey to diagnosis' and 'the positive SS (and dry mouth) coping process' that were derived from the empirical findings are a proposed extension of an existing model, and need further testing in different ways to demonstrate value, and to explore their transferability to other samples (or illnesses).

The subjective component of dry mouth means that the insight from this research is applicable in understanding trial-related changes in the dry mouth (and SS) symptom burden, or for the comparison of symptom burden between intervention arms in

clinical trials, and may provide sufficient self-reported data for clinical trial consumers to make treatment choices or to evaluate new therapies (Cleeland, 2007).

Moreover, the findings from the current research support the significance of the influence of characteristics of the individual and the environment, and of patients' symptom interpretation on HRQoL and OHRQoL. These domains should be included in the current measures for a more accurate assessment of HRQoL/OHRQoL. An example of an item could be, "How would you rate dry mouth amidst your other symptoms?" or "Has your dry mouth affected your interaction with your family?" or "Has your HCP been helpful in co-managing your dry mouth with you?". Moreover, it is important to understand that QoL is a dynamic perception that is affected by the coping process. This is an important consideration when assessing changes in HRQoL/OHRQoL measure scores with time. These will allow a more realistic consideration of patients' perception of the impact of dry mouth on their QoL.

The findings have described in detail the dry mouth experience of SS patients. The participants' experiences revealed a very unique relationship with SS (and its other manifestations). The pertinent points may be compiled into an original SS-specific dry mouth inventory. Given the severity of dry mouth as measured by the SXI score, this group of patients definitely requires more understanding and assessment of their dry mouth status. It is likely that existing measures (such as the SXI) do not have sufficient content validity for routine use in SS, but that needs to be explored quantitative research.

This understanding of HRQoL and OHRQoL is also significant to health planning and policy development. There is an increasing focus on patient-based outcomes in an environment of ever-greater demands on scarce oral health resources, meaning that identification of the conditions with the most potential to compromise OHRQoL is a matter of some urgency (Locker, 2003a). Dry mouth is such a significant oral impairment, a 'hidden disability', and a costly ongoing condition for the participants in this research (and in other population samples), and so specific healthcare funding and (perhaps) compensation is definitely indicated.

6.6 Conclusion

In conclusion, the current research made a major contribution to knowledge in this area, but the limitations inherent within the study were recognised (as described). It had addressed an important gap in the existing knowledge, and had made significant contributions to the general body of knowledge in improving the understanding of SS patients' dry mouth experience, and hence providing practical clinical applications of this insight. In terms of theoretical contribution, the current study has examined two very important domains of HRQoL/OHRQoL: the role of characteristics of the individual and the environment in relation to the daily experience of dry mouth by these SS patients. This was the first study to put a qualitative approach to the revised Wilson and Cleary model by Ferrans et al (2005), with a resulting reconceptualisation of patients' perspectives of the model. Tentative theoretical models in relation to 'the journey to diagnosis' and 'the positive SS (and dry mouth) coping process' have been drawn from the findings, and require further exploration to establish their possible contribution to knowledge. More research in future is required to advance the patient, clinical and theoretical understanding of HRQoL and OHRQoL in SS sufferers.

Chapter 7 Overview

This thesis was a presentation of the current study titled: *Living with dry mouth – Sjögren's patients' perspectives*. To begin with, the literature review individually explored the conditions of dry mouth and SS. Dry mouth is a complex condition that is relatively prevalent. There has been research to quantify and understand the aetiology of dry mouth. Dry mouth negatively impacts upon sufferers' OHRQoL and is one of primary findings in SS. Moreover, there is no cure for it. SS is a relatively uncommon (but not rare) disease that has a complicated network of aetiology and pathophysiology that still requires more research. Owing to its broad clinical manifestations, the diagnosis of SS is often delayed. Both dry mouth and SS have severe and adverse effects on the QoL of patients.

There is now abundant literature on the measurable negative impact of dry mouth and SS on the QoL of patients. However, patients' perspectives on disease impact go beyond HRQoL and OHRQoL measures. The revised Wilson and Cleary model (2005) conceptualises the various (clinical and non-clinical) levels that can affect HRQoL/OHRQoL, but is unable to fully describe the complex patient experience of dry mouth and SS. This gap in knowledge is best addressed using a different research paradigm (the interpretivist) that is behind the qualitative approach. The research question was: what is the ongoing lived experience of dry mouth for SS patients, and how does each individual cope with dry mouth amid a spectrum of symptoms (with consideration to individual and environmental characteristics)?

Participants were recruited because of their knowledge (to answer the research question), and the data collected from diaries, interviews, and SXI scores were used to understand the lived experience of dry mouth in these SS patients. The collected data were analysed using thematic content analysis, an approach that has been well-established in psychology research (Braun and Clarke, 2006).

The SXI scores provided a description of the severity of the dry mouth experienced by the research participants. Compared to a range of estimates from samples of older

people, the current research participants had more than twice their mean score. Such a score could provide HCPs with insight into the severity of their patients' symptoms.

An original model was drawn from the empirical findings to depict 'the journey to diagnosis'. The model was based on the participants' interpretations of a spectrum of initial symptoms (in different combinations), that depended on their: (1) life stage; (2) disease experience; and (3) functional status affected. These led them to seek medical help, making the symptom interpretation open to HCPs. This interpretation in turn, depended on the HCPs' awareness of, and acknowledgement of, the SS symptoms. The various routes to diagnosis had an effect on QoL, and was reflected in the range of time taken (1 to 28 years). The findings emphasised the importance of GPs' awareness of SS symptoms, because they were often the first port of call for these participants.

The theme 'Interactions with HCPs' explored the bi-directional link between the two levels 'characteristics of the individual' and 'characteristics of the environment' in the revised Wilson and Cleary model (2005). The qualitative approach revealed aspects of these interactions that were not represented by the model. The current research participants tended towards either active or passive patient roles, and, together with their personalities, lifestyles, preferences, and perceptions, these had an influence on their interactions with HCPs. The pertinent aspects of these interactions were described in the context of: (1) referral to specialists; (2) prescription medications; (3) patient education; (4) HCP accessibility; (5) financial aid; and (6) empathy. The perspectives of the current research participants provided HCPs with insight into the impact of their behavior as part of the environmental support for patients and hence their QoL.

The theme 'Disease impact spectrum' described how the participants perceived their dry mouth and SS experience as a 'hidden disability' that impaired their QoL. Their dry mouth was dynamic and relative to the other SS manifestations. A 'revised revised' Wilson and Cleary model was a established from the empirical findings to reflect the participants' perspectives on what was significant in their perception of disease impact. There was a strong sense of how the characteristics of the individual

and the environment influenced their interpretation of how dry mouth and SS affected their QoL.

An original model of the positive SS (and dry mouth) coping process was established from the empirical findings. The coping process for the participants was dynamic and personalised, and had emphasis on psychosocial and cognitive aspects. In view of the lack of effective treatment options, such insight into the participants' coping processes can improve open dialogue and partnership to enable patient-HCP collaboration in managing the disease.

The discussion considered the strengths and limitations of the current research. The study was original, trustworthy, and transferable. It had made significant contributions to the general body of knowledge in improving the understanding of SS patients' dry mouth experience, and therefore providing practical clinical applications of this insight. The theoretical contribution of the current study had examined two very important domains of HRQoL/OHRQoL: the role of characteristics of the individual and the environment in relation to the SS patients' lived experience of dry mouth. This is a pioneering study that has adopted a qualitative approach to the revised Wilson and Cleary model by Ferrans et al (2005), and has provided a reconceptualisation of patients' perspectives of the model. The original models drawn from the findings in relation to the themes 'the journey to diagnosis' and 'the positive SS (and dry mouth) coping process' require further exploration. Future research was recommended to advance the patient, clinical and theoretical knowledge of HRQoL and OHRQoL in SS sufferers.

In conclusion, dry mouth is not a trivial symptom for these participants. In order to truly understand the dry mouth experience for SS sufferers, HCPs must examine them in the context of their individual characteristics (such as personality, lifestyle, preferences, and perceptions) and the environment in which he/she lives (and which HCPs are a part).

Appendices

Appendix 1

Shortened Xerostomia Inventory

The following statements refer to your experiences of mouth dryness during the last 4 weeks. For each statement, please circle the response which applies to you.

My mouth feels dry	NEVER	HARDLY EVER	OCCASION-ALLY	FREQUENTLY	ALWAYS
I have difficulty in eating dry foods	NEVER	HARDLY EVER	OCCASION-ALLY	FREQUENTLY	ALWAYS
My mouth feels dry when eating a meal	NEVER	HARDLY EVER	OCCASION-ALLY	FREQUENTLY	ALWAYS
I have difficulties swallowing certain foods	NEVER	HARDLY EVER	OCCASION-ALLY	FREQUENTLY	ALWAYS
My lips feel dry	NEVER	HARDLY EVER	OCCASION-ALLY	FREQUENTLY	ALWAYS

Global question:
How often does your mouth feel dry? (circle the appropriate response)

NEVER OCCASIONALLY FREQUENTLY ALWAYS

Appendix 2.

Revised international classification criteria for Sjögren's syndrome

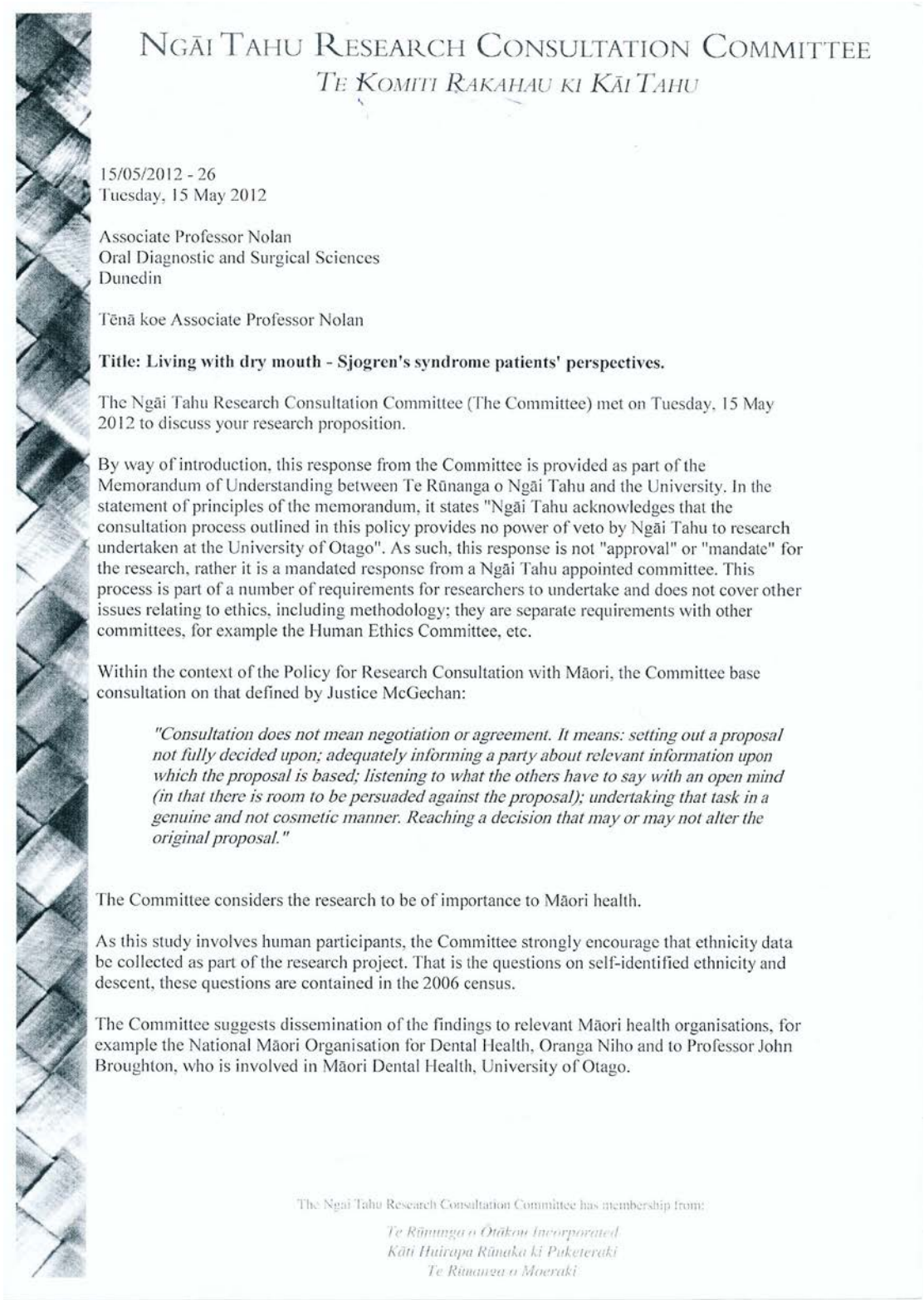
I. Ocular symptoms: a positive response to at least one of the following questions:
1. Have you had daily, persistent, troublesome dry eyes for more than 3 months?
2. Do you have a recurrent sensation of sand or gravel in the eyes?
3. Do you use tear substitutes more than 3 times a day?
II. Oral symptoms: a positive response to at least one of the following questions:
1. Have you had a daily feeling of dry mouth for more than 3 months?
2. Have you had recurrently or persistently swollen salivary glands as an adult?
3. Do you frequently drink liquids to aid in swallowing dry food?
III. Ocular signs—that is, objective evidence of ocular involvement defined as a positive result for at least one of the following two tests:
1. Schirmer's I test, performed without anaesthesia (≤5 mm in 5 minutes)
2. Rose bengal score or other ocular dye score (≥4 according to van Bijsterveld's scoring system)
IV. Histopathology: In minor salivary glands (obtained through normal-appearing mucosa) focal lymphocytic sialoadenitis, evaluated by an expert histopathologist, with a focus score ≥1, defined as a number of lymphocytic foci (which are adjacent to normal-appearing mucous acini and contain more than 50 lymphocytes) per 4 mm^2 of glandular tissue
V. Salivary gland involvement: objective evidence of salivary gland involvement defined by a positive result for at least one of the following diagnostic tests:
1. Unstimulated whole salivary flow (≤1.5 ml in 15 minutes)
2. Parotid sialography showing the presence of diffuse sialectasias (punctate, cavitary or destructive pattern), without evidence of obstruction in the major ducts
3. Salivary scintigraphy showing delayed uptake, reduced concentration and/or delayed excretion of tracer
VI. Autoantibodies: presence in the serum of the following autoantibodies:
1. Antibodies to Ro(SSA) or La(SSB) antigens, or both

Revised rules for classification

For primary SS
In patients without any potentially associated disease, primary SS may be defined as follows:
a. The presence of any 4 of the 6 items is indicative of primary SS, as long as either item IV (Histopathology) or VI (Serology) is positive
b. The presence of any 3 of the 4 objective criteria items (that is, items III, IV, V, VI)
c. The classification tree procedure represents a valid alternative method for classification, although it should be more properly used in clinical-epidemiological survey
For secondary SS
In patients with a potentially associated disease (for instance, another well defined connective tissue disease), the presence of item I or item II plus any 2 from among items III, IV, and V may be considered as indicative of secondary SS
Exclusion criteria:
Past head and neck radiation treatment
Hepatitis C infection
Acquired immunodeficiency disease (AIDS)
Pre-existing lymphoma
Sarcoidosis
Graft versus host disease
Use of anticholinergic drugs (since a time shorter than 4-fold the half life of the drug)

Appendix 3.

