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World Workshop on Oral Medicine VIII: Development of a core outcome set for dry mouth: the patient perspective

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Objective. We conducted a qualitative study of patients' perspectives on dry mouth outcomes to explore their personal experiences and investigate which outcomes are most important to them. This work was part of the WONDER initiative (World Workshop on Oral Medicine Outcomes Initiative for the Direction of Research) exploring Core Outcome Measures in Effectiveness Trials.

Study Design. Using a study-specific topic guide, we conducted digitally recorded, semi-structured interviews of focus groups of patients with dry mouth secondary to Sjögren syndrome and head and neck radiotherapy. We conducted interviews until data saturation had been achieved and evaluated all transcripts for accuracy before we anonymized the data.

Results. Two focus groups consisting of 4 participants per group identified 4 distinct themes: (1) impact on oral health and function, (2) social isolation and withdrawal, (3) frustration with dry mouth management, and (4) limited knowledge of the medical community and lack of understanding of family and friends.

Conclusions. The diversity of self-reported outcomes and the complexity of patient perceptions identified in our work may represent additional barriers to successful dry mouth management that should be considered in the design of future clinical trials. (Oral Surg Oral Med Oral Pathol Oral Radiol 2023;135:876–884)

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Human saliva is a unique biofluid produced by major and minor salivary glands and secreted into the oral cavity. In terms of composition, saliva holds around 99% of water enriched by mucin, enzymes (lysozyme, lipase, and amylase), proteins, electrolytes (sodium, calcium, potassium, magnesium, bicarbonate, and phosphates), secretory immunoglobulin A, growth factors (epidermal growth factor, nerve growth factor, basic fibroblast growth factor, transforming growth factor-alpha, and insulin/insulin-like growth factor), and nitrogenous products (urea and ammonia), among other components. After being secreted into the oral cavity, saliva also contains crevicular fluid and a pool of epithelial and inflammatory cells.¹ Saliva protects the dentition from caries and provides lubrication of the oral tissues, allowing proper speech, chewing, and swallowing, among other functions. Moreover, salivary enzymes initiate the digestion process in the oral cavity.

Statement of Clinical Relevance

This qualitative study of patients' perspectives of dry mouth outcomes explored their lived experiences and identified relevant patient-reported outcomes to be potentially included in clinical trials of dry mouth management.

ity.¹ Due to the fundamental role of saliva in maintaining oral health, the impact of dry mouth on patients' daily lives cannot be underestimated.²

Subjective complaints of dry mouth, in professional and diagnostic terminology, also known as *xerostomia*, correspond to the symptom of oral dryness. A quantitative reduction in salivary flow rate does not necessarily accompany dry mouth. Nevertheless, salivary gland hypofunction or hyposalivation diagnosis refers to a quantified decrease in the salivary volume. The underlying causes of salivary gland hypofunction and xerostomia may include medication intake; Sjögren syndrome, also known as Sjögren disease; radiation therapy to the head and neck; senility; and dehydration.³

Although dry mouth is a common condition globally, management options that provide sustained relief of symptoms are limited. Although multiple clinical trials have been carried out, systematic reviews have highlighted limited evidence with which to recommend any sufficiently robust treatment modality. Furthermore, the comparison of the efficacy of treatment modalities and pooling of results of clinical trials have been inconsistent due to a lack of consensus on which outcomes should be recorded in clinical trials for dry mouth.^{4,5}

A core outcome set (COS) is an agreed-upon, standardized set of outcomes that should be measured and reported, as a minimum requirement, in all clinical trials in specific areas of health or health care. According to the Core Outcome Measures in Effectiveness Trials initiative,⁶ the generation of a comprehensive list of outcomes identifying and rectifying deficiencies in existing knowledge should be carried out when designing clinical trials.⁷⁻¹¹ Qualitative interviews with patients experiencing the condition(s) to be explored are also pivotal to identifying outcomes that are important to them, allowing the patient's voice to be incorporated into the COS. This is now considered an integral part of developing a COS¹² and has been adopted by outcome sets developed in allied medical specialties, such as neurology, rheumatology, and respiratory medicine.^{13,14}

