Ill by mouth? Patients' experiences of the oral and dental manifestations of scleroderma.

Abstract

Scleroderma is a rare chronic multi-system disease characterised by fibrotic changes in the skin, connective tissues and internal organs. The high mortality rate associated with the condition means that clinical attention is often focused exclusively in these contexts. This paper reports on a mixed methods study which explored patients' and practitioners' experiences of the oral and dental manifestations of scleroderma – other aspects of the condition that, patients state, also have life-changing, and limiting, effects. The overarching research questions underpinning the study were, what are patients' and practitioners' experiences of scleroderma-related dental problems and what strategies can be developed to improve patients' quality of life?

Here we focus exclusively on patients, who reported a significant disjuncture between their own experiences and professionals' understanding and intervention, such that their scleroderma related oral and dental problems were perceived to be clinically ignored and/or trivialised. This evident mismatch generates a range of keenly felt absences wherein the patient, the mouth, and the oral cavity more broadly, can be seen to both literally, and metaphorically, dis/dysappear. We draw on a range of theoretical and empirical perspectives to recognise the complex interplay of priorities and expectations that play out between patient and practitioner, concluding that far greater clinical attention should be afforded to the patient's own account of their condition whether or not this fully aligns with practitioners' perspectives.

Scleroderma affects in excess of 2.5 million people globally, but the issues we explore here have a resonance far beyond this one rare condition. A range of other auto-immune and chronic conditions, such as type 1 diabetes and rheumatoid arthritis, also prominently feature oral and dental problems and, as such, this work has the potential to help shape the treatment and understanding of the illness experiences of significant numbers of people.

Key Words

Scleroderma, mouth, dental, dys/disappearance, quality-of-life

Introduction

As key features of human appearance, communication, and oral experience, the mouth, and its attendant teeth, play critically important parts in our lives. Thus, whilst oral and dental health are essential for good physical health, they are also intrinsic to a person's sense of self and psychological well-being – such that Rousseau *et al*., (2013:463) termed the mouth "symbolic of the self". Further, the place occupied by the mouth, in the centre of the face and, ordinarily, the hub around which communication is centred, is both highly visible and a key site of intrinsic, and extrinsic, judgements made for, and about, an individual (Exley, 2009; Fiske *et al*., 1998; Steele, *et al*., 2000). The mouth, in this context, is, therefore, fundamentally 'socially symbolic' (Khalid and Quiñonez, 2015) in that good oral health is a key identifier, not just of an evident commitment to dental hygiene and oral health care, but also of a person's social class, social standing and, in a dental industry that relies, increasingly, on private practice, access to conspicuous disposable income.

Whilst there is a plethora of work which focuses on the clinical contexts of dental disorders, there has been relatively little research that has focused on the "mouth as an embodied reality in everyday life" (Gibson and Exley, nd: 53). Similarly, there has been scant attention paid to the ways in which people understand and experience what happens when the mouth 'fails' and requires professional intervention. Further, the sociology of the mouth, and of dentistry in particular, remains limited (Exley, 2009; Graham, 2006; Neville, 2023) when compared to the sociology of health and illness, within which it is located, more broadly (Exley, 2009).

There are, however, a number of notable exceptions, including the work of Fiske *et al*., (1998) and Davis, *et al*., (2000) who focused on the emotional and practical effects of tooth loss and Graham *et al*., (2006) who explored how partial dentures are employed in the context of regaining a sense of dental normality following tooth loss. Durham, *et al*.'s (2010) later qualitative study explored the experiences of people living with temporomandibular disorders, and Peters, et al., (2015) focused on the experience of living with chronic pain, whilst Rousseau, *et al*., (2014) employed the concept of biographical disruption (Bury, 1982) as a theoretical tool to explore the experience and meaning of dental pain, tooth loss and replacement.

More recent work has begun to explore the oral health experiences of people living with long-term illness and disability. The work of Bogner, *et al*., (2024) and Bogner and Faulks, (2023), for example, explored the experiences of people with intellectual disabilities and complex medical conditions, whose oral and dental health problems added further dimensions of complexity and potential burden to already challenging health and social circumstances. Likewise for people living with a diagnosis of dementia, whose oral health can be compromised by a systematic neglect of the mouth which is underpinned by a lack of social, economic, and cultural capital (Scambler, *et al*., 2023, Daly, *et al*., 2017).

There is also a developing awareness of the complex relationships between dental health and long-term illness but this, to date, has tended to focus most explicitly on the clinical manifestations of these associations (see, for example, Fu, *et al*., 2025, Kane, 2017).This relatively small literature has highlighted what is a critical lack of attention to the sociology of dentistry whilst, most importantly, generating an evolving dialogue at the critical intersection between medicine and dentistry – the mouth and oral cavity. The imperative for this dialogue is increasingly important in the context of the rapid increase of people living with multiple long-term conditions globally and it is at this particular intersection that we situate this paper.

The paper reports on a collaborative, mixed-methods study, funded by Scleroderma & Reynaud's UK (SRUK) (Grant Call: Quality of Life), that extends this area, exploring patients' understandings and experiences of the oral and dental manifestations of scleroderma (ODMS), a rare, chronic, multisystem autoimmune condition that primarily affects the internal organs, skin and connective tissues. The study aimed to explore and understand patients' experiences of scleroderma-related dental problems and to document rheumatologists' and dental professionals' awareness of the condition and its oral and dental manifestations. We also worked with patients and practitioners to co-produce resources to help optimise referral mechanisms and improve preventive oral and dental care in scleroderma. The overarching research questions underpinning the study were, what are patients' and practitioners' experiences of scleroderma-related dental problems and what strategies can be developed to improve patients' quality of life?