Maori consultation



NGĀI TAHU RESEARCH CONSULTATION COMMITTEE
TE KOMITI RAKAHAU KI KĀI TAHU

15/05/2012 - 26
Tuesday, 15 May 2012

Associate Professor Nolan
Oral Diagnostic and Surgical Sciences
Dunedin

Tēnā koe Associate Professor Nolan

Title: Living with dry mouth - Sjogren's syndrome patients' perspectives.

The Ngāi Tahu Research Consultation Committee (The Committee) met on Tuesday, 15 May 2012 to discuss your research proposition.

By way of introduction, this response from the Committee is provided as part of the Memorandum of Understanding between Te Rūnanga o Ngāi Tahu and the University. In the statement of principles of the memorandum, it states "Ngāi Tahu acknowledges that the consultation process outlined in this policy provides no power of veto by Ngāi Tahu to research undertaken at the University of Otago". As such, this response is not "approval" or "mandate" for the research, rather it is a mandated response from a Ngāi Tahu appointed committee. This process is part of a number of requirements for researchers to undertake and does not cover other issues relating to ethics, including methodology; they are separate requirements with other committees, for example the Human Ethics Committee, etc.

Within the context of the Policy for Research Consultation with Māori, the Committee base consultation on that defined by Justice McGechan:

> *"Consultation does not mean negotiation or agreement. It means: setting out a proposal not fully decided upon; adequately informing a party about relevant information upon which the proposal is based; listening to what the others have to say with an open mind (in that there is room to be persuaded against the proposal); undertaking that task in a genuine and not cosmetic manner. Reaching a decision that may or may not alter the original proposal."*

The Committee considers the research to be of importance to Māori health.

As this study involves human participants, the Committee strongly encourage that ethnicity data be collected as part of the research project. That is the questions on self-identified ethnicity and descent, these questions are contained in the 2006 census.

The Committee suggests dissemination of the findings to relevant Māori health organisations, for example the National Māori Organisation for Dental Health, Oranga Niho and to Professor John Broughton, who is involved in Māori Dental Health, University of Otago.

The Ngāi Tahu Research Consultation Committee has membership from:

Te Rūnanga o Ōtākou Incorporated
Kāti Huirapa Rūnaka ki Puketeraki
Te Rūnanga o Moeraki

NGĀI TAHU RESEARCH CONSULTATION COMMITTEE
TE KOMITI RAKAHAU KI KĀI TAHU

We wish you every success in your research and the Committee also requests a copy of the research findings.

This letter of suggestion, recommendation and advice is current for an 18 month period from Tuesday, 15 May 2012 to 15 November 2013.

The recommendations and suggestions above are provided on your proposal submitted through the consultation website process. These recommendations and suggestions do not necessarily relate to ethical issues with the research, including methodology. Other committees may also provide feedback in these areas.

Nāhaku noa, nā

pp NTRCC

Mark Brunton
Kaitakawaenga Rangahau Māori
Facilitator Research Māori
Research Division
Te Whare Wānanga o Otāgo
Ph: +64 3 479 8738
email: mark.brunton@otago.ac.nz
Web: www.otago.ac.nz

The Ngai Tahu Research Consultation Committee has membership from:

Te Rūnunga o Ōtākou Incorporated
Kāti Huirapa Rūnaka ki Puketeraki
Te Rūnanga o Moeraki

Appendix 4.

Ethics approval

22 May 2012

12/129

Dear Assoc. Prof. Nolan, I am writing to let
Ethics Committee considered your

proposal entitled "Living **with dry mouth - Sjögren's syndrome patients' perspectives".**

As a result of that consideration, the current status of your proposal is:- **Approved**

For your future reference, the Ethics Committee's reference code for this project is:- **12/129.**

The comments and views expressed by the Ethics Committee concerning your proposal are as follows:-

While approving the application, the Committee would be grateful if you would respond to the following:

Please provide the Committee with a list of references you have cited in your application.

Approval is for up to three years from the date of this letter. If this project has not been completed within three years from the date of this letter, re-approval must be requested. If the nature, consent, location, procedures or personnel of your approved application change, please advise me in writing.

Yours sincerely,

Mr Gary Witte

Manager, Academic Committees

Tel: 479 8256 Email: gary.witte@otago.ac.nz

c.c. Department of Oral Diagnostic and Surgical Sciences

Appendix 5.

Ethical approval amendment letter 2012

4 December 2012

9 8256
Email: gary.witte@otago.ac.nz

c.c. Professor R M Love Head Department of Oral Diagnostic and Surgical Sciences

Appendix 6.

Ethical approval amendment letter 2013

4 April 2013

writing. I hope all goes well for you with your upcoming research.

Appendix 7.

NZDAMOH grant approval

The total amount required for this study is proposed to be $6,256, and the full amount has been awared by the NZDAMOH oral health research fund.

NZDA House
Building 1, 195 Main Highway
Ellerslie, Auckland 1051
PO Box 28084, Remuera 1541
Ph +64 9 579 8001
Fx +64 9 580 0010
Email: research@nzda.org.nz

NEW ZEALAND
DENTAL ASSOCIATION

ADVICE OF RESEARCH FUNDING GRANT APPLICATION AS ASSESSED BY THE PANEL OF THE MINISTRY OF HEALTH ORAL HEALTH RESEARCH FUND WEDNESDAY 18 July 2012

Date of Advice	13 August 2012
Name of Applicant/s	**Ngo J**, Nolan, Ferguson S, Thomson M
Title of Research	Living with dry mouth – Sjogren's syndrome patients' perspectives.
Amount Awarded	$6,256
Condition/s of Award	Funding of this project is subject to ethics approval being obtained and is made on the condition that the Assessment Panel receives a satisfactory Progress Report by 01 June 2013 and annually by 1 June each year beyond 2013. A Final Report and copy of any publications / reports are required at the completion of the study. Note: The format and content requirements of Progress and Final Reports are currently being reviewed. You will be advised of the revised requirements in due course.
General Comments The Panel congratulates the recipients on the award of this grant and looks forward to a progress report by 01 June 2013.	
Signed: Malcolm McMillan	*Malcolm D. McMillan* (Chair, Ministry of Health Oral Health Research Fund Panel)

YOU ARE REQUIRED TO SUBMIT A PROGRESS REPORT ON YOUR RESEARCH BY 1 JUNE 2013
(Please email your report to – research@nzda.org.nz)

The Principal Researcher should sign, date and return a **COPY of** this advice notice (in the panel below) to acknowledge the conditions and enable receipt of the Award. If the Principal Researcher is a post-graduate student then the student's supervisor should sign and return this form. Thank you.

Name: _____ **Signed:** _____ **Date:** ___/___/2012
*Principal Researcher **OR** Student Supervisor*

Page 1 of 1

Appendix 8.

Information sheet

12/129
INFORMATION SHEET FOR PARTICIPANTS

Thank you for showing an interest in this project. Please read this information sheet carefully before deciding whether or not to participate. If you decide to participate we thank you. If you decide not to take part there will be no disadvantage to you and we thank you for considering our request.

What is the Aim of the Project?

The aim of this qualitative research project is to explore how Sjögren's Syndrome(SS) patients live with dry mouth. Diary and interview research methods will be used to understand the daily experiences of participants coping with dry mouth. The results are likely to provide a better understanding of the impact of dry mouth on SS patients' lives. This project is being undertaken as part of the requirements for the Doctorate of Clinical Dentistry Special Needs Dentistry.

What Type of Participants are being sought?

We would like people with Sjögren's Syndrome who suffer from dry mouth to share
their experiences in their diaries, and discussions.

What will Participants be Asked to Do?

Should you agree to take part in this project, you will be asked to maintain a month-long diary entry about how dry mouth affects your daily life. This dairy can be organised in any style, including writing or scrapbook form. The amount of time dedicated to diary entry is entirely up to your own discretion. Some of the points which you may wish to include in the diary entries are sleep pattern, social meals, relationship with others, occupation, dental problems, positive and negative emotions

about dry mouth. During this month, the interviewer will contact you weekly by email/phone to discuss the progress of the diary entries, and answer any queries you may have. Later, you will be asked to participate in a face-to-face/skype interview which will last about 1 hour. The main themes from your diary entries will be discussed further during the interview. All these will be carried out in the dental school meeting room. Should an alternative be required, discussions can be made. Please be aware that you may decide not to take part in the project without any disadvantage to yourself of any kind.

What Data or Information will be Collected and What Use will be Made of it?
Data will be collected as mentioned above. The interviews will be audio-taped and transcribed. The diaries and the interviews will also be analysed for emerging themes. The analysis will produce insights on your experiences coping with dry mouth. For the final thesis report and any scientific journal publication, every attempt will be made to preserve your anonymity. The data will be accessible by researchers, supervisors, transcribers and examiners of the thesis. The data collected will be securely stored in such a way that only those mentioned below will be able to gain access to it. Data obtained as a result of the research will be retained for **at least 5 years** in secure storage. Any personal information held on the participants [*such as contact details, audio tapes, after they have been transcribed etc*,] may be destroyed at the completion of the research even though the data derived from the research will, in most cases, be kept for much longer or possibly indefinitely. The results of the project may be published and will be available in the University of Otago Library (Dunedin, New Zealand) but every attempt will be made to preserve your anonymity. During the follow up interview post-diary, there will be an opportunity for us to discuss whether the themes drawn from the provided data are interpreted as the original meaning as you have expressed. Any clarification or amendments can be made during the interview. This project involves an open-questioning technique. The general line of questioning includes how dry mouth may affect your daily life in aspects including social meals, relationships with others, sleep pattern, occupation, emotions, and dental problems stress. The precise nature of the questions which will be asked have not been determined in advance, but will depend on the way in which the interview develops. Consequently, although the University of Otago Human Ethics Committee is aware of the general areas to be explored in the interview, the

Committee has not been able to review the precise questions to be used. In the event that the line of questioning does develop in such a way that you feel hesitant or uncomfortable you are reminded of your right to decline to answer any particular question(s) and also that you may withdraw from the project at any stage without any disadvantage to yourself of any kind.

Can Participants Change their Mind and Withdraw from the Project?

You may withdraw from participation in the project at any time and without any disadvantage to yourself of any kind.

What if Participants have any Questions?

If you have any questions about our project, either now or in the future, please feel free to contact either:

Joanna Ngo and/or Anita Nolan

Department of Oral Diagnostic and Surgical Sciences, School of Dentistry, University of Otago

University Telephone Number: (470) 3555

University Telephone Number: (470) 3556

Email Address: ssresearch2012@yahoo.co.nz
Email Address: anita.nolan@otago.ac.nz

This study has been approved by the University of Otago Human Ethics Committee. If you have any concerns about the ethical conduct of the research you may contact the Committee through the Human Ethics Committee Administrator (ph 03 479 8256). Any issues you raise will be treated in confidence and investigated and you will be informed of the outcome.

Appendix 9.

Participants consent form

12/129

Living with dry mouth - Sjögren's syndrome patients' perspectives

CONSENT FORM FOR PARTICIPANTS

I have read the Information Sheet concerning this project and understand what it is about. All my questions have been answered to my satisfaction. I understand that I am free to request further information at any stage.
I know that:

1. My participation in the project is entirely voluntary;

2. I am free to withdraw from the project at any time without any disadvantage;

3. Personal identifying information such as audio-tapes and diary entries will be destroyed at the conclusion of the project but any raw data on which the results of the project depend will be retained in secure storage for at least five years;

4. This project involves an open-questioning technique. The general line of questioning includes social meals, relationships with others, sleep pattern, occupation, emotions, and dental problems stress. The precise nature of the questions which will be asked have not been determined in advance, but will depend on the way in which the interview develops and that in the event that the line of questioning develops in such a way that I feel hesitant or uncomfortable I may decline to answer any particular question(s) and/or may withdraw from the project without any disadvantage of any kind.

5. The results of the project may be published and will be available in the University of Otago Library (Dunedin, New Zealand) but every attempt will be made to preserve my anonymity should I choose to remain anonymous.

6. There will be a reimbursement for participation with a $50 new world voucher.

I agree to take part in this project.

....

(Signature of participant) (Date)

This study has been approved by the University of Otago Human Ethics Committee. If you have any concerns about the ethical conduct of the research you may contact the Committee through the Human Ethics Committee Administrator (ph 03 479 8256). Any issues you raise will be treated in confidence and investigated and you will be informed of the outcome.

Appendix 10.

Guidelines for diary entries

Guidelines for diary entry about the impact of Sjögren's Syndrome (SS) on your daily life.
The diary may be inclusive of, but not exclusive to the following points

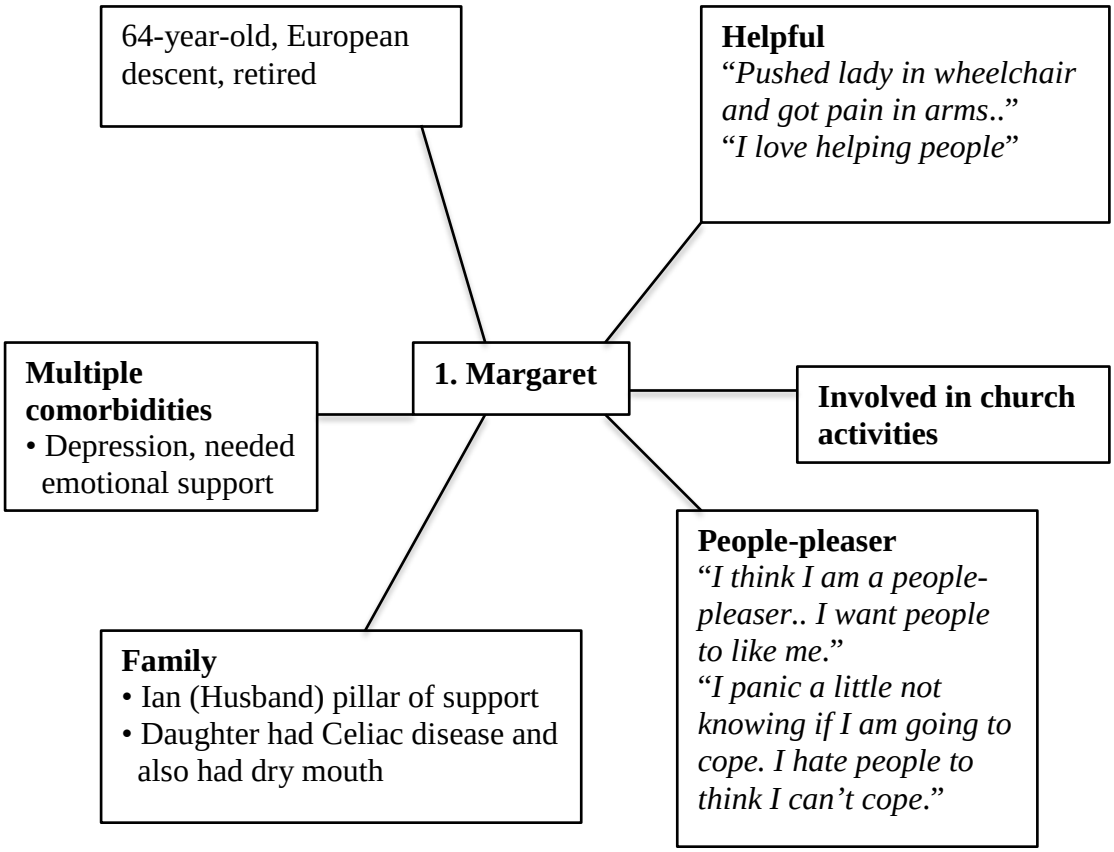
- Personal experience leading to diagnosis of SS (beginning of diary)
- Interaction with healthcare professionals
- Family responses
- Social interactions
- Daily routines
- Positive and negative impact on emotions, psychology, and body function
- Coping mechanisms