Young and Bagley highlighted the importance and benefits of patient involvement in COS design and determining which outcomes should be included in a minimum outcome set.¹⁵ The authors also discussed the various methods used and indicated that the current methods used to generate clinician or researcher opinions might not be comprehensive without patient input. They emphasized the merits of using qualitative research methods to add to the material generated from the initial systematic review's "long list" of outcomes. Therefore, as part of the World Workshop on Oral Medicine Outcomes Initiative for the Direction of

Research (WONDER), this study aimed to explore the personal experience of patients with dry mouth, focusing on the outcomes of care, and to investigate which outcomes were important to the cohort of patients examined.¹⁶

MATERIALS AND METHODS

A qualitative study incorporating focus groups of patients with dry mouth was conducted. These focus groups were semi-structured interviews, using a study specific topic guide. In the reporting of the study, they followed guidance from the Standard for Reporting Qualitative Research.¹⁷ The researchers held each focus group in 2 parts: In Part 1, they explored patients' personal experiences with dry mouth; in Part 2, they allowed the patients to review a summary of the outcomes used in the dry mouth literature. From the patient's perspective, a discussion ensued regarding the importance of the various outcomes found in the literature and which outcome domains of importance to them were absent from the list presented.

Ethical Considerations

The researchers received ethical approval for this study from the Clinical Research Ethics Committee of Cork Teaching Hospitals, Ireland (ECM 3 [rrr] 01/06/2021). The researchers gave the participants a written study-specific information leaflet and asked them to sign an informed consent form.

The Research Team and Reflexivity

The research team comprised 2 researchers, RNiR (Researcher 1) and MO'C (Researcher 2). Researcher 1 was responsible for running the focus groups, whereas Researcher 2 was responsible for recruiting participants. The researchers shared data analysis between them and held a debriefing meeting after each focus group to encourage reflexivity.¹⁸

Participants and Sampling Strategy

The inclusion criteria were age older than 18 years, diagnosis of dry mouth, attendance at the Oral Medicine Unit of Cork University Dental School and Hospital, and ability to speak fluent English. There were no other inclusion or exclusion criteria. The research team asked participants older than 18 years with a diagnosis of dry mouth who attended the Oral Medicine Unit of Cork University Dental School and Hospital whether they would be willing to participate in the study. The sampling strategy was purposive to include participants who had dry mouths of varying etiologies (medication-induced, Sjögren syndrome, or head and neck radiotherapy) and severity. Time since diagnosis was also considered when recruiting participants, with some

recently diagnosed patients and others being long-standing patients of the host Oral Medicine Unit.

The research team contacted 20 patients by telephone between March and April 2022, from which they formed 2 focus groups of 4 patients each (a total of 8 participants) who met in person. The remaining 12 patients, including 4 patients with medication-induced dry mouth, 5 patients with dry mouth post head and neck radiotherapy, and 3 patients with Sjögren syndrome, declined to participate. Among these patients, 2 patients with medication-induced dry mouth declined because they became unwell on the day of the focus group, and 10 because of prior family or work commitments. Each focus group lasted between 40 and 50 minutes. Data analysis was an iterative process, and collection ceased once saturation had been achieved. Data saturation was defined as the moment the data collected had sufficient breadth and depth to answer the research questions and at which no new themes were emerging.¹⁹

Data Collection and Analysis

The research team developed a structured topic guide, with minor edits following feedback from participants in the initial focus group (Table I). Although the topic guide indicated a similar scope of areas to be covered in each focus group, the facilitator allowed conversations to flow between the participants with minimal intervention.²⁰ Therefore, the focus group methodology encouraged group interactions, ensuring participants had the opportunity to share personal experiences

and collectively explore outcomes of dry mouth treatments that were significant to the group.²¹ The focus group discussions were audio recorded and transcribed verbatim.

The researchers used an interpretive methodology to identify themes regarding the patients' personal experiences, keeping in mind the conceptual framework of Locker's Model of Oral Health.^{22,23} They analyzed the transcripts line-by-line and coded them to define the data to be analyzed. They manually performed the data analysis using Microsoft Office Word software (Microsoft Corporation, Redmond, WA, USA), coding identified passages of text that exemplified a descriptive idea and linking them under a single idea or code. This process allowed the transcripts to be categorized and facilitated the identification of key themes. The researchers selected anonymized quotes from the transcripts per key themes and presented them as an emblematic part of the qualitative results.

