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World Workshop on Oral Medicine VIII: What outcomes domains are measured in studies for dry mouth? Part III: A qualitative study on the patient perspective of dry mouth outcomes. A WONDER study.

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Abstract

Objectives: We conducted a qualitative study on patients' perspectives of dry mouth outcomes to explore their lived experiences and investigate what 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 (COMET). **Study Design:** This was a qualitative study based on digitally recorded, semi-structured interviews using a study-specific topic guide incorporating focus groups of patients with dry mouth secondary to Sjögren's syndrome, and head and neck radiotherapy. The interviews were performed until data saturation, all transcripts were evaluated for accuracy and then anonymized. **Results:** Two focus groups consisting of four participants per group identified four distinct themes: (1) impact on oral health and function; (2) social isolation and withdrawal; (3) frustrations 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 patients' 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.

Introduction

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 (e.g. lysozyme, lipase, and amylase), electrolytes, and secretory immunoglobulin A, and after being secreted into the oral cavity, saliva also contains crevicular fluid and a pool of human cells (epithelial cells and inflammatory cells). Saliva protects the dentition from decay and provide lubrication of the oral tissues, allowing proper speech, chewing and swallowing, among other functions. The enzymes initiate the digesting process in the oral cavity (Dodds et al., 2015). Due to the fundamental role of saliva in maintaining oral health, the impact of dry mouth on patients' daily lives cannot be underestimated (Ship 2002).

Subjective complaints of dry mouth, in professional and diagnostic terminology also known as 'xerostomia', correspond to the symptom of oral dryness. Xerostomia is not necessarily accompanied by a quantitative reduction in salivary flow rate. On the other hand, the diagnoses of salivary gland hypofunction or hyposalivation refer to a quantified decrease in the salivary volume. The underlying causes of salivary gland hypofunction and xerostomia include medication intake, Sjögren's syndrome, also known as Sjögren's disease, radiation therapy to the head and neck, senility, and dehydration (Wolff et al., 2017).

Although dry mouth is a common condition globally, management options that provide sustained relief of symptoms are limited. Multiple clinical trials have been carried out in this setting; however, systematic reviews have highlighted limited evidence to recommend any sufficiently robust treatment modality. Furthermore, the comparison of the efficacy of treatment modalities or pooling of results of clinical trials has been inconsistent due to a lack of consensus

on what outcomes should be recorded in clinical trials for dry mouth (Furness et al., 2011; Riley et al., 2017).

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 (COMET) initiative, the generation of a comprehensive list of outcomes identifying and rectifying deficiencies in existing knowledge should be carried out when designing clinical trials (Sinha et al., 2008; Sinha et al., 2012; Ní Ríordáin et al., 2015; Williamson et al., 2017; <https://www.comet-initiative.org/>). Qualitative interviews with patients experiencing the conditions to be explored are also pivotal to enabling the identification of outcomes that are important to them, allowing the patient voice to be incorporated into the COS. This is now seen as an integral part of the development of a COS (Kirkham et al., 2017), and has been adopted by outcome sets developed in allied medical specialities, such as neurology, rheumatology, and respiratory medicine (Kirwan et al., 2003; Sinha et al., 2012; Noble, Marson 2016).

Young and Bagley highlighted the importance and benefits of patient involvement in COS design, and in determining which outcomes should be included in a minimum outcome set (Young, Bagley 2016). The authors also discussed the various methodologies used and the fact that the methods used to generate clinician or researcher opinions may not be as fruitful without patient's input. They emphasized the merits of using qualitative research methods to add to the material generated from the initial systematic review 'longlist' 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 lived experience of patients with dry mouth, with

an emphasis on the treatments used and the outcomes of care, and investigate what outcomes were important to this cohort of patients.