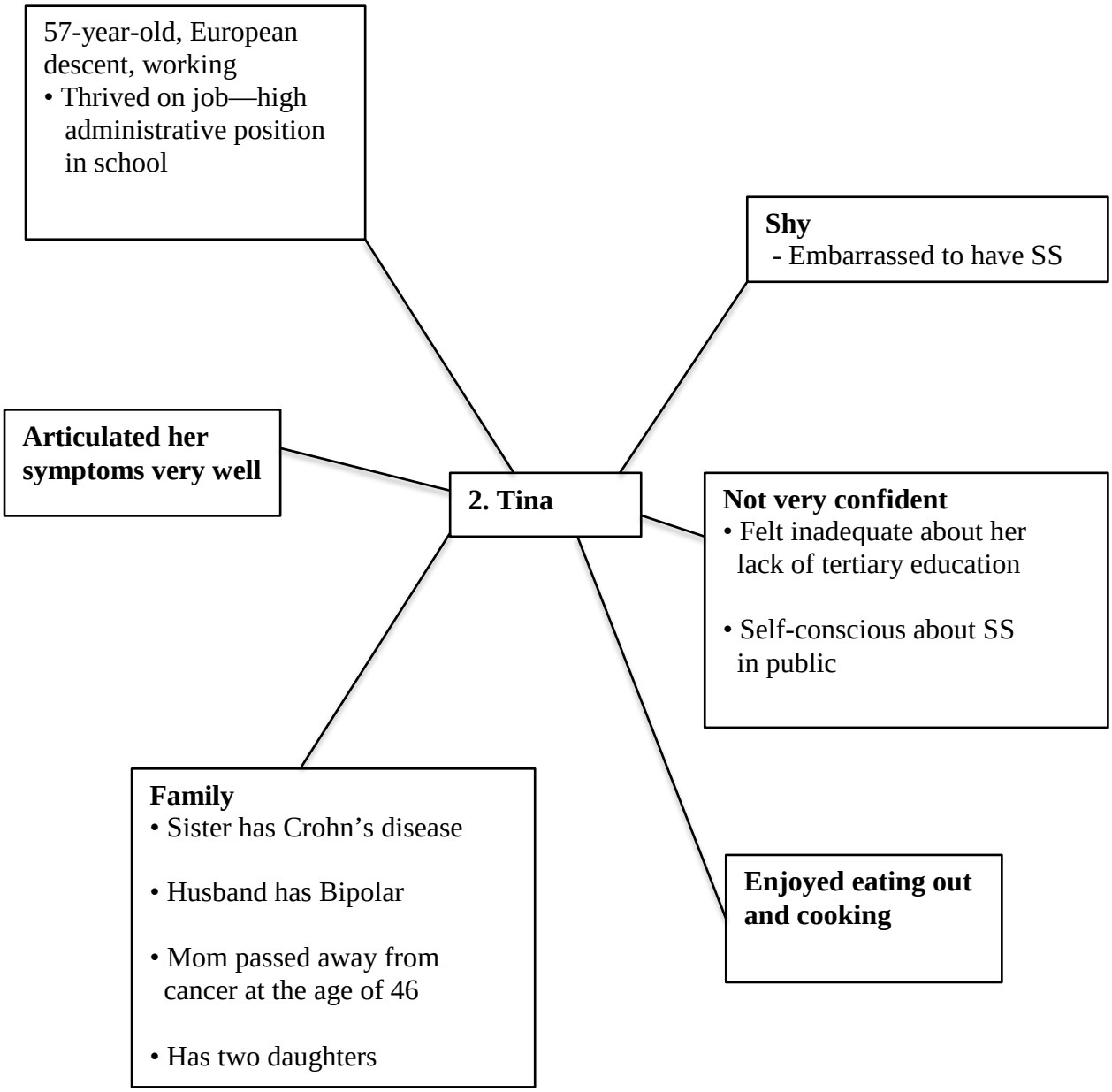
The aim of the diary entries is for clinicians to gain insight into the daily lives of patients with SS. An entry each day would be great, multiple entries regarding significant events would be wonderful, however, please do not feel obliged to write for the sake of it. The absence of entries on certain days is acceptable, both positive and negative entries are encouraged.

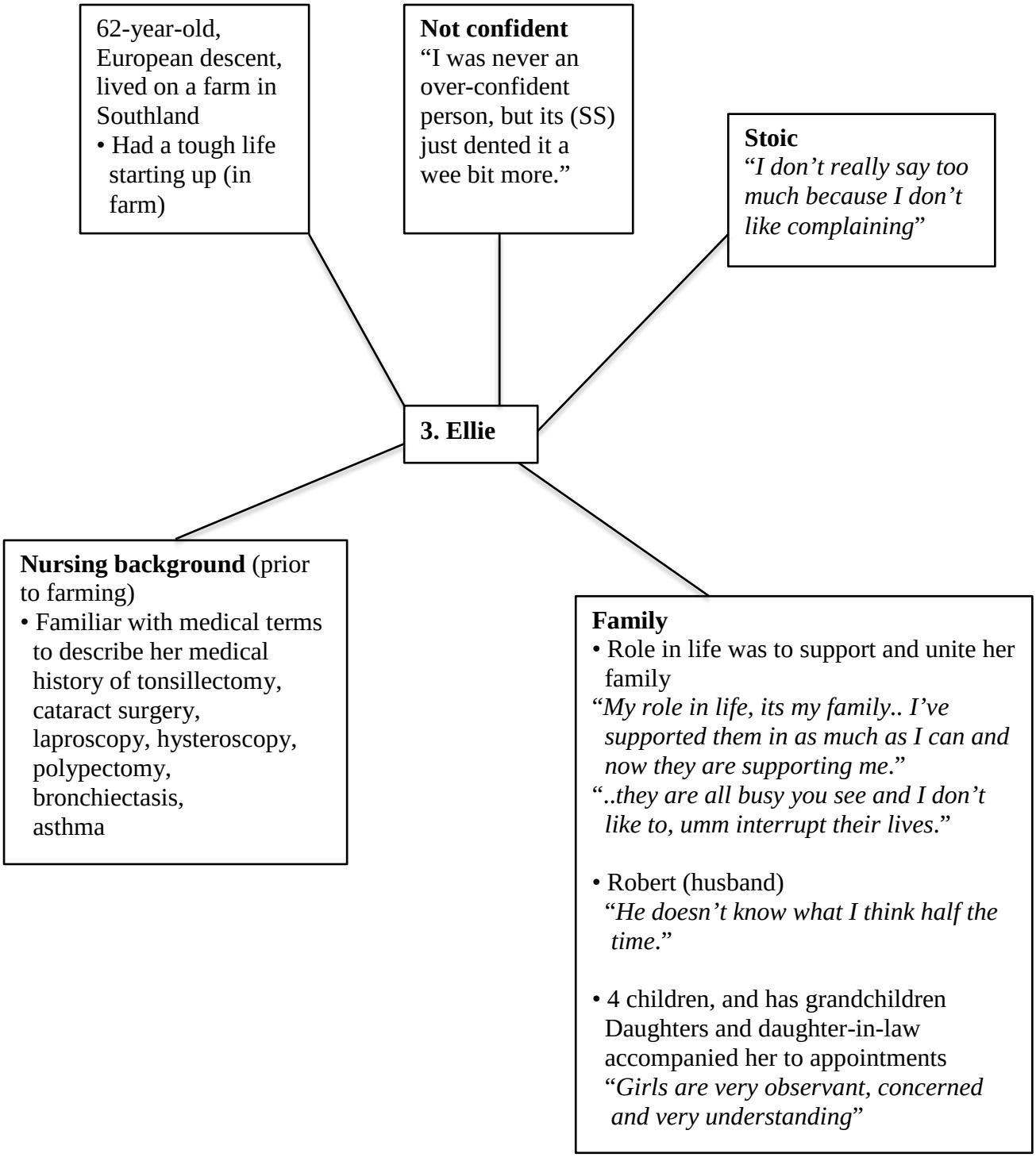
Entries may be made via email or hard copy. Thank you.
ssresearch2012@yahoo.co.nz

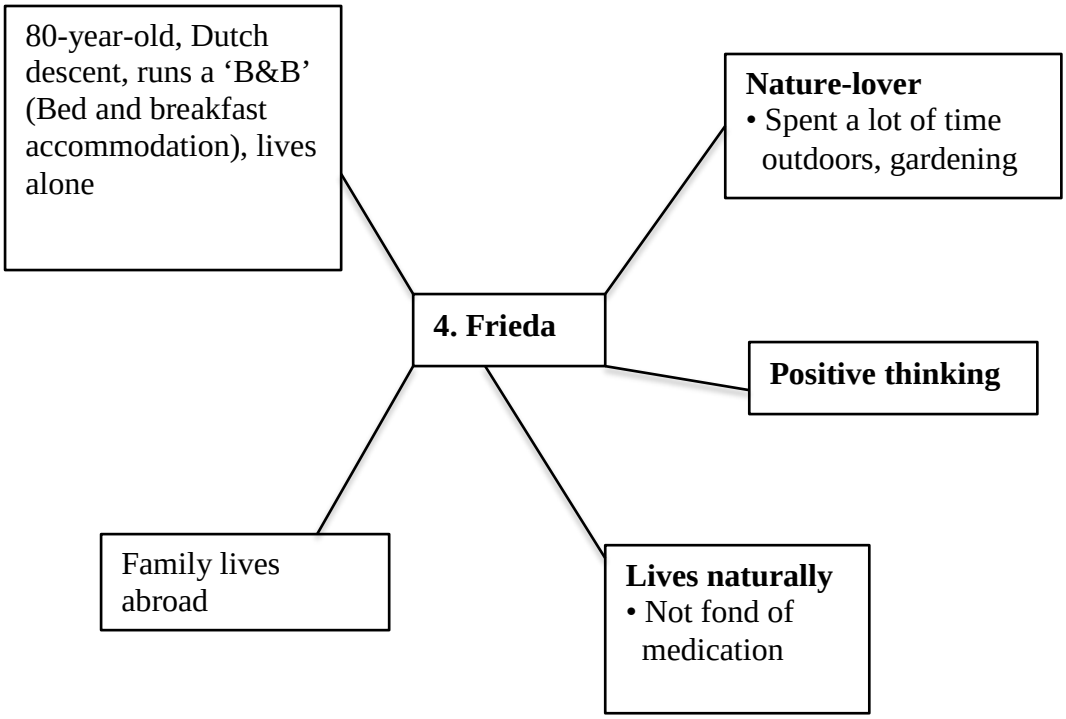
Appendix 11

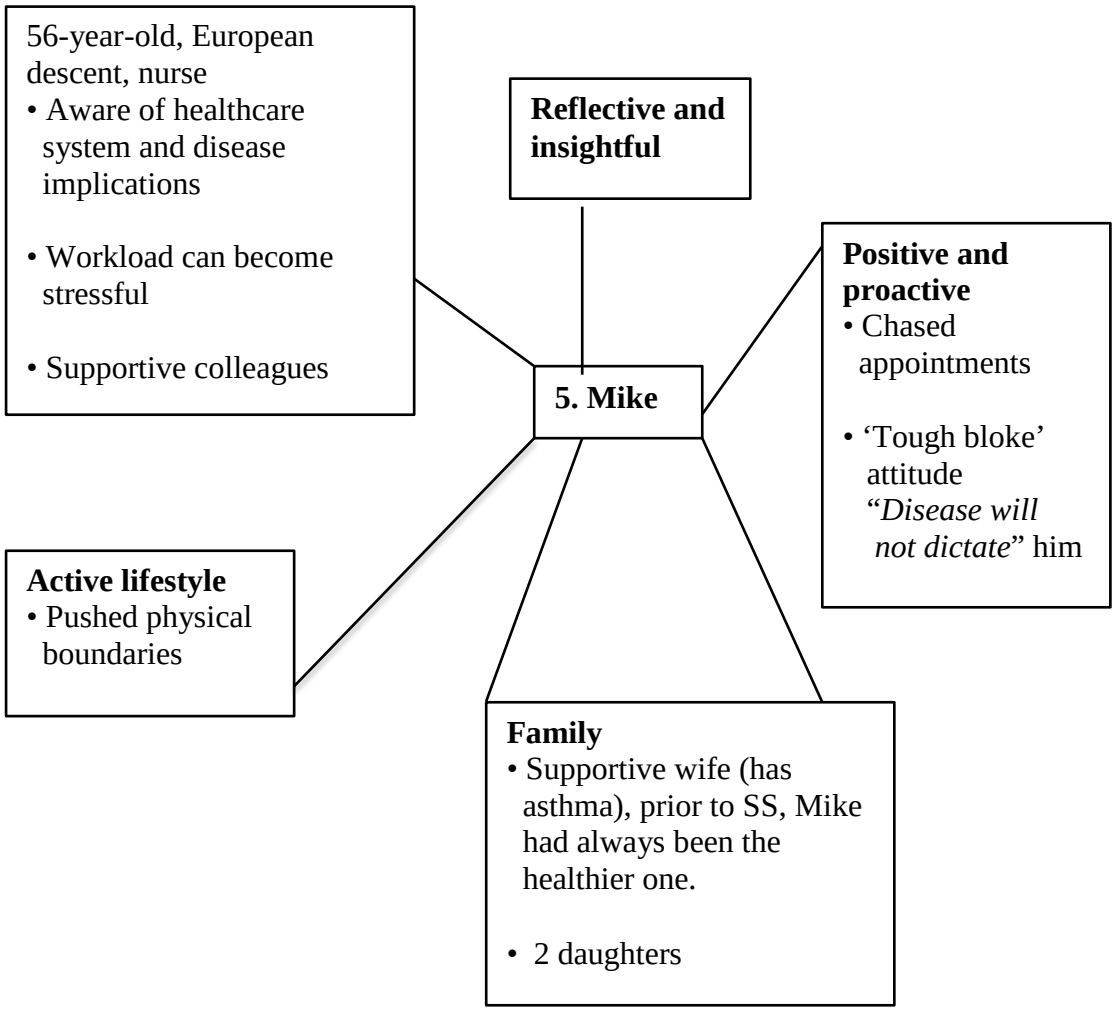
Summary diagrams of each participant's individual and environmental characteristics