RESULTS

The patients were aged 45 to 77 years and lived across different parts of Ireland. The demographic and clinico-pathologic data of the participants are summarized in Table II. Two in-focus groups of 4 participants each (8 participants total) met in person and covered 4 different themes (Figure 1 and Table III). The study recruited more women than men (6 versus 2, respectively) with no apparent differences in self-reported dry mouth experiences. Among all 8 patients, 5

Table I. Structured Topic Guide Used for Data Collection

<i>Stages</i>	<i>Approaches</i>
Personal history and knowledge about dry mouth	What do you understand about what has caused your dry mouth? How long have you been living with dry mouth?
Experience of living with dry mouth (how dry mouth has impacted your life)	Quality of life—activity limitations: Have you had problems while eating, drinking, and speaking? Mood: Have you stopped doing things due to the emotional burden of dry mouth? Have you been worrying? Do you think that your mood has any influence on your pain? Family and social interactions: Has anything changed with regard to social interactions? Have you stopped making plans with your friends or family? Have you stopped going out for meals? Productivity: Have you had to take time off of work?
Treatment of dry mouth	What treatments have you had? Are you satisfied with your treatment? Was it successful? What do you understand about the success of treatment?
Treatment outcomes	When you think about your dry mouth: What do you consider to be the most important treatment outcome? When you think about your treatment: Did you consider the side effects/adverse effects before choosing a treatment?
If you had to think about a list of the most important treatment outcomes, what would they be?	Open questions

Table II. Demographic and Clinicopathologic Characteristics of Study Participants

Group	Patient	Age (y)	Sex	Dry mouth etiology
Focus Group 1	1	70	Female	Sjögren syndrome
	2	63	Female	Sjögren syndrome
	3	45	Female	Sjögren syndrome
	4	66	Female	Head and neck radiotherapy
Focus Group 2	1	61	Female	Sjögren syndrome
	2	77	Male	Head and neck radiotherapy
	3	69	Male	Head and neck radiotherapy
	4	73	Female	Sjögren syndrome

experienced dry mouths because of Sjögren syndrome, and 3 experienced it after head and neck radiation therapy.

The patients reported that dry mouth outcomes had a deep multidimensional impact on them, including severity and their everyday life. After categorizing the themes, the researchers selected anonymized verbatim quotes exploring each theme from the transcripts and presented them below, followed by the patient’s study code.

Part 1: Personal Experiences with Treatments And Outcomes

Theme 1: Impact on oral health and function. The discussions in both focus groups focused on the participants’ limited ability to speak, eat, taste, and swallow:

Saliva is so important. I realize now. It does everything. It breaks down your food and everything for you. I miss it so much. For that, to help even to lubricate your mouth, to swallow, to taste (Focus Group 2, Participant 3 [FG2Pt3]).

Taste is completely different. You just don’t taste things (FG1Pt4).

I’d have to have water or milk or something if I wanted to eat. It has to be there. Something to help you when you swallow, even though you are slow at eating, sometimes it just gets lodged. Then especially if you are with some people, you don’t want to panic, you try to keep yourself calm (FG1Pt2).

Digestion for me lately is hard. I know it is from the lack of saliva. . .The problem is breaking down of the food. I feel like I will choke (FG2Pt4).

It is embarrassing with my son and the kids; when I cough, it is like a family emergency: “Get water for her, get water for her, don’t choke.” It really is embarrassing; you feel like a child (FG1Pt2).

The participants discussed the concerns they have about the impact of dry mouth on their dentition and oral mucosa, along with the financial implications of the dental care required:

Everything is so dry, and then the teeth are so sharp that I bite my tongue. It is very sore. It stings and burns after biting. If you took any bite of fruit, it is awful (FG1Pt4).

The plaque forms on the teeth so quickly that it feels horrible. It is a real nuisance. My teeth are getting sharper and sharper. I bite my cheek and tongue (FG1Pt2).

My teeth are so discolored, prone to cavities, my teeth are chipping. I’m constantly going to the dentist; there is a huge financial aspect to it. It big time gets me down (FG2Pt4).