Materials and Methods

This was a qualitative study based on recorded semi-structured interviews using a study-specific topic guide and incorporating focus groups of participants with dry mouth. The reporting of the study follows guidance from the Standard for Reporting Qualitative Research (SRQR) (O'Brien et al., 2014). Each focus group was held in two parts: part 1 explored the lived experience of patients with dry mouth, and part 2 allowed patients to review a summary of the outcomes used in the dry mouth literature. A discussion ensued regarding the importance, from the patient perspective, of the various outcomes found in the literature and which outcome domains of importance to patients were absent from the list presented.

Ethical considerations

This study received ethical approval from the Clinical Research Ethics Committee of Cork Teaching Hospitals, Ireland [ECM 3 (rrr) 01/06/2021]. Participants were provided with the written study specific information leaflet and signed an informed consent form.

The research team and reflexivity

The research team consisted of two researchers [RNiR (Researcher 1) and MO'C (Researcher 2)]. Researcher 1 was responsible for running the focus groups whilst Researcher 2 was responsible for the recruitment of participants. Data analysis was shared between both

researchers. A debriefing meeting was held after each focus group to encourage reflexivity (Dodgson et al., 2019).

Participants and sampling strategy

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 if they were willing to participate in the study. Only English speakers were considered for the study. There were no other inclusion or exclusion criteria.

The sampling strategy was purposive to include participants who had dry mouth of varying etiologies (medication-induced, Sjögren's syndrome; head and neck radiotherapy), and severity. Time since diagnosis was also considered when recruiting participants, with some recently diagnosed and other longstanding patients of the host Oral Medicine Unit.

A total of 20 patients were contacted by telephone between March and April 2022. Two focus groups with a total number of eight participants met face-to-face. The remaining 12 patients (including four patients with medication-induced xerostomia, five patients with xerostomia post head and neck radiotherapy, and three patients with Sjögren's syndrome) declined the invitation due to the following reasons - two patients with medication-induced xerostomia became unwell the day of the focus group, others had prior family or work commitments. Each focus group lasted between 40-50 minutes. Data analysis was an iterative process and collection ceased once saturation was achieved. Data saturation was defined as the moment at which the data collected had enough breadth and depth, was adequate to answer research questions, and no new themes were emerging (O'Reilly, Parkeret, 2012).

Data collection and analysis

A structured topic guide was developed by the research team, with minor edits following feedback from participants in the initial focus group (**Table 1**). The topic guide allowed a similar scope of areas to be covered in each focus group, however, the facilitator allowed conversations to flow between participants with minimal intervention (Braun, Clarke, 2013). The focus group methodology used allowed group interactions, ensuring participants had the opportunity to share lived experiences and collectively explore outcomes of the treatment of dry mouth that were of significance to the group (Bryman, 2016). The focus group discussions were audio recorded and transcribed *verbatim*.

An interpretive methodology was utilized to identify themes of the lived experience of the patients, keeping in mind the conceptual framework of Locker's Model of Oral Health (Locker 1988; Nowell et al. 2017). The transcripts were analyzed on a line-by-line basis and coded, to define the data to be analyzed. This data analysis was done manually using Microsoft Office Word software® (Microsoft Corporation, Redmond, WA, USA). Coding involved identifying passages of text that exemplify a descriptive idea and linking them together under a single idea or code. This process then allowed the transcripts to be categorized and facilitated the identification of key themes. Selected anonymized quotes were taken from the transcripts, per key themes, and presented as an emblematic part of the qualitative results.

Results

Four different themes (**Figure 1**) were covered in two focus groups with four participants per focus group (**Table 2**), totaling eight face-to-face participants. The study recruited more women (six women vs two men) with no apparent differences when compared to men with self-

reported dry mouth experiences. Of these, five patients had xerostomia because of Sjögren's syndrome, and three patients had xerostomia following head and neck radiation therapy. Patients were aged 45 to 77 years and lived across different parts of Ireland. Demographic and clinicopathologic data of the participants are summarized in **Table 3**.