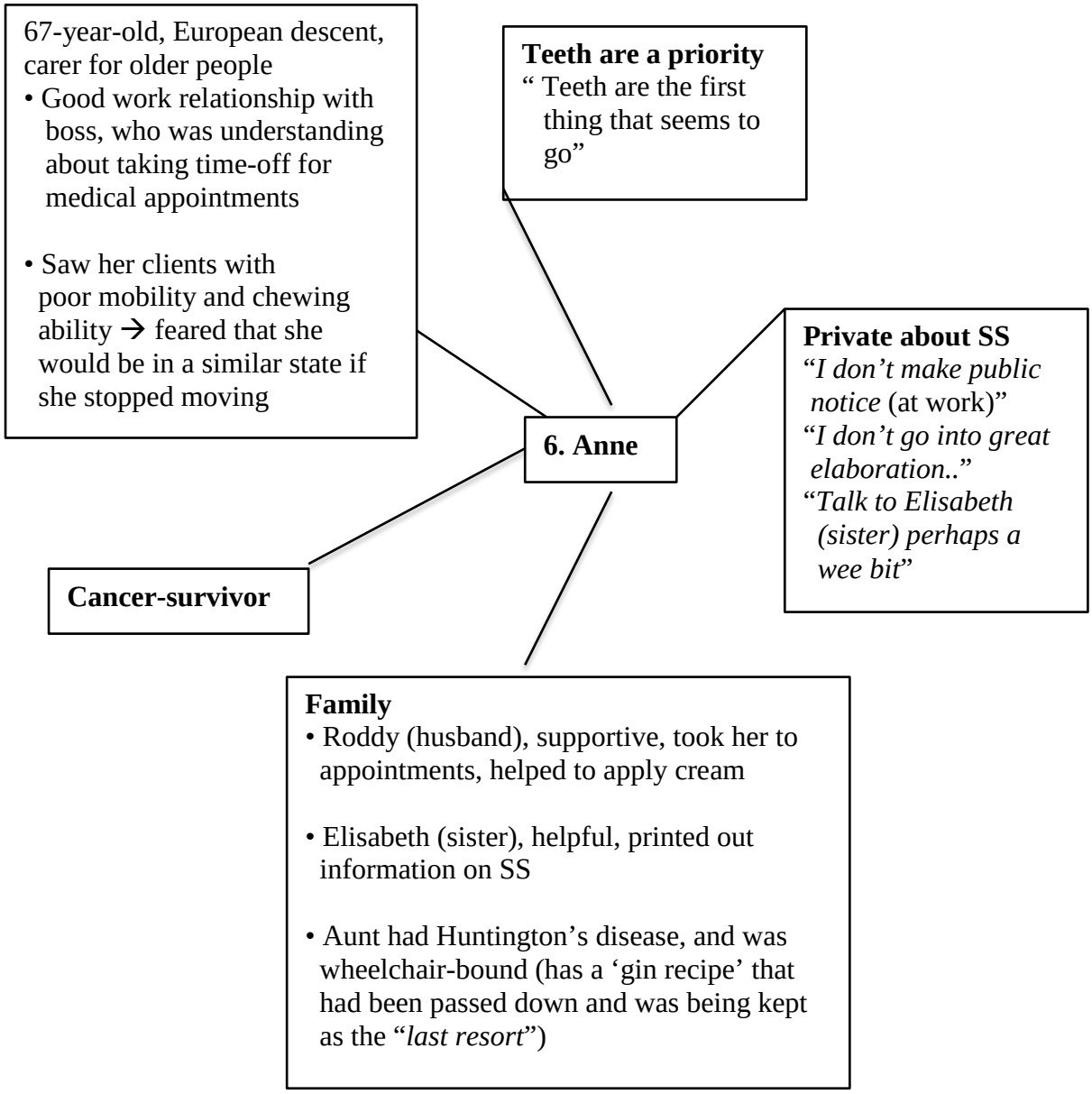


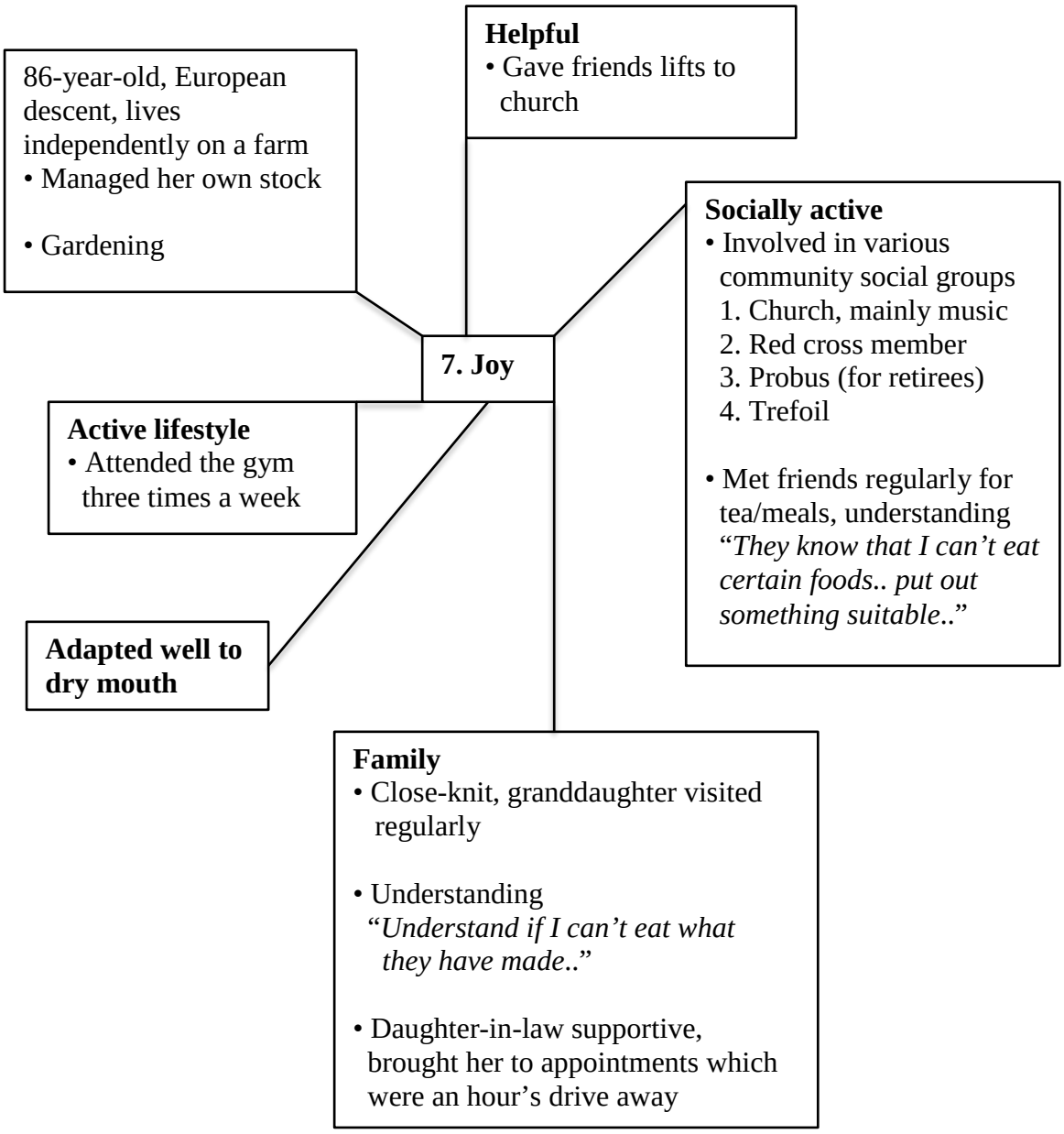

57-year-old, European descent, working
• Thrived on job—high administrative position in school
Shy
- Embarrassed to have SS
Articulated her symptoms very well
2. Tina
Not very confident
• Felt inadequate about her lack of tertiary education
• Self-conscious about SS in public
Family
• Sister has Crohn's disease
• Husband has Bipolar
• Mom passed away from cancer at the age of 46
• Has two daughters
Enjoyed eating out and cooking

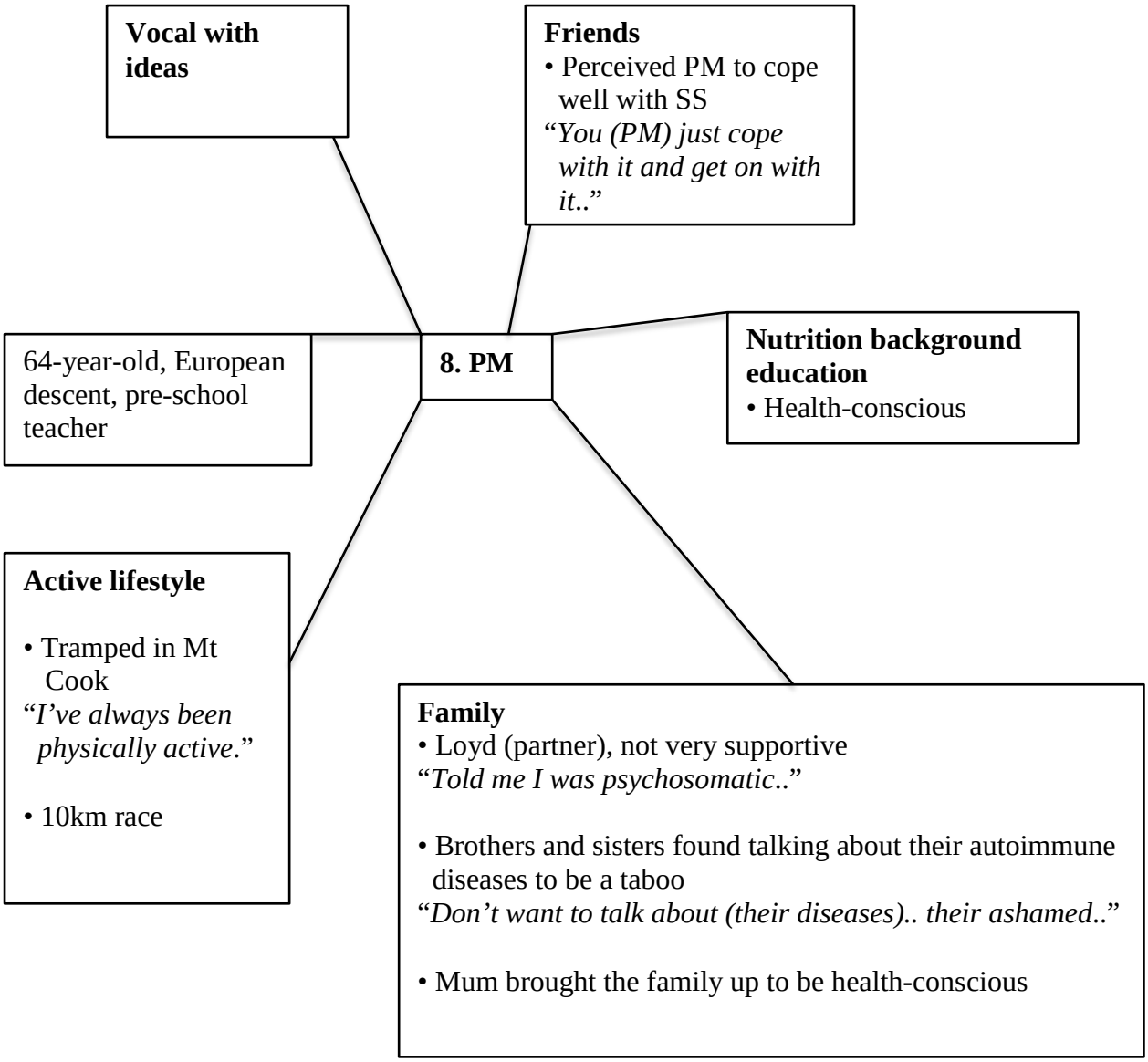

62-year-old, European descent, lived on a farm in Southland
• Had a tough life starting up (in farm)
Not confident
"I was never an over-confident person, but its (SS) just dented it a wee bit more."
Stoic
"I don't really say too much because I don't like complaining"
3. Ellie
Nursing background (prior to farming)
• Familiar with medical terms to describe her medical history of tonsillectomy, cataract surgery, laproscopy, hysteroscopy, polypectomy, bronchiectasis, asthma
Family
• Role in life was to support and unite her family
"My role in life, its my family.. I've supported them in as much as I can and now they are supporting me."
"..they are all busy you see and I don't like to, umm interrupt their lives."
• Robert (husband)
"He doesn't know what I think half the time."
• 4 children, and has grandchildren
Daughters and daughter-in-law accompanied her to appointments
"Girls are very observant, concerned and very understanding"

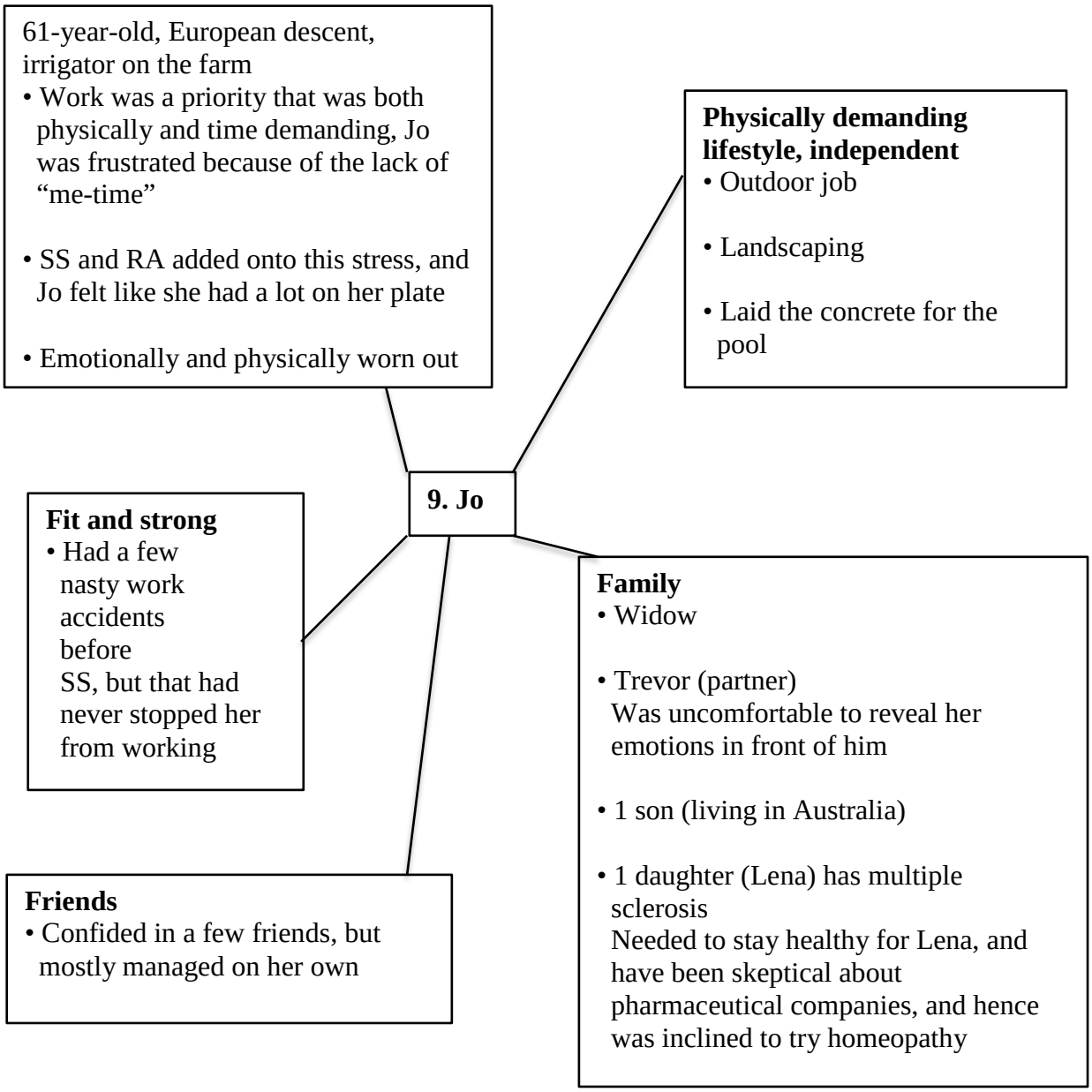

80-year-old, Dutch descent, runs a ‘B&B’ (Bed and breakfast accommodation), lives alone
Nature-lover
• Spent a lot of time outdoors, gardening
4. Frieda
Positive thinking
Family lives abroad
Lives naturally
• Not fond of medication