It is very uncomfortable. When you wake in the morning and your tongue is stuck to the roof of your mouth, it is very uncomfortable (FG1Pt1).

What I find as well. . .if you get a small nick in the mouth, I get a huge mouth ulcer then and it takes so

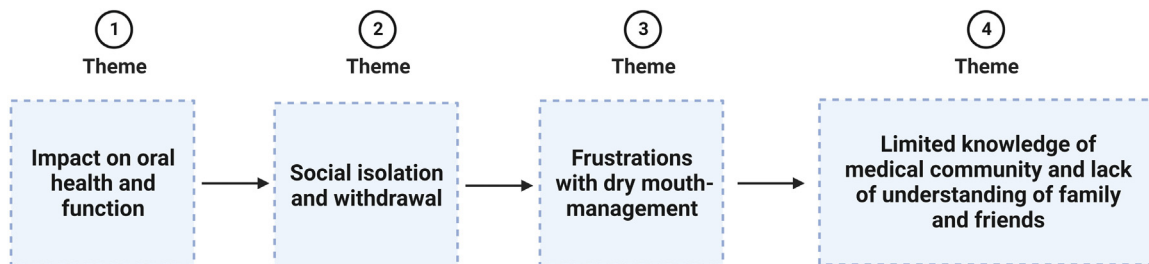


Figure 1. Self-reported most impacting outcome domains experienced by participants with dry mouth included in this qualitative study.

Table III. Outline of Study Themes, Subthemes, and Descriptors

Themes	Subthemes
Theme 1: Impact on oral health and function (impairment—discomfort and pain; functional limitation)	Impact on ability to eat Impact on ability to swallow Impact on taste Impact on ability to speak Oral discomfort Fragility of oral tissues Increasing dental care needs and associated costs
Theme 2: Social impact: isolation, withdrawal and worry for the future (disability, social and psychological)	Withdrawal from social interactions Impact on mood Limitation of social circle Worry about worsening of the condition Pessimism for future
Theme 3: Frustration with dry mouth management (disability, psychological impact)	Limited availability of treatment options Minimal relief of symptoms of concern, such as coughing and choking, with treatments available
Theme 4: Limited knowledge of medical community and lack of understanding of family and friends	Frustration with limited knowledge of health care professionals of the impact of dry mouth Social embarrassment associated with poor understanding when trying to explain oral dryness

long to heal. I have one now on my tongue. It's awful (FG1Pt2).

Theme 2: Social isolation and withdrawal. Withdrawal from social events and family interactions were discussed at length, with participants reflecting on how much smaller their social circle had become and how gradually they had to step back from society more and more each year:

For me it has cut off my life really. I can't walk much; if I meet people, I can't talk to them. . . . I met people and I just couldn't talk to them, my mouth was so dry (FG1Pt1).

Talking to people when I'm out for a walk is a real problem. . . . Rather than start to try to explain about my dry mouth, it was after a shower and there was water on the side of the road, so I just bent down and had some to try to lubricate my mouth. Then I thought, is that what my life has come to, drinking rainwater so I can just talk? (FG2Pt3).

If you do go out socializing, you hope there is a real talker in the room so you can leave them at it. I almost need to take a time out (FG1Pt4).

You have to trick yourself into doing things; otherwise, you would never go out. Then when you go out, you are mindful of your mouth all the time. I withdraw a little bit more each year that I have this dry mouth (FG2Pt1).

I used to do a little charity work, but I just couldn't do that anymore because I had to talk. I just withdraw from a lot of things (FG2Pt4).

It has a big effect on my mood. I don't go out. I haven't been going out for the last while. I used to like going somewhere and meeting someone for lunch or tea, but I would really think twice now about that. I wouldn't meet someone I didn't know very, very well, so it limits your social circle (FG1Pt2).

It stops you doing things (FG2Pt2).

There was a general sense of concern for the future, especially if the degree of oral dryness would worsen over time:

You just feel like why this is all happening to me. . . . You wake up in the morning and you are just thinking, "Here we go again" (FG1Pt2).

It gets me down big time. You begin to feel like "Why me and when will this end?" (FG1Pt4).