Dry mouth outcomes had a deep multidimensional impact on participants, including the severity and impact on everyday life. After categorization, selected anonymized quotes were taken *ipsis litteris* from the transcripts exploring each of the study themes and are presented below, followed by patient's respective study code.

Part 1: Lived experience with an emphasis on the treatment used and the outcomes of care

Theme 1: Impact on oral health and function

Discussions in both focus groups concentrated on their participant's limited ability to speak, eat, taste and swallow:

...saliva is so important. I realise 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, Patient 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.

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 bit 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 discoloured, prone to cavities, my teeth are chipping. I’m constantly going to the dentist – there is a huge financial aspect to it. It big times 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 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 has become and how gradually over time they 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 socialising, 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: Frustrations with dry mouth management

The limited number of treatment options to relieve the oral symptoms 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 cope for find 'work-arounds':

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

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 the impact of the oral dryness on their everyday lives was not being acknowledged by healthcare professionals:

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's syndrome, they still look at you. Even the medical professionals don't understand, and some haven't even heard of it... – FG1Pt2.

Ya, 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's 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's 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 the existing outcomes used

A considerable part of both focus groups was dedicated to a discussion about the social limitations of having a dry mouth and the frustrations with current treatments. However, participants were keen to highlight that although the social isolation was important, it would be more important to record practical measures such as how much saliva is available in the mouth as this is what you would hope would really improve in a clinical trial.

Socialising 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 socialising will follow - FG2Pt3.

An additional outcome of importance, according to the patients, were 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's 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.

Ya, 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

This is the first study to explore the lived experience of patients with dry mouth, with an emphasis on the treatments used and the outcomes of care, and investigate what outcomes were important to this cohort of patients.

Although there's previous evidence about the fact 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 on dry mouth outcomes (Cioffi 1991; Aliko et al., 2015; Villa et al., 2015; Villa et al., 2016; Wolff et al., 2017; Mercadante et al., 2021). To fill this gap of scientifically based information, this WONDER study assessed real-world experiences lived by patients with dry mouth to investigate what outcomes are most important from a patient-centered perspective, achieving information for the improvement of COMET and ultimately guiding changes that benefit patients.

Most of the previously published qualitative studies on patients with dry mouth have focused on oral symptoms, and patient's strategies to cope with it (Gibson et al. 2020), successfully demonstrating that dry mouth often impacts the quality-of-life outcomes more severely than dental-related disease (Locker et al., 2003; Rohr et al., 2010; Owens et al., 2014; Gouvêa et al., 2020; de Pauli Paglioni et al., 2020). Hence, this WONDER study has expanded assessment tools to cover all the outcomes that cluster from dry mouth to provide original data about how dry-mouth-related symptoms can escalate in real-world scenarios and become an “aggravating misery” among participants (Folke et al., 2009).

When assessing previous qualitative studies, Gibson et al. 2020 highlighted the lack of condition-specific measures targeting non-physical dimensions of oral health-related quality of life outcomes, including restrictions on social impacts. In our study, social isolation and withdrawal imposed by dry mouth was an unanimous perception among the participants that deserve to be fully explored and measured in future clinical trials. Most of the participants mentioned that dry mouth negatively impacted everyday life situations, such as walking, talking, going out for lunch or tea, and even charity work, imposing severe social restrictions. The impact of the “noticeability” of a certain condition has been previously shown as a more direct measure of the intrusiveness of symptoms among vitiligo patients (Batchelor et al., 2016) and was

validated herein among patients with dry mouth. This will be important in future trials because it suggests that patients with dry mouth may not be able to 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 making treatment advancements (Atkinson et al., 2012; van den Hurk et al., 2022). The current understanding of outcomes associated with dry mouth is mostly based on clinician-reported data (Gibson et al. 2020) which explains the discrepancy between provider-based assessments and the experiences reported by the participants of this study concerning their frustrations with treatment options available and most importantly, their perceptions about the quality of support received by medical providers.