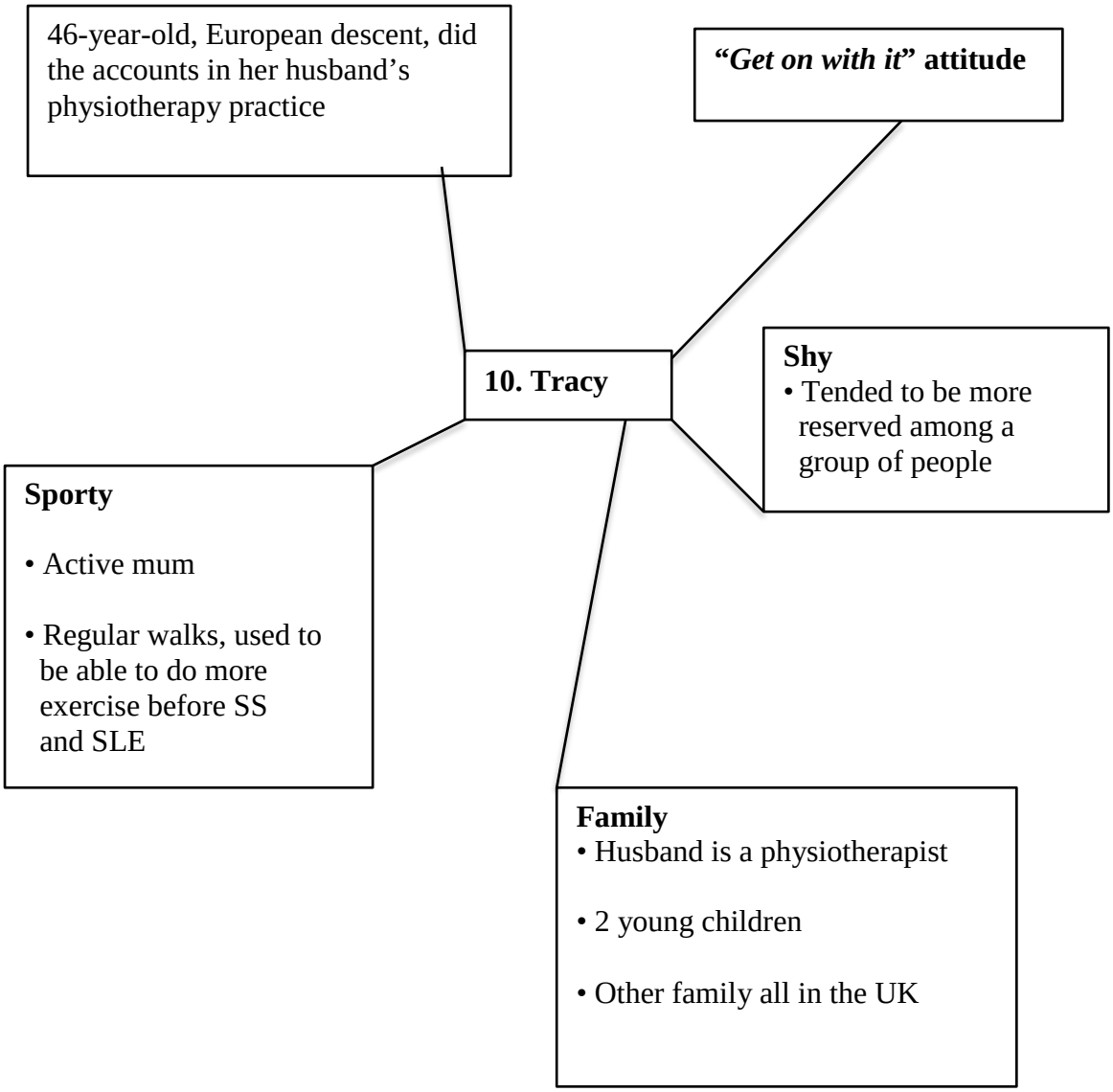

56-year-old, European descent, nurse
• Aware of healthcare system and disease implications
• Workload can become stressful
• Supportive colleagues
Reflective and insightful
Positive and proactive
• Chased appointments
• 'Tough bloke' attitude
"Disease will not dictate" him
5. Mike
Active lifestyle
• Pushed physical boundaries
Family
• Supportive wife (has asthma), prior to SS, Mike had always been the healthier one.
• 2 daughters


67-year-old, European descent, carer for older people
• Good work relationship with boss, who was understanding about taking time-off for medical appointments
• Saw her clients with poor mobility and chewing ability → feared that she would be in a similar state if she stopped moving
Teeth are a priority
" Teeth are the first thing that seems to go"
Private about SS
"I don't make public notice (at work)"
"I don't go into great elaboration.."
"Talk to Elisabeth (sister) perhaps a wee bit"
6. Anne
Cancer-survivor
Family
• Roddy (husband), supportive, took her to appointments, helped to apply cream
• Elisabeth (sister), helpful, printed out information on SS
• Aunt had Huntington's disease, and was wheelchair-bound (has a 'gin recipe' that had been passed down and was being kept as the "last resort")


86-year-old, European descent, lives independently on a farm
• Managed her own stock
• Gardening
Helpful
• Gave friends lifts to church
Socially active
• Involved in various community social groups
1. Church, mainly music
2. Red cross member
3. Probus (for retirees)
4. Trefoil
• Met friends regularly for tea/meals, understanding "They know that I can't eat certain foods.. put out something suitable.."
7. Joy
Active lifestyle
• Attended the gym three times a week
Adapted well to dry mouth
Family
• Close-knit, granddaughter visited regularly
• Understanding "Understand if I can't eat what they have made.."
• Daughter-in-law supportive, brought her to appointments which were an hour's drive away


Vocal with ideas
Friends
• Perceived PM to cope well with SS
"You (PM) just cope with it and get on with it.."
64-year-old, European descent, pre-school teacher
8. PM
Nutrition background education
• Health-conscious
Active lifestyle
• Tramped in Mt Cook
"I've always been physically active."
• 10km race
Family
• Loyd (partner), not very supportive
"Told me I was psychosomatic.."
• Brothers and sisters found talking about their autoimmune diseases to be a taboo
"Don't want to talk about (their diseases).. their ashamed.."
• Mum brought the family up to be health-conscious


61-year-old, European descent, irrigator on the farm
• Work was a priority that was both physically and time demanding, Jo was frustrated because of the lack of "me-time"
• SS and RA added onto this stress, and Jo felt like she had a lot on her plate
• Emotionally and physically worn out
Physically demanding lifestyle, independent
• Outdoor job
• Landscaping
• Laid the concrete for the pool
9. Jo
Fit and strong
• Had a few nasty work accidents before SS, but that had never stopped her from working
Family
• Widow
• Trevor (partner)
Was uncomfortable to reveal her emotions in front of him
• 1 son (living in Australia)
• 1 daughter (Lena) has multiple sclerosis
Needed to stay healthy for Lena, and have been skeptical about pharmaceutical companies, and hence was inclined to try homeopathy
Friends
• Confided in a few friends, but mostly managed on her own


46-year-old, European descent, did the accounts in her husband's physiotherapy practice
"Get on with it" attitude
10. Tracy
Shy
• Tended to be more reserved among a group of people
Sporty
• Active mum
• Regular walks, used to be able to do more exercise before SS and SLE
Family
• Husband is a physiotherapist
• 2 young children
• Other family all in the UK

Appendix 12

CD with verbatim transcripts

Attached to the back cover of the thesis

References

Abdelghany A, Nolan A, Freeman R (2011). Treating patients with dry mouth: general dental practitioners' knowledge, attitudes and clinical management. *British Dental Journal* 211(10)*:*E21-E21.

Adair JG (1984). The Hawthorne effect: A reconsideration of the methodological artifact. *Journal of Applied Psychology* 69(2)*:*334.

Agha-Hosseini F, Mirzaii-Dizgah I, Mirjalili N (2012). Relationship of stimulated whole saliva cortisol level with the severity of a feeling of dry mouth in menopausal women. *Gerodontology* 29(1)*:*43-47.

Ainamo J, Barmes D, Beagrie G, Cutress T, Martin J, Sardo-Infirri J (1982). Development of the World Health Organization (WHO) community periodontal index of treatment needs (CPITN). *International Dental Journal* 32(3)*:*281.

Al-Hashimi I (2001). The management of Sjögren's syndrome in dental practice. *The Journal of the American Dental Association* 132(10)*:*1409-1417.

Al-Hashimi I, Khuder S, Haghighat N, Zipp M (2001). Frequency and predictive value of the clinical manifestations in Sjögren's syndrome. *Journal of Oral Pathology and Medicine* 30(1)*:*1-6.

Al-Hashimi I (2005). Xerostomia secondary to Sjögren's syndrome in the elderly. *Drugs & Aging* 22(11)*:*887-899.

Albrecht GL, Devlieger PJ (1999). The disability paradox: high quality of life against all odds. *Social Science & Medicine* 48(8)*:*977-988.

Allen PF, Locker D (1997). Do item weights matter? An assessment using the oral health impact profile. *Community Dental Health* 14(3)*:*133-138.

Allen PF, McMillan AS, Locker D (2001). An assessment of sensitivity to change of the oral health impact profile in a clinical trial. *Community Dentistry and Oral Epidemiology* 29(3)*:*175-182.

Allen PF (2003). Assessment of oral health related quality of life. *Health and Quality of Life Outcomes* 1(1)*:*40.

Allison PJ, Locker D, Feine JS (1997). Quality of life: a dynamic construct. *Social Science & Medicine* 45(2)*:*221-230.

Anderson LA, Dedrick RF (1990). Development of the trust in physician scale: A measure to assess interpersonal trust inpatient-physician relationships. *Psychological Reports* 67(3f)*:*1091-1100.

Andersson SI, Manthorpe R, Theander E (2001). Situational patterns in coping with primary Sjo¨gren's syndrome. *Psychology, Health & Medicine* 6(1)*:*29-40.

Anttila SS, Knuuttila M, Sakki TK (1998). Depressive symptoms as an underlying factor of the sensation of dry mouth. *Psychosomatic Medicine* 60(2)*:*215-218.

Arora NK, McHorney CA (2000). Patient preferences for medical decision making: who really wants to participate? *Medical Care* 38(3)*:*335-341.

Arora NK, Finney Rutten LJ, Gustafson DH, Moser R, Hawkins RP (2007). Perceived helpfulness and impact of social support provided by family, friends, and health care providers to women newly diagnosed with breast cancer. *Psycho-Oncology* 16(5)*:*474-486.

Astin HS (1984). The meaning of work in women's lives: A sociopsychological model of career choice and work behavior. *The Counseling Psychologist*.

Atchison KA, Dolan TA (1990). Development of the geriatric oral health assessment index. *Journal of Dental Education* 54(11)*:*680-687.

Bakas T, McLennon SM, Carpenter JS, Buelow JM, Otte JL, Hanna KM *et al.* (2012). Systematic review of health-related quality of life models. *Health and Quality of Life Outcomes* 10(1)*:*134.

Baker SR, Pankhurst CL, Robinson PG (2007). Testing relationships between clinical and non-clinical variables in xerostomia: a structural equation model of oral health-related quality of life. *Quality of Life Research* 16(2)*:*297-308.

Baker SR, Pearson NK, Robinson PG (2008). Testing the applicability of a conceptual model of oral health in housebound edentulous older people. *Community Dentistry and Oral Epidemiololgy* 36(3)*:*237-248.

Baldini C, Talarico R, Tzioufas AG, Bombardieri S (2012). Classification criteria for Sjögren's syndrome: a critical review. *Journal of Autoimmunity* 39(1)*:*9-14.

Bax H, Vriesendorp T, Kallenberg C, Kalk W (2002). Fatigue and immune activity in Sjögren's syndrome. *Annals of the Rheumatic diseases* 61(3)*:*284-284.

Bayetto K, Logan RM (2010). Sjögren's syndrome: a review of aetiology, pathogenesis, diagnosis and management. *Australian Dental Journal* 55 Suppl 1(39-47.

Belenguer R, Ramos-Casals M, Brito-Zeron P, Del Pino J, Sentís J, Aguiló S *et al.* (2004). Influence of clinical and immunological parameters on the health-related quality of life of patients with primary Sjögren's syndrome. *Clinical and Experimental Rheumatology* 23(3)*:*351-356.

Berzon R, Hays R, Shumaker S (1993). International use, application and performance of health-related quality of life instruments. *Quality of Life Research* 2(6)*:*367-368.

Binard A, Devauchelle-Pensec V, Fautrel B, Jousse S, Youinou P, Saraux A (2007). Epidemiology of Sjögren's syndrome: where are we now? *Clinical and Experimental Rheumatolology* 25(1)*:*1-4.

Binard A, Le Pottier L, Devauchelle-Pensec V, Saraux A, Youinou P, Pers J-O (2009). Is the blood B-cell subset profile diagnostic for Sjögren syndrome? *Annals of the Rheumatic Diseases* 68(9)*:*1447-1452.

Bird CM (2005). How I stopped dreading and learned to love transcription. *Qualitative Inquiry* 11(2)*:*226-248.

Bodenheimer T, Lorig K, Holman H, Grumbach K (2002). Patient self-management of chronic disease in primary care. *The Journal of the American Medical Association* 288(19)*:*2469-2475.

Bøge Christensen L, Petersen PE, Thorn JJ, Schiødt M (2001). Dental caries and dental health behavior of patients with primary Sjögren syndrome. *Odontologica Scandinavica* 59(3)*:*116-120.

Bolger N, Davis A, Rafaeli E (2003). Diary methods: Capturing life as it is lived. *Annual Review of Psychology* 54(1)*:*579-616.

Bolstad AI, Haga H-J, Wassmuth R, Jonsson R (2000). Monozygotic twins with primary Sjögren's syndrome. *Journal of Rheumatology* 27(9)*:*2264.

Bots CP, Brand HS, Veerman EC, Korevaar JC, Valentijn-Benz M, Bezemer PD *et al.* (2005). Chewing gum and a saliva substitute alleviate thirst and xerostomia in patients on haemodialysis. *Nephrology Dialysis Transplantation* 20(3)*:*578-584.

Bowling A (2005). Just one question: If one question works, why ask several? *Journal of Epidemiology and Community Health* 59(5)*:*342-345.

Bowman S, Ibrahim G, Holmes G, Hamburger J, Ainsworth J (2004). Estimating the prevalence among Caucasian women of primary Sjögren's syndrome in two general practices in Birmingham, UK. *Scandinavian Journal of Rheumatology* 33(1)*:*39-43.

Braun V, Clarke V (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology* 3(2)*:*77-101.

Brazier J-, Harper R, Jones N, O'cathain A, Thomas K, Usherwood T *et al.* (1992). Validating the SF-36 health survey questionnaire: new outcome measure for primary care. *British Medical Journal* 305(6846)*:*160.

Brito-Zerón P, Sisó-Almirall A, Bové A, Kostov BA, Ramos-Casals M (2013). Primary Sjögren syndrome: an update on current pharmacotherapy options and future directions. *Expert Opinion on Pharmacotherapy* 14(3)*:*279-289.

Britten N (1995). Qualitative interviews in medical research. *British Medical Journal* 311(6999)*:*251-253.

Broom A, Willis E (2007). Competing paradigms and health research. *Researching Health: Qualitative, Quantitative and Mixed Methods:*16-31.

Brown RF, Butow PN, Henman M, Dunn SM, Boyle F, Tattersall MH (2002). Responding to the active and passive patient: flexibility is the key. *Health Expectations* 5(3)*:*236-245.

Bullock A (2010). Dental education as a social science: some implications for research. *European Journal of Dental Education* 14(2)*:*65-68.

Bylund CL, Makoul G (2002). Empathic communication and gender in the physician-patient encounter. *Patient Education and Counseling* 48(3)*:*207-216.