As long as things don't get worse. I'm afraid of that every day. It really concerns me (F2G2Pt1).

For me, looking forward, when I read things online, it all worries me. Where is it going to end? I'm young. Where is it going to end? (FG1Pt3).

Theme 3: Frustration with dry mouth management. The limited number of treatment options to relieve the oral symptoms of dry mouth and aid with oral function emerged as a real source of frustration for participants. Many were resigned to never having relief of symptoms and just needing to find "workarounds" to cope:

There must be something better out there. Now I take a small spray with me as a backup, but it doesn't do me much good (FG1Pt1).

Because there isn't much available to help, all the gels and sprays, I just can't use any of them because they are too sticky (FG2Pt3).

There is no help. Nothing is going to fix this (FG1Pt2).

Nothing works, but you just have to say get on with it. Nothing is going to change, and nobody can help (FG2Pt2).

You are conscious that if you get coughing, you get stressed and then everything gets worse. You even have to be conscious about breathing. I have to breathe more through my nose and keep my mouth shut to try to keep my mouth a little less dry. Everything is an effort, and nothing can help me. Sprays, gels, mouthwashes—they are all useless (FG1Pt4).

Theme 4: Limited knowledge of medical community and lack of understanding of family and friends. Some participants were upset and embarrassed about the lack of understanding or knowledge of those around them regarding their ongoing oral dryness. Others were frustrated that healthcare professionals were not acknowledging the impact that oral dryness was having on their everyday lives:

When you tell people you have something wrong with your saliva glands, they just look at you; it is like you are speaking a different language. Even if you say you have Sjögren syndrome, they still look at you. Even the medical professionals don't understand, and some haven't even heard of it (FG1Pt2).

Yeah, a lot of people just don't understand. It gets embarrassing. You try to explain, but it is pointless so better to keep your mouth shut (FG1Pt4).

It is so much worse when you are with people. What are you meant to say? I suffer from dry mouth, and the majority of people don't know what you are talking about (FG2Pt2).

I see Rheumatology, and some of the young doctors don't know that dry mouth is part of Sjögren, so they don't want to know about how bad my dry mouth is or try to help me with it. . . not their responsibility (FG2Pt1).

You have Sjögren syndrome, they still look at you. Some of the medical professionals don't understand

how bad the dryness is, and some haven't even heard of it (FG2Pt4).

Part 2: Review of Existing Outcomes

A considerable part of both focus groups was dedicated to discussing the social limitations of having a dry mouth and frustration with current treatments. However, the participants were keen to highlight that although attention to social isolation was important, it was more important to focus on practical measures, such as recording how much saliva is available in the mouth, as the use of these measures would help patients with dry mouth anticipate how they may improve with participation in a clinical trial.

Socializing is a secondary thing. . . There is an importance attached to it for all of us, but I would rather they would accurately record how much saliva is actually in your mouth. Once they improve that in research, then the socializing will follow (FG2Pt3).

An additional outcome of importance reported by the participants was the impact of dry mouth on overall health and oral health specifically.

Lots of us have this dry mouth because of another condition, like Sjögren syndrome or after cancer treatment, so why wouldn't you want to record how the person's overall health is in a study for dry mouth? It doesn't make sense otherwise (FG1Pt3).

It has a big effect on the health of my teeth, and because of that it has an effect on my finances. It might not be the same for people all over the world, but visiting the dentist costs lots here (FG1Pt2).

Yeah, what if my teeth start to break and fall out. . . I won't be able to eat or anything. Then it will have an effect on everything. It is really important to record out the health of your teeth and mouth, I think (FG1Pt1).

DISCUSSION

To our knowledge, this was the first study to explore patients' personal experiences with dry mouth, focusing on their perspectives on their treatments and outcomes of care to investigate which outcomes are important to the cohort of patients examined. Despite evidence that salivary gland hypofunction and xerostomia impact nearly all physical, psychological, spiritual, social, financial, and family domains, among other recognized components of quality-of-life outcomes, only

a few clinical studies have explored the perception of patients regarding dry mouth outcomes.^{3,24-28} To fill this gap in scientifically based information, this WONDER study assessed the real-world experiences of patients with dry mouth to investigate which outcomes are most important from a patient-centered perspective, eliciting information for the improvement of the Core Outcome Measures in Effectiveness Trials and ultimately guiding changes that benefit patients.