The current findings reinforce the need for “real-world” data from patients with dry mouth providing an additional chance to understand the symptoms’ burden, and possibly uncover the urgent need to develop personalized interventions. Since 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 in this sense should include strategies at the public and private health levels targeting venues to improve their atmospheres (with private or closed spaces, humidifiers, and/or free drinking water), encourage 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 (Gibson et al. 2020).

Although data saturation was achieved, this was a relatively small group of participants from a single site, the opinions and impacts represented in the study may not be truly representative of all patients with dry mouth worldwide. Future studies, out with the Republic of Ireland, could

add to the current qualitative data and enhance our understanding of cultural differences in multicultural populations (Asia, Africa, North America, South America, Antarctica, other European regions, the United Kingdom, and Australia, among others) affected by other dry mouthing conditions, such as senile and medication-induced dry mouth. In terms of limitations, it is also important mentioning that intimacy was not broached specifically during the focus groups (just the topic of relationships with family); and that dry mouth severity was not objectively quantified, so only the more severe end of the dry mouth spectrum of symptoms was covered because the recruitment happened from secondary care patients.

In conclusion, we explored the lived experience of patients with dry mouth, emphasizing the outcomes of care, and found a degree of hopelessness, despair with treatment options available and how current treatments have so little impact on their lives. This further emphasizes the need for the development of a patient-centered COS for dry mouth, to generate more robust evidence for treatment interventions. The role of the patient as the main stakeholder in COS development has also been highlighted in the present study with the identification of emerging social outcomes to be considered in the design of clinical trials.

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

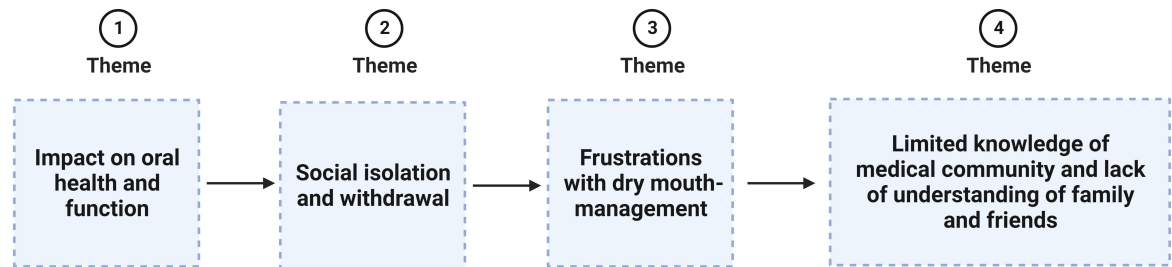


Table 1. Structured topic guide used for data collection.

Stages	Approaches
<i>Personal history and knowledge about dry mouth</i>	<p><i>What do you understand about you what has caused your dry mouth?</i></p> <p><i>Number of years/months living with dry mouth</i></p>
<i>Experience of living with dry mouth (how has dry mouth impacted on your life)</i>	<p><i>Quality of life – activity limitations – Have you had problems while eating, drinking, speaking?</i></p> <p><i>Mood: have you stopped doing things due to the emotional burden of dry mouth?</i></p> <p><i>Have you worried? Do you think that your mood has any influence on your pain?</i></p> <p><i>Family and social interactions – has anything changed with regards to social interactions? Have you stopped making plans with your friends or family? Have you stopped going out for meals?</i></p> <p><i>Productivity – Have you had to take time off work?</i></p>
<i>Treatment of dry mouth</i>	<i>What treatments have you had?</i>

Satisfaction with treatment

Was it successful?

What do you understand about success of treatment?

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?

Treatment outcomes

If you had to think about a list of the most important treatment outcomes, what would they be?

Open questions

Table 2. Outline of the 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: Frustrations with dry mouth management (Disability – psychological)	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 healthcare professional of the impact of dry mouth Social embarrassment associated with poor understanding when trying to explain oral dryness

Table 3. Demographic and clinicopathologic characteristics of study participants.

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