Cameron L, Leventhal EA, Leventhal H (1995). Seeking medical care in response to symptoms and life stress. *Psychosomatic Medicine* 57(1)*:*37-47.

Carey JW (1993). Linking qualitative and quantitative methods: Integrating cultural factors into public health. *Qualitative Health Research* 3(3)*:*298-318.

Carr A, Ng W-F, Figueiredo F, Macleod R, Greenwood M, Staines K (2012). Sjögren's syndrome–an update for dental practitioners. *British Dental Journal* 213(7)*:*353-357.

Carr AJ, Gibson B, Robinson PG (2001). Measuring quality of life: Is quality of life determined by expectations or experience? *British Medical Journal* 322(7296)*:*1240.

Carson D, Gilmore A, Perry C, Gronhaug K (2001). Qualitative marketing research: Sage.

Casebeer AL, Verhoef MJ (1997). Combining qualitative and quantitative research methods: considering the possibilities for enhancing the study of chronic diseases. *Chronic Diseases in Canada* 18(3)*:*18-13.

Chisholm D, Mason D (1968). Labial salivary gland biopsy in Sjögren's disease. *Journal of Clinical Pathology* 21(5)*:*656-660.

Chrischilles EA, Rubenstein LM, Voelker MD, Wallace RB, Rodnitzky RL (2002). Linking clinical variables to health-related quality of life in Parkinson's disease. *Parkinsonism & Related Disorders* 8(3)*:*199-209.

Christiansen CH (1999). Defining lives: Occupation as identity: An essay on competence, coherence, and the creation of meaning. *American Journal of Occupational Therapy* 53(6)*:*547-558.

Cleeland CS (2007). Symptom burden: multiple symptoms and their impact as patient-reported outcomes. *Journal of the National Cancer Institute Monographs* 2007(37)*:*16-21.

Corbett J, Pullen A, Rhodes C (2009). What a kerfuffle about a truffle: Food and the incorporation of identity. *Bits of Organization:*119-140.

Coulter A (1997). Partnerships with patients: the pros and cons of shared clinical decision-making. *Journal of Health Services Research* 2(2)*:*112-121.

D'Zurilla TJ, Nezu AM (1986). Problem-solving therapy: A social competence approach to clinical intervention: Springer Publishing Company New York.

Daniels S, Glorieux I, Minnen J, van Tienoven TP (2012). More than preparing a meal? Concerning the meanings of home cooking. *Appetite* 58(3)*:*1050-1056.

de Ridder D, Geenen R, Kuijer R, van Middendorp H (2008). Psychological adjustment to chronic disease. *The Lancet* 372(9634)*:*246-255.

Deák M, Szvetnik A, Balog A, Sohár N, Varga R, Pokorny G *et al.* (2012). Neuroimmune Interactions in Sjögren's Syndrome: Relationship of Exocrine Gland Dysfunction with Autoantibodies to Muscarinic Acetylcholine Receptor-3 and Mental Health Status Parameters. *Neuroimmunomodulation* 20(2)*:*79-86.

Delalande S, De Seze J, Fauchais A-L, Hachulla E, Stojkovic T, Ferriby D *et al.* (2004). Neurologic manifestations in primary Sjögren syndrome: a study of 82 patients. *Medicine* 83(5)*:*280-291.

Delli K, Vissink A, Spijkervet FK (2014). Salivary Gland Biopsy for Sjögren's Syndrome. *Oral and Maxillofacial Surgery Clinics of North America* 26(1)*:*23-33.

Dellve L, Samuelsson L, Tallborn A, Fasth A, Hallberg LRM (2006). Stress and well-being among parents of children with rare diseases: a prospective intervention study. *Journal of Advanced Nursing* 53(4)*:*392-402.

DiBonaventura M, Sun SX, Bolge SC, Wagner J-S, Mody R (2011). Health-related quality of life, work productivity and health care resource use associated with constipation predominant irritable bowel syndrome. *Current Medical Research & Opinion* 27(11)*:*2213-2222.

Diener E, Suh EM, Lucas RE, Smith HL (1999). Subjective well-being: Three decades of progress. *Psychological Bulletin* 125(2)*:*276.

Dodd M, Janson S, Facione N, Faucett J, Froelicher ES, Humphreys J *et al.* (2001). Advancing the science of symptom management. *Journal of Advanced Nursing* 33(5)*:*668-676.

Dolan TA, Peek CW, Stuck AE, Beck JC (1998). Three-year changes in global oral health rating by elderly dentate adults. *Community Dentistry and Oral Epidemiology* 26(1)*:*62-69.

Durham J, Steele J, Moufti MA, Wassell R, Robinson P, Exley C (2011). Temporomandibular disorder patients' journey through care. *Community Dentistry and Oral Epidemiology* 39(6)*:*532-541.

Ebert EC (2012). Gastrointestinal and hepatic manifestations of Sjögren syndrome. *Journal of Clinical Gastroenterology* 46(1)*:*25-30.

Edmunds S, Brown G (2012). Doing qualitative research in dentistry and dental education. *European Journal of Dental Education* 16(2)*:*110-117.

Elliott H (1997). The use of diaries in sociological research on health experience. *Sociological Research Online* 2(2).

Eltas A, Tozoğlu Ü, Keleş M, Canakci V (2012). Assessment of oral health in peritoneal dialysis patients with and without diabetes mellitus. *Peritoneal Dialysis International* 32(1)*:*81-85.

Emanuel EJ, Fairclough DL, Slutsman J, Emanuel LL (2000). Understanding economic and other burdens of terminal illness: the experience of patients and their caregivers. *Annals of Internal Medicine* 132(6)*:*451-459.

Engel GL (1977). The need for a new medical model: a challenge for biomedicine. *Science* 196(4286)*:*129-136.

Enger TB, Palm O, Garen T, Sandvik L, Jensen JL (2011). Oral distress in primary Sjögren's syndrome: implications for health-related quality of life. *European Journal of Oral Sciences* 119(6)*:*474-480.

Eyler AE, Wilcox S, Matson-Koffman D, Evenson KR, Sanderson B, Thompson J *et al.* (2002). Correlates of physical activity among women from diverse racial/ethnic groups. *Journal of Women's Health & Gender-based Medicine* 11(3)*:*239-253.

Felix DH, Luker J, Scully C (2012). Oral medicine: 4. Dry mouth and disorders of salivation. *Dental Update* 39(10)*:*738-743.

Felton BJ, Revenson TA (1984). Coping with chronic illness: a study of illness controllability and the influence of coping strategies on psychological adjustment. *Journal of Consulting and Clinical Psychology* 52(3)*:*343.

Fernandes MJ, Ruta DA, Ogden GR, Pitts NB, Ogston SA (2006). Assessing oral health-related quality of life in general dental practice in Scotland: validation of the OHIP-14. *Community Dentistry and Oral Epidemiology* 34(1)*:*53-62.

Ferrans CE, Zerwic JJ, Wilbur JE, Larson JL (2005). Conceptual Model of Health-Related Quality of Life. *Journal of Nursing Scholarship* 37(4)*:*336-342.

Fitzpatrick R, Fletcher A, Gore S, Jones D, Spiegelhalter D, Cox D (1992). Quality of life measures in health care. I: Applications and issues in assessment. *British Medical Journal* 305(6861)*:*1074.

Fleck M, Kern ER, Zhou T, Lang B, Mountz JD (1998). Murine cytomegalovirus induces a Sjögren's syndrome-like disease in C57Bl/6 ‐ lpr/lpr mice. *Arthritis & Rheumatism* 41(12)*:*2175-2184.

Folke S (2009). The subjective meaning of xerostomia—an aggravating misery. *International Journal of Qualitative Studies on Health and Well-being* 4(4)*:*245-255.

Fox PC, Busch K, Baum B (1987). Subjective reports of xerostomia and objective measures of salivary gland performance. *The Journal of the American Dental Association* 115(4)*:*581-584.

Fox PC (2007). Autoimmune Diseases and Sjögren's Syndrome. *Annals of the New York Acadamy of Science* 1098(1)*:*15-21.

Fox PC, Bowman SJ, Segal B, Vivino FB, Murukutla N, Choueiri K *et al.* (2008). Oral involvement in primary Sjögren syndrome. *The Journal of the American Dental Association* 139(12)*:*1592-1601.

Fox RI, Robinson CA, Curd JG, Kozin F, Howelly FV (1986). Sjögren's syndrome. Proposed criteria for classification. *Arthritis & Rheumatism* 29(5)*:*577-585.

Fox RI, Michelson P (2000). Approaches to the treatment of Sjögren's syndrome. *The Journal of Rheumatology Supplement* 61(15.

Fox RI (2005). Sjögren's syndrome. *The Lancet* 366(9482)*:*321-331.

Frisch MB, Cornell J, Villanueva M, Retzlaff PJ (1992). Clinical validation of the Quality of Life Inventory. A measure of life satisfaction for use in treatment planning and outcome assessment. *Psychological Assessment* 4(1)*:*92.

Garcia-Carrasco M, Ramos-Casals M, Rosas J, Pallares L, Calvo-Alen J, Cervera R *et al.* (2002). Primary Sjögren syndrome: clinical and immunologic disease patterns in a cohort of 400 patients. *Medicine (Baltimore)* 81(4)*:*270-280.

Gerdin EW, Einarson S, Jonsson M, Aronsson K, Johansson I (2005). Impact of dry mouth conditions on oral health ‐ related quality of life in older people. *Gerodontology* 22(4)*:*219-226.

Gessain A, Vernant J, Maurs L, Barin F, Gout O, Calender Ad *et al.* (1985). Antibodies to human T-lymphotropic virus type-I in patients with tropical spastic paraparesis. *The Lancet* 326(8452)*:*407-410.

Glassner B, Moreno JD (1989). The Qualitative-quantitative Distinction in the Social sciences. Netherlands: Springer.

González S, Sung H, Sepúlveda D, González M, Molina C (2013). Oral manifestations and their treatment in Sjögren's syndrome. *Oral Diseases* 20(2)*:*153-61.

Gottenberg J-E, Cinquetti G, Larroche C, Combe B, Hachulla E, Meyer O *et al.* (2013). Efficacy of rituximab in systemic manifestations of primary Sjögren's syndrome: results in 78 patients of the AutoImmune and Rituximab registry. *Annals of the Rheumatic Diseases* 72(6)*:*1026-1031.

Greenfield S, Kaplan SH, Ware Jr JE, Yano EM, Frank HJ (1988). Patients' participation in medical care. *Journal of General Internal Medicine* 3(5)*:*448-457.

Grimm P (2010). Social desirability bias. *Wiley International Encyclopedia of Marketing*. Wiley online library.

Grueninger UJ (1995). Arterial hypertension: lessons from patient education. *Patient Education and Counseling* 26(1)*:*37-55.

Guba EG (1990). The paradigm dialog: Sage.

Guba EG, Lincoln YS (1994). Competing paradigms in qualitative research. *Handbook of Qualitative Research* 2:163-194.

Guggenheimer J (2003). Xerostomia etiology, recognition and treatment. *The Journal of the American Dental Association* 134(1)*:*61.

Guyatt GH, Feeny DH, Patrick DL (1993). Measuring health-related quality of life. *Annals of Internal Medicine* 118(8)*:*622-629.

Haase JE, Myers ST (1988). Reconciling paradigm assumptions of qualitative and quantitative research. *Western Journal of Nursing Research* 10(2)*:*128-137.

Haddad J, Trinchet J, Pateron D, Mal F, Beaugrand M, Munz-Gotheil C *et al.* (1992). Lymphocytic sialadenitis of Sjögren's syndrome associated with chronic hepatitis C virus liver disease. *The Lancet* 339(8789)*:*321-323.

Halse A-K, Marthinussen M, Wahren-Herlenius M, Jonsson R (2000). Isotype distribution of anti-Ro/SS-A and anti-La/SS-B antibodies in plasma and saliva of patients with Sjögren's syndrome. *Scandinavian Journal of Rheumatology* 29(1)*:*13-19.

Hamburger J (2001). Sjögren's syndrome as seen by an oral physician. *Scandinavian Journal of Rheumatology* 30(115)*:*34-39.

Hartz MB, Beal JR (2000). Patients' attitudes and comfort levels regarding medical students' involvement in obstetrics-gynecology outpatient clinics. *Academic Medicine* 75(10)*:*1010-1014.

Hay K, Morton R, Wall C (2001). Quality of life and nutritional studies in Sjögren's syndrome patients with xerostomia. *The New Zealand Dental Journal* 97(430)*:*128-131.

Haynes K (2006). A therapeutic journey?: Reflections on the effects of research on researcher and participants. *Qualitative Research in Organizations and Management: An International Journal* 1(3)*:*204-221.

Heo S, Moser DK, Riegel B, Hall LA, Christman N (2005). Testing a published model of health-related quality of life in heart failure. *Journal of Cardiac Failure* 11(5)*:*372-379.

Heslin KC, Stein JA, Heinzerling KG, Pan D, Magladry C, Hays RD (2011). Clinical correlates of health-related quality of life among opioid-dependent patients. *Quality of Life Research* 20(8)*:*1205-1213.

Hida A, Akahoshi M, Takagi Y, Ashizawa K, Imaizumi M, Soda M *et al.* (2008). Prevalence of Sjögren syndrome among Nagasaki atomic bomb survivors. *Annals of the Rheumatic Diseases* 67(5)*:*689-695.

Hochberg MS (2007). The Doctor's White Coat--an Historical Perspective. *Virtual Mentor* 9(4)*:*310.

Holman H, Lorig K (2000). Patients as partners in managing chronic disease. *British Medical Journal* 320(7234)*:*526-527.

Hopcraft M, Tan C (2010). Xerostomia: an update for clinicians. *Australian Dental Journal* 55(3)*:*238-244.

Humphrey SP, Williamson RT (2001). A review of saliva: normal composition, flow, and function. *The Journal of Prosthetic Dentistry* 85(2)*:*162-169.

Ikebe K, Matsuda K, Morii K, Wada M, Hazeyama T, Nokubi T *et al.* (2007). Impact of dry mouth and hyposalivation on oral health-related quality of life of elderly Japanese. *Oral Surgery Oral Medicine Oral Pathology Oral Radiolology and Endodontology* 103(2)*:*216-222.

Ito I, Nagai S, Kitaichi M, Nicholson AG, Johkoh T, Noma S *et al.* (2005). Pulmonary manifestations of primary Sjögren's syndrome: a clinical, radiologic, and pathologic study. *American Journal of Respiratory and Critical Care Medicine* 171(6)*:*632-638.

Jagosh J, Donald Boudreau J, Steinert Y, MacDonald ME, Ingram L (2011). The importance of physician listening from the patients' perspective: Enhancing diagnosis, healing, and the doctor–patient relationship. *Patient Education and Counseling* 85(3)*:*369-374.

Janz NK, Janevic MR, Dodge JA, Fingerlin TE, Schork MA, Mosca LJ *et al.* (2001). Factors influencing quality of life in older women with heart disease. *Medical Care* 39(6)*:*588-598.

Jenkins JB (2010). Factors influencing quality of life in older adults with osteoarthritis prior to total knee replacement. Digital online collection of knowledge and scholarship. The University of North Carolina at Greensboro.

Jensen SB, Vissink A (2014). Salivary Gland Dysfunction and Xerostomia in Sjögren's Syndrome. *Oral and Maxillofacial Surgery Clinics of North America* 26(1)*:*35-53.