Most previously published qualitative studies of patients with dry mouth, which focused on oral symptoms and patient coping strategies,²⁹ have demonstrated that dry mouth often impacts quality-of-life outcomes more severely than dental-related disease.³⁰⁻³⁴ Hence, this WONDER study expanded the assessment tools used to cover all the outcomes relevant to dry mouth to provide original data regarding how dry-mouth-related symptoms can escalate in real-world scenarios and become an “aggravating misery” among patients.³⁵

A previous qualitative study²⁹ highlighted the lack of condition-specific measures targeting nonphysical dimensions of oral health-related quality of life outcomes, including negative social impacts. In our study, participants unanimously reported that the social isolation and withdrawal imposed by dry mouth deserve to be fully explored and measured in future clinical trials. Most participants mentioned that dry mouth negatively impacted everyday situations, such as enjoying walks, talking, going out for lunch or tea, and even doing charity work, thereby imposing severe social restrictions. The impact of how “noticeable” a certain condition is to others has been shown to be a more direct measure of the intrusiveness of symptoms among vitiligo patients,³⁶ and was validated among patients with dry mouth in this study. Such validation is important in future trials because it suggests that patients with dry mouths cannot cope with their symptoms privately.

The merits of qualitative data in exploring the perspective of patients with chronic conditions cannot be underestimated. Utilizing patient data to determine the nature, severity, and frequency of oral diseases is the first step in providing treatment.^{37,38} The current understanding of outcomes associated with dry mouth is mostly based on clinician-reported data,²⁸ which explains the discrepancy between provider-based assessments and the patient-reported experiences in this study concerning patients’ frustration with the treatment options available and, most importantly, perceptions of the quality of support they received by medical providers.

The current findings reinforce the need for collecting “real-world” data from patients with dry mouth to provide more opportunities to understand symptom burden and uncover the urgent need to develop personalized

interventions. Because dry mouth may lead to social disability, creating new measures to prevent its multidimensional consequences may improve interventions for patients struggling to manage their suffering without proper support from health care providers. New approaches should include public and private health strategies targeting venues to improve the patient environment. Examples of these strategies include providing private or closed spaces, humidifiers, free drinking water, and encouraging restaurants to consider menu options for patients with dry mouth and digital media managers to raise awareness about the contemporary social impact of dry mouth.²⁹

Although achieving data saturation, the researchers examined only a small group of adult participants from a single center in the Republic of Ireland. Thus, the opinions and impacts reported may not truly represent all patients with dry mouths worldwide. However, once data saturation has been achieved in qualitative research, no more patients need to be recruited. Future studies with more patients performed in multiple centers outside the Republic of Ireland could add to the qualitative data evidence base and enhance our understanding of cultural differences in multicultural populations within and among countries. Regarding limitations, because this study did not objectively quantify dry mouth, only the more severe end of the dry mouth spectrum of symptoms was covered. Furthermore, the conclusions were limited because patients with Sjögren syndrome and patients who have undergone head and neck radiotherapy encompass very different populations. For the latter, quality of life domains are distinctly affected and enhanced by radiotherapy due to multiple adverse effects, such as mucosal sensitivity, recurrent oral infections, dysgeusia, and rampant caries risk, which overlap with reported dry mouth impact.^{33,34}

Once the COS for dry mouth has been finalized, the next development phase will be to determine which instruments or tools should be used to record this core set of outcome domains. Existing instruments employed to measure dry mouth will be rigorously evaluated following a specific methodology to ensure that the most suitable measurement tools are finally selected (<https://www.cosmin.nl/tools/guideline-selecting-proms-cos/>).

CONCLUSIONS

In conclusion, our exploration of the personal experience of patients with dry mouth focusing on outcomes of care identified a degree of hopelessness and despair that the treatment options available have so little impact on their lives. The role of the patient as the main stakeholder in COS development was highlighted with the identification of emerging social outcomes to

be considered in the design of clinical trials. This finding emphasizes the need for developing a patient-centered COS for dry mouth to generate more robust evidence for treatment interventions.

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PRESENTATION

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