Johnstone PA, Peng YP, May BC, Inouye WS, Niemtzow RC (2001). Acupuncture for pilocarpine-resistant xerostomia following radiotherapy for head and neck malignancies. *International Journal of Radiation Oncology* Biology* Physics* 50(2)*:*353-357.

Jonsson R, Bolstad AI, Brokstad KA, Brun JG (2007). Sjögren's syndrome—A plethora of clinical and immunological phenotypes with a complex genetic background. *Annals of the New York Acadamy of Sciences* 1108(1)*:*433-447.

Jonsson R, Brun JG (2010). Sjögren's syndrome: Wiley Online Library.

Jonsson R, Vogelsang P, Volchenkov R, Espinosa A, Wahren-Herlenius M, Appel S (2011). The complexity of Sjögren's syndrome: novel aspects on pathogenesis. *Immunology Letters* 141(1)*:*1-9.

Jonsson R, Theander E, Sjöström B, Brokstad K, Henriksson G (2013). Autoantibodies Present Before Symptom Onset in Primary Sjögren Syndrome. *The Journal of the American Medical Association* 310(17)*:*1854-1855.

Kabasakal Y, Kitapcioglu G, Turk T, Öder G, Durusoy R, Mete N *et al.* (2006). The prevalence of Sjögren's syndrome in adult women. *Scandinavian Journal of Rheumatology* 35(5)*:*379-383.

Kassan S, Thomas T, Moutsopoulos H, Hoover R, Kimberly R, Budman D *et al.* (1978). Increased risk of lymphoma in sicca syndrome. *Annals of Internal Medicine* 89(6)*:*888-892.

Kassan SS, Moutsopoulos HM (2004). Clinical manifestations and early diagnosis of Sjögren syndrome. *Archives of Internal Medicine* 164(12)*:*1275-1284.

Kennedy AP, Rogers AE (2002). Improving patient involvement in chronic disease management: the views of patients, GPs and specialists on a guidebook for ulcerative colitis. *Patient Education and Counseling* 47(3)*:*257-263.

Klein H, Palmer CE, Knutson JW (1938). Studies on dental caries. I. dental status and dental needs of elementary school children. *Public Health Reports* 53:751-756.

Knafl K, Breitmayer B, Gallo A, Zoeller L (1996). Family response to childhood chronic illness: Description of management styles. *Journal of Pediatric Nursing* 11(5)*:*315-326.

Kolavic SA, Gibson G, Al‐Hashimi I, Guo IY (1997). The level of cariogenic microlorganisms in patients with Sjögren's syndrome. *Special Care in Dentistry* 17(2)*:*65-69.

Konttinen YT, Kasna-Ronkainen L (2002). Sjögren's syndrome: viewpoint on pathogenesis. One of the reasons I was never asked to write a textbook chapter on it. *Scandinavian Journal of Rheumatology Supplement* 116(15-22.

Kralik D, Brown M, Koch T (2001). Women's experiences of 'being diagnosed' with a long-term illness. *Journal of Advanced Nursing* 33(5)*:*594-602.

Krupp LB, LaRocca NG, Muir-Nash J, Steinberg AD (1989). The fatigue severity scale: application to patients with multiple sclerosis and systemic lupus erythematosus. *Archives of Neurology* 46(10)*:*1121.

Kruszka P, O'Brian RJ (2009). Diagnosis and management of Sjögren syndrome. *American Family Physician* 79(6).

Lapadat JC, Lindsay AC (1999). Transcription in research and practice: From standardization of technique to interpretive positionings. *Qualitative Inquiry* 5(1)*:*64-86.

Larsson K, Lööf L, Rönnblom A, Nordin K (2008). Quality of life for patients with exacerbation in inflammatory bowel disease and how they cope with disease activity. *Journal of Psychosomatic Research* 64(2)*:*139-148.

Leidy NK (1994). Functional status and the forward progress of merry-go-rounds: toward a coherent analytical framework. *Nursing Research* 43(4)*:*196-202.

Leone CW, Oppenheim FG (2001). Physical and chemical aspects of saliva as indicators of risk for dental caries in humans. *Journal of Dental Education* 65(10)*:*1054-1062.

Lessard CJ, Li H, Adrianto I, Ice JA, Rasmussen A, Grundahl KM *et al.* (2013). Variants at multiple loci implicated in both innate and adaptive immune responses are associated with Sjögren's syndrome. *Nature Genetics* 45(11)*:*1284-1292.

Lincoln YS (1985). Naturalistic Inquiry. Beverly Hills, CA: Sage.

Lincoln YS, Lynham SA, Guba EG (2011). Paradigmatic controversies, contradictions, and emerging confluences, revisited. *Denzin HK, Lincoln YS Handbook of Qualitative Research USA: SAGE Publications, Inc*:97.

Lindsay S (2009). Prioritizing illness: lessons in self-managing multiple chronic diseases. *Canadian Journal of Sociology* 34(4)*:*983-1002.

Locker D (1988). Measuring oral health: a conceptual framework. *Community Dental Health* 5(1)*:*3-18.

Locker D (1995). Xerostomia in older adults: a longitudinal study. *Gerodontology* 12(1)*:*18-25.

Locker D (1998). Issues in measuring change in self-perceived oral health status. *Community Dentistry and Oral Epidemiology* 26(1)*:*41-47.

Locker D (2001). *Oral health indicators and determinants for population oral health surveys*. Toronto: University of Toronto.

Locker D, Matear D, Stephens M, Lawrence H, Payne B (2001). Comparison of the GOHAI and OHIP-14 as measures of the oral health-related quality of life of the elderly. *Community Dentistry and Oral Epidemiology* 29(5)*:*373-381.

Locker D (2003a). Dental status, xerostomia and the oral health-related quality of life of an elderly institutionalized population. *Special Care in Dentistry* 23(3)*:*86-93.

Locker D, Gibson B (2005). Discrepancies between self‐ratings of and satisfaction with oral health in two older adult populations. *Community Dentistry and Oral Epidemiology* 33(4)*:*280-288.

Locker D, Allen F (2007). What do measures of 'oral health-related quality of life' measure? *Community Dentistry and Oral Epidemiology* 35(6)*:*401-411.

Locker D, Berka E, Jokovic A, Tompson B (2007). Does self-weighting of items enhance the performance of an oral health-related quality of life questionnaire? *Community Dentistry and Oral Epidemiology* 35(1)*:*35-43.

Locker D, Quinonez C (2009). Functional and psychosocial impacts of oral disorders in Canadian adults: a national population survey. *Journal of the Canadian Dental Association* 75(7)*:*521a-e.

Loiseau P, Lepage V, Djelal F, Busson M, Tamouza R, Raffoux C *et al.* (2001). HLA class I and class II are both associated with the genetic predisposition to primary Sjögren syndrome. *Human Immunology* 62(7)*:*725-731.

Lorig K, Chastain RL, Ung E, Shoor S, Holman HR (1989). Development and evaluation of a scale to measure perceived self-efficacy in people with arthritis. *Arthritis & Rheumatism* 32(1)*:*37-44.

Lovelace TL, Fox NF, Sood AJ, Nguyen SA, Day TA (2014). Management of radiotherapy-induced salivary hypofunction and consequent xerostomia in patients with oral or head and neck cancer: meta-analysis and literature review. *Oral surgery, Oral Medicine, Oral Pathology and Oral Radiology* 117(5)*:*595-607.

Lundström I, Lindström F (1994). Subjective and clinical oral symptoms in patients with primary Sjögren's syndrome. *Clinical and Experimental Rheumatology* 13(6)*:*725-731.

Lutz W, Sanderson W, Scherbov S (2008). The coming acceleration of global population ageing. *Nature* 451(7179)*:*716-719.

MacKinnon DP, Lockwood CM, Hoffman JM, West SG, Sheets V (2002). A comparison of methods to test mediation and other intervening variable effects. *Psychological Methods* 7(1)*:*83.

Mandl T, Ekberg O, Wollmer P, Manthorpe R, Jacobsson L (2007). Dysphagia and dysmotility of the pharynx and oesophagus in patients with primary Sjögren's syndrome. *Scandinavian Journal of Rheumatology* 36(5)*:*394-401.

Manthorpe R, Oxholm P, Prause J, Schiødt M (1985). The Copenhagen criteria for Sjögren's syndrome. *Scandinavian Journal of Rheumatology Supplement* 61:19-21.

Martín-Piedra M, Gómez-Moreno G, Herrera D, Aguilar-Salvatierra A (2011). Effectiveness of a recent topical sialogogue in the management of drug-induced xerostomia. *Journal of Clinical and Experimental Dentistry* 3(4)*:*e268-273.

Mavragani CP, Moutsopoulos HM (2010). The geoepidemiology of Sjögren's syndrome. *Autoimmunity Reviews* 9(5)*:*A305-A310.

Maxwell JA (2012). Qualitative Research Design: An Interactive Approach: An Interactive Approach. Thousand Oaks, CA: Sage.

Mays N, Pope C (1995). Qualitative research: rigour and qualitative research. *British Medical Journal* 311(6997)*:*109-112.

McCracken LM, Eccleston C (2003). Coping or acceptance: what to do about chronic pain? *Pain* 105(1)*:*197-204.

McGrath C, Bedi R (2004). Why are we 'weighting' ? An assessment of a self-weighting approach to measuring oral health-related quality of life. *Community Dentistry and Oral Epidemiology* 32(1)*:*19-24.

McLeroy KR, Bibeau D, Steckler A, Glanz K (1988). An ecological perspective on health promotion programs. *Health Education & Behavior* 15(4)*:*351-377.

Meijer JM, Meiners PM, Slater JJH, Spijkervet FK, Kallenberg CG, Vissink A *et al.* (2009). Health-related quality of life, employment and disability in patients with Sjögren's syndrome. *Rheumatology* 48(9)*:*1077-1082.

Meijer JM, Meiners P, Vissink A, Spijkervet F, Abdulahad W, Kamminga N *et al.* (2010). Effectiveness of rituximab treatment in primary Sjögren's syndrome: A randomized, double-blind, placebo-controlled trial. *Arthritis & Rheumatism* 62(4)*:*960-968.

Mengshoel AM, Norheim KB, Omdal R (2013). Primary Sjögren's syndrome‐Fatigue is an ever ‐ present, fluctuating and uncontrollable lack of energy. *Arthritis Care & Research*.

Mignogna M, Fedele S, Russo LL, Muzio LL, Wolff A (2005). Sjögren's syndrome: the diagnostic potential of early oral manifestations preceding hyposalivation/xerostomia. *Journal of Oral Pathology & Medicine* 34(1)*:*1-6.

Miljanović B, Dana R, Sullivan DA, Schaumberg DA (2007). Impact of dry eye syndrome on vision-related quality of life. *American Journal of Ophthalmology* 143(3)*:*409-415.e402.

Napeñas JJ, Brennan MT, Fox PC (2009). Diagnosis and treatment of xerostomia (dry mouth). *Odontology* 97(2)*:*76-83.

Napeñas JJ, Rouleau TS (2014). Oral Complications of Sjögren's Syndrome. *Oral and Maxillofacial Surgery Clinics of North America* 26(1)*:*55-62.

Närhi T (1994). Prevalence of subjective feelings of dry mouth in the elderly. *Journal of Dental Research* 73(1)*:*20-25.

Navazesh M (1993). Methods for Collecting Saliva. *Annals of the New York Acadamy of Science* 694(1)*:*72-77.

Navazesh M, Kumar SK (2008). Measuring salivary flow Challenges and opportunities. *The Journal of the American Dental Association* 139(Supplement 2)*:*35S-40S.

Nederfors T, Isaksson R, Mörnstad H, Dahlöf C (1997). Prevalence of perceived symptoms of dry mouth in an adult Swedish population-relation to age, sex and pharmacotherapy. *Community Dentistry and Oral Epidemiology* 25(3)*:*211-216.

Nederfors T (2000). Xerostomia and hyposalivation. *Advances in Dental Research* 14(1)*:*48-56.

Nordenram G, Davidson T, Gynther G, Helgesson G, Hultin M, Jemt T *et al.* (2013). Qualitative studies of patients' perceptions of loss of teeth, the edentulous state and prosthetic rehabilitation: A systematic review with meta-synthesis. *Acta Odontologica Scandinavica* 71(3-4)*:*937-951.

Northern KP (2001). Effect of a self-management program on patients with chronic disease. *Effective Clinical Practice* 4(6)*:*256-262.

O'Neill I, Scully C (2013). Biologics in oral medicine: Sjögren syndrome. *Oral Diseases* 19(2)*:*121-127.

Ochsendorf F, Runne U (1991). Chloroquine and hydroxychloroquine: side effect profile of important therapeutic drugs. *Der Hautarzt; Zeitschrift fur Dermatologie, Venerologie, und verwandte Gebiete* 42(3)*:*140-146.

Orellana MF, Lagravere MO, Boychuk DG, Major PW, Flores-Mir C (2006). Prevalence of xerostomia in population-based samples: a systematic review. *Journal of Public Health Dentistry* 66(2)*:*152-158.

Orzeł-Gryglewska J (2010). Consequences of sleep deprivation. *International Journal of Occupational Medicine and Environmental Health* 23(1)*:*95-114.

Österberg T, Landahl S, Hedegård B (1984). Salivary flow, saliva, pH and buffering capacity in 70-year-old men and women. *Journal of Oral Rehabilitation* 11(2)*:*157-170.

Owens J, Gibson BJ, Periyakaruppiah K, Baker SR, Robinson PG (2014). Impairment effects, disability and dry mouth: Exploring the public and private dimensions. *Health* 18(5)*:*509-525.

Pai S, Ghezzi EM, Ship JA (2001). Development of a Visual Analogue Scale questionnaire for subjective assessment of salivary dysfunction. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 91(3)*:*311-316.

Panja S, Starr B, Colleran KM (2005). Patient knowledge improves glycemic control: is it time to go back to the classroom? *Journal of Investigative Medicine* 53(5)*:*264-266.

Patterson A (2005). Processes, relationships, settings, products and consumers: the case for qualitative diary research. *Qualitative Market Research* 8(2)*:*142-156.
Patton MQ (1987). How to use qualitative methods in evaluation. Newbury Park, CA: Sage.

Pons-Estel GJ, Pons-Estel BA, Alarcón GS (2012). Musculoskeletal Involvement. In: Sjögren's Syndrome. London: Springer.

Quandt SA, Savoca MR, Leng X, Chen H, Bell RA, Gilbert GH *et al.* (2011). Dry mouth and dietary quality in older adults in North Carolina. *Journal of the American Geriatrics Society* 59(3)*:*439-445.

Ramos-Casals M, Brito-Zerón P, Perez-De-Lis M, Jimenez I, Blanco M-J, Bove A *et al.* (2010a). Sjögren syndrome or Sjögren disease? The histological and immunological bias caused by the 2002 criteria. *Clinical Reviews in Allergy & Immunology* 38(2-3)*:*178-185.

Ramos-Casals M, Tzioufas AG, Stone JH, Sisó A, Bosch X (2010b). Treatment of Primary Sjögren SyndromeA Systematic Review. *The Journal of the American Medical Association* 304(4)*:*452-460.

Reveille JD, Wilson RW, Provost TT, Bias WB, Arnett FC (1984). Primary Sjögren's syndrome and other autoimmune diseases in families. Prevalence and immunogenetic studies in six kindreds. *Annals of Internal Medicine* 101(6)*:*748-756.

Rubin HJ, Rubin IS (2011). Qualitative interviewing: The art of hearing data. Thousand Oaks, CA: Sage.

Rydholm M, Strang P (2002). Physical and psychosocial impact of xerostomia in palliative cancer care: a qualitative interview study. *International Journal of Palliative Nursing* 8(7)*:*318-323.

Saito I, Servenius B, Compton T, Fox RI (1989). Detection of Epstein-Barr virus DNA by polymerase chain reaction in blood and tissue biopsies from patients with Sjögren's syndrome. *The Journal of Experimental Medicine* 169(6)*:*2191-2198.

Sardenberg F, Goursand D, Polletto LT, Vale MP, Zarzar PM, Paiva SM (2010). Oral Manifestations and Treatment of a Child with Sjögren's Syndrome. *Journal of Dentistry for Children* 77(2)*:*102-105.

Scharloo M, Kaptein A, Weinman J, Hazes J, Willems L, Bergman W *et al.* (1998). Illness perceptions, coping and functioning in patients with rheumatoid arthritis,

chronic obstructive pulmonary disease and psoriasis. *Journal of Psychosomatic Research* 44(5)*:*573-585.

Schoofs N (2001). Seeing the glass half full: Living with Sjögren's Syndrome. *Journal of Professional Nursing* 17(4)*:*194-202.

Schwartz CE, Sendor RM (1999). Helping others helps oneself: response shift effects in peer support. *Social Science & Medicine* 48(11)*:*1563-1575.

Scully C, Felix D (2005). 3: Oral Medicine—Update for the dental practitioner: Dry mouth and disorders of salivation. *British Dental Journal* 199(7)*:*423-427.
Scully C (2010). Medical Problems in Dentistry. UK: Elsevier Health Sciences.

Segal B, Thomas W, Rogers T, Leon JM, Hughes P, Patel D *et al.* (2008). Prevalence, severity, and predictors of fatigue in subjects with primary Sjögren's syndrome. *Arthritis Care & Research* 59(12)*:*1780-1787.

Segal B, Bowman SJ, Fox PC, Vivino FB, Murukutla N, Brodscholl J *et al.* (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.

Seror R, Ravaud P, Bowman SJ, Baron G, Tzioufas A, Theander E *et al.* (2010). EULAR Sjögren's syndrome disease activity index: development of a consensus systemic disease activity index for primary Sjögren's syndrome. *Annals of the Rheumatic Diseases* 69(6)*:*1103-1109.

Seror R, Ravaud P, Mariette X, Bootsma H, Theander E, Hansen A *et al.* (2011). EULAR Sjögren's Syndrome Patient Reported Index (ESSPRI): development of a consensus patient index for primary Sjögren's syndrome. *Annals of the Rheumatic Diseases* 70(6)*:*968-972.

Seror R, Bootsma H, Bowman SJ, Dörner T, Gottenberg J-E, Mariette X *et al.* (2012). Outcome measures for primary Sjögren's syndrome. *Journal of Autoimmunity* 39(1)*:*97-102.

Shiboski SC, Shiboski CH, Criswell L, Baer A, Challacombe S, Lanfranchi H *et al.* (2012). American College of Rheumatology classification criteria for Sjögren's syndrome: a data-driven, expert consensus approach in the Sjögren's International Collaborative Clinical Alliance cohort. *Arthritis Care & Research (Hoboken)* 64(4)*:*475-487.

Skopouli FN, Dafni U, Ioannidis J, Moutsopoulos HM Clinical evolution, and morbidity and mortalityof primary Sjögren's syndrome. Seminars in Arthritis and Rheumatology 2000: Elsevier.

Slade GD, Spencer AJ (1994). Development and evaluation of the oral health impact profile. *Community Dental Health* 11(1)*:*3-11.

Slade GD Measuring oral health and quality of life. Background and Rationale for the Conference 1996. Chapel Hill: University of North Carolina.

Slade GD, Spencer AJ, Locker D, Hunt R, Strauss R, Beck JD (1996). Variations in the social impact of oral conditions among older adults in South Australia, Ontario, and North Carolina. *Journal of Dental Research* 75(7)*:*1439-1450.

Slade GD (1997). Derivation and validation of a short-form oral health impact profile. *Community Dentistry and Oral Epidemiolology* 25(4)*:*284-290.

Slade GD (1998). Assessing change in quality of life using the oral health impact profile. *Community Dentistry and Oral Epidemiology* 26(1)*:*52-61.
Slevin ML, Plant H, Lynch Da, Drinkwater J, Gregory W (1988). Who should measure quality of life, the doctor or the patient? *British Journal of Cancer* 57(1)*:*109.

Soto-Rojas AE, Kraus A (2002). The oral side of Sjögren syndrome. Diagnosis and treatment. A review. *Archives of Medical Research* 33(2)*:*95-106.

Sousa KH, Kwok O-M (2006). Putting Wilson and Cleary to the test: analysis of a HRQOL conceptual model using structural equation modeling. *Quality of Life Research* 15(4)*:*725-737.

Southwood TR, Malleson PN, Roberts-Thomson PJ, Mahy M (1990). Unconventional remedies used for patients with juvenile arthritis. *Pediatrics* 85(2)*:*150-154.

Soy M, Piskin S (2007). Cutaneous findings in patients with primary Sjögren's syndrome. *Clinical Rheumatology* 26(8)*:*1350-1352.

Spirduso WW, Cronin DL (2001). Exercise dose–response effects on quality of life and independent living in older adults. *Medicine & Science in Sports & Exercise*.

Sprangers MA, Schwartz CE (1999). Integrating response shift into health-related quality of life research: a theoretical model. *Social Science & Medicine* 48(11)*:*1507-1515.

Sreebny LM, Schwartz SS (1997). A reference guide to drugs and dry mouth–2nd edition. *Gerodontology* 14(1)*:*33-47.

Sreebny LM (2000). Saliva in health and disease: an appraisal and update. *International Dental Journal* 50(3)*:*140-161.

Stanton AL, Revenson TA, Tennen H (2007). Health psychology: psychological adjustment to chronic disease. *Annual Review of Psychology* 58(565-592.

Stewart A, Ware JE (1992). Measuring functioning and well-being. Durham: Duke University Press.

Stewart CM, Berg KM, Cha S, Reeves WH (2008). Salivary dysfunction and quality of life in Sjögren syndrome: a critical oral-systemic connection. *The Journal of the American Dental Association (1939)* 139(3)*:*291-299; quiz 358-299.

Stineman M, Lollar D, Üstün T (2005). The International Classification of Functioning, Disability, and Health: ICF empowering rehabilitation through an operational bio-psycho-social model. *Physical Medicine and Rehabilitation: Principles and Practice:*1099-1108.

Stoves J, Connolly J, Cheung CK, Grange A, Rhodes P, O'Donoghue D *et al.* (2010). Electronic consultation as an alternative to hospital referral for patients with chronic kidney disease: a novel application for networked electronic health records to improve the accessibility and efficiency of healthcare. *Quality and Safety in Health Care* 19(5)*:*e54-e54.

Street Jr RL, Haidet P (2011). How well do doctors know their patients? Factors affecting physician understanding of patients' health beliefs. *Journal of General Internal Medicine* 26(1)*:*21-27.

Strietzel FP, Lafaurie GI, Mendoza GRB, Alajbeg I, Pejda S, Vuletić L *et al.* (2011). Efficacy and safety of an intraoral electrostimulation device for xerostomia relief: a multicenter, randomized trial. *Arthritis & Rheumatism* 63(1)*:*180-190.

Taiym S, Haghighat N, Al-Hashimi I (2004). A comparison of the hormone levels in patients with Sjögren's syndrome and healthy controls. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 97(5)*:*579-583.

Tasaki K, Maskarinec G, Shumay DM, Tatsumura Y, Kakai H (2002). Communication between physicians and cancer patients about complementary and alternative medicine: exploring patients' perspectives. *Psycho-Oncology* 11(3)*:*212-220.

Taylor SE, Armor DA (1996). Positive illusions and coping with adversity. *Journal of Personality* 64(4)*:*873-898.

Terada K, Katamine S, Moriuchi R, Miyamoto T, Eguchi K, Kita M *et al.* (1994). Prevalence of serum and salivary antibodies to HTLV-1 in Sjögren's syndrome. *The Lancet* 344(8930)*:*1116-1119.

Theander E, Vasaitis L, Baecklund E, Nordmark G, Warfvinge G, Liedholm R *et al.* (2011). Lymphoid organisation in labial salivary gland biopsies is a possible predictor for the development of malignant lymphoma in primary Sjögren's syndrome. *Annals of the Rheumatic Diseases* 70(8)*:*1363-1368.

Theander E, Wollheim FA (2012). A Note from Sweden: Recollection of Henrik Sjögren. In: Sjögren's Syndrome. Oklahoma: Springer.

Thomas E, Hay E, Hajeer A, Silman A (1998). Sjögren's syndrome: a community-based study of prevalence and impact. *Rheumatology* 37(10)*:*1069-1076.

Thomson WM, Williams S (2000). Further testing of the xerostomia inventory. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 89(1)*:*46-50.

Thomson WM, Chalmers JM, Spencer AJ, Ketabi M (1999a). The occurrence of xerostomia and salivary gland hypofunction in a population‐based sample of older South Australians. *Special Care in Dentistry* 19(1)*:*20-23.

Thomson WM, Chalmers JM, Spencer AJ, Williams SM (1999b). The Xerostomia Inventory: a multi-item approach to measuring dry mouth. *Community Dental Health* 16(1)*:*12-17.

Thomson WM (2005). Issues in the epidemiological investigation of dry mouth. *Gerodontology* 22(2)*:*65-76.

Thomson WM, Chalmers JM, John Spencer A, Slade GD, Carter KD (2006a). A longitudinal study of medication exposure and xerostomia among older people. *Gerodontology* 23(4)*:*205-213.

Thomson WM, Poulton R, Broadbent JM, Al-Kubaisy S (2006b). Xerostomia and medications among 32-year-olds. *Acta Odontolologica Scandinavica* 64(4)*:*249-254.

Thomson WM, Lawrence HP, Broadbent JM, Poulton R (2006c). The impact of xerostomia on oral-health-related quality of life among younger adults. *Health and Quality of Life Outcomes* 4*:*86.

Thomson WM (2007). Measuring change in dry-mouth symptoms over time using the Xerostomia Inventory. *Gerodontology* 24(1)*:*30-35.

Thomson WM, van der Putten GJ, de Baat C, Ikebe K, Matsuda K, Enoki K *et al.* (2011). Shortening the xerostomia inventory. *Oral Surgery, Oral Medicine, Oral Pathololology, Oral Radiolology, and Endodontology* 112(3)*:*322-327.

Thomson WM, Mejia GC, Broadbent JM, Poulton R (2012). Construct validity of Locker's global oral health item. *Journal of Dental Research* 91(11)*:*1038-42

Thomson WM. *Subjective aspects of dry mouth*. In: *Dry mouth: a clinical guide on causes, effects and treatments*. Edited by Guy Carpenter. Springer Science and Business Media, in press.

Tobón GJ, Renaudineau Y, Hillion S, Cornec D, Devauchelle-Pensec V, Youinou P *et al.* (2010). The Fms‐like tyrosine kinase 3 ligand, a mediator of B cell survival, is also a marker of lymphoma in primary Sjögren's syndrome. *Arthritis & Rheumatism* 62(11)*:*3447-3456.

Tsifetaki N, Kitsos G, Paschides C, Alamanos Y, Eftaxias V, Voulgari P *et al.* (2003). Oral pilocarpine for the treatment of ocular symptoms in patients with Sjögren's syndrome: a randomised 12 week controlled study. *Annals of Rheumatic Diseases* 62(12)*:*1204-1207.

Tsubota K (2012). New Approaches for the Management of Dry Mouth and Dry Eye in Sjögren's Syndrome in Japan. In: Sjögren's Syndrome: Springer, pp. 415-421.

Turner MD, Ship JA (2007). Dry mouth and its effects on the oral health of elderly people. *The Journal of the American Dental Association* 138(suppl 1)*:*15S-20S.

Tzioufas AG, Moutsopoulos H (2003). Sjögren's syndrome. In: European Handbook of Dermatological Treatments. Berlin: Springer.

Valtýsdóttir ST, Gudbjörnsson B, Lindqvist U, Hällgren R, Hetta J (2000). Anxiety and depression in patients with primary Sjögren's syndrome. *The Journal of Rheumatology* 27(1*)*:165-169.

Vitali C, Bombardieri S, Moutsopoulos HM, Balestrieri G, Bencivelli W, Bernstein RM *et al.* (1993). Preliminary criteria for the classification of Sjögren's syndrome. Results of a prospective concerted action supported by the European Community. *Arthritis & Rheumatism* 36(3)*:*340-347.

Vitali C, Bombardieri S, Jonsson R, Moutsopoulos H, Alexander E, Carsons S *et al.* (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(6)*:*554-558.

von Bultzingslowen I, Sollecito TP, Fox PC, Daniels T, Jonsson R, Lockhart PB *et al.* (2007). Salivary dysfunction associated with systemic diseases: systematic review and clinical management recommendations. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiolology, and Endodontology* 103 Supplement S57*:*e51-15.

Wade DT, Halligan PW (2004). Do biomedical models of illness make for good healthcare systems? *British Medical Journal* 329(7479)*:*1398.

Ware JE (2004). SF-36 health survey update. *The Use of Psychological Testing for Treatment Planning and Outcomes Assessment* 3(693-718.

Ware JE, Jr., Sherbourne CD (1992). The MOS 36-item short-form health survey (SF-36). I. Conceptual framework and item selection. *Medical Care* 30(6)*:*473-483.

Waterman SA, Gordon TP, Rischmueller M (2000). Inhibitory effects of muscarinic receptor autoantibodies on parasympathetic neurotransmission in Sjögren's syndrome. *Arthritis & Rheumatism* 43(7)*:*1647-1654.

Westhoff G, Dörner T, Zink A (2012). Fatigue and depression predict physician visits and work disability in women with primary Sjögren's syndrome: results from a cohort study. *Rheumatology* 51(2)*:*262-269.

Wettergren L, Björkholm M, Axdorph U, Langius-Eklöf A (2004). Determinants of health-related quality of life in long-term survivors of Hodgkin's lymphoma. *Quality of Life Research* 13(8)*:*1369-1379.

Williams MV, Baker DW, Parker RM, Nurss JR (1998). Relationship of functional health literacy to patients' knowledge of their chronic disease: a study of patients with hypertension and diabetes. *Archives of Internal Medicine* 158(2)*:*166-172.

Wilson IB, Cleary PD (1995). Linking clinical variables with health-related quality of life. *The Journal of the American Medical Association* 273(1)*:*59-65.

World Health Organization (1948). WHO Constitution; 1948. *Official Records of the World Health Organization* 2:100.

World Health Organization (1980). International classification of impairments, disabilities, and handicaps: a manual of classification relating to the consequences of disease, published in accordance with resolution WHA29. 35 of the Twenty-ninth World Health Assembly, May 1976.

Yan Z, Young AL, Hua H, Xu Y (2011). Multiple Oral Candida Infections in Patients with Sjögren's Syndrome—Prevalence and Clinical and Drug Susceptibility Profiles. *The Journal of Rheumatology* 38(11)*:*2428-2431.

Zhang N, Shi CS, Yao QP, Pan G, Wang L, Wen Z *et al.* (1995). Prevalence of primary Sjögren's syndrome in China. *The Journal of Rheumatology* 22(4)*:*659-661.

Zigmond AS, Snaith RP (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica* 67(6)*:*361-370.

Zufferey P, Meyer O, Grossin M, Kahn M (1995). Primary Sjögren's syndrome (SS) and malignant lymphoma: a retrospective cohort study of 55 patients with SS. *Scandinavian Journal of Rheumatology* 24(6)*:*342-345.