This paper, which focuses primarily on patients' experiences, presents a range of original theoretical insights whilst also presenting new empirical material which promises to help

shape the future treatment of oral health care in scleroderma. Moreover, we suggest that a focus on the specificities of scleroderma allows for a wide-ranging analysis of clinical policy and practice which transcend the particularities of this rare condition, contributing to the wider literature on rare autoimmune diseases.

Scleroderma

Scleroderma is an auto-immune condition characterised by the production of excess collagen leading to fibrosis of the skin and internal organs causing thickening and hardening. In this context, the immune system, often perceived as entirely benign and focused only on actively engaging with 'foreign' invaders (viruses, bacteria, parasites and the like), begins to perceive normal tissue as pathogenic. Felstiner (2007:8), in the context of another autoimmune condition, rheumatoid arthritis, articulates this process thus – the immune system "keeps treating body parts as aliens, keeps arousing immune cells until they are attacking familiars as suspects, picking on bones they are supposed to be keeping safe". As such, in autoimmune disease, the body effectively produces an immune response to itself.

There is a vast array of autoimmune conditions, with up to 10% of the (UK) adult population affected at any one time (Conrad *et al*., 2023). They include commonly known conditions such as rheumatoid arthritis, multiple sclerosis, and type I diabetes and less common diseases such as lupus, Sjogren's syndrome and scleroderma itself. What unites these conditions, however, is that they cause chronic long-term morbidity (and in the case of lupus and scleroderma, in particular, early mortality), and significant incapacity in a range of contexts.

One of the defining features of autoimmune conditions is their symptomatic flux and how their impacts shift in and out of focus as symptoms wax and wane, sometimes on a daily basis. Diagnosis is often a protracted process (lupus, for example, takes an average of seven years to diagnose) and these conditions are characterised by insistent clinical and existential uncertainty and a pervasive range of biological, psychological, and social effects. Taken together, autoimmune conditions generate profoundly enigmatic illness experiences for both patients and practitioners (Price and Walker, 2015). As such, scleroderma, like other autoimmune conditions, constitutes the site of significant embodied and biographical

disruption (Bury, 1982), primarily impacting upon those (usually women) in their most productive years (Ngo, *et al*., 2014; Dou, *et al*., 2024) and having significant negative effects on quality-of-life.

There are 2.5 million people worldwide who are diagnosed with scleroderma, including, approximately, 19,000 in the UK. Given that cumulative survival from diagnosis has been estimated at 89.3% at 3 years, 74.9% at 5 years and 62.5% at 10 years (Elhai, *et al*., 2017; Rubio-Rivas *et al*., 2014), it is perhaps not surprising that, at the point of diagnosis, clinicians tend to focus on the significant threats scleroderma poses to the internal organs. Much time, clinical concern and effort, is spent, particularly early in the disease, testing for, and treating, internal organ involvement and damage and the variety of ways in which scleroderma impacts upon the skin and connective tissues more generally.

There are, however, other significant aspects of this condition that tend to be poorly understood or addressed by clinicians, but which also have a profoundly negative impact on people's quality of life. In the context of this paper, these include ODMS which may be experienced by up to 80% of people living with scleroderma (Authors' own, 2023). These can include microstomia (limited mouth opening), muscular atrophy, thin lips, xerostomia (dry mouth), oral fibrosis, ulcers, restricted tongue mobility, temporomandibular joint problems, and salivary gland disorders. In addition, people may also experience problems maintaining their oral health due to physical limitations they experience as a result of scleroderma more broadly, such as contractures in the hands.

These issues, unsurprisingly, are accompanied by acute assaults on a person's sense of self and identity and, as our data will suggest, ODMS can be very much the focus of patients' concerns. This fact is reflected and validated through a recent campaign mounted by SRUK - 'scleroderma won't take my smile', referencing the very real experiences people living with scleroderma have in the context of a 'disappearing' mouth, as their lips recede, and the oral cavity shrinks and hardens.

As noted above, the concerns people have in this context, however, tend not to be equally shared, recognised, or validated by the clinicians involved in their care (both dentists and rheumatologists - the clinical specialism most closely associated with autoimmune conditions such as scleroderma). This can result in a profound sense of invalidation for

patients and missed opportunities for clinicians to holistically address the effects of this systemic disease.

In the following sections of this paper, we explore patients' perspectives on, and experiences of, what has, historically, been a contested space in the context of clinical policy and practice – the mouth. Our analysis centres, in particular, around the concept of dis(and dys)appearance. In so doing, we address the varied metaphorical, and very real, absences generated when clinicians and patients approach the problems associated with conditions such as scleroderma in markedly different ways. We will argue that these issues have been shaped by a range of historical, policy-driven and professional prerogatives, all of which culminate in a variety of negative effects on patients' quality of life.

Methods

The study utilised a concurrent mixed methods study design comprised of four elements:

1. A systematic scoping review of the literature (Authors' own, 2023)
2. Three online surveys with (i) patients (ii) rheumatologists (iii) primary care dental professionals (Authors' own, 2024)
3. In-depth qualitative interviews with people living with scleroderma
4. Co-production of project outputs – the project team have worked with a PPI (Patient and Public Involvement) group based at the University of XX to collaboratively explore awareness-raising, information tools and referral mechanisms to optimise the patient care pathway in scleroderma. Participants were recruited through SRUK's social media and an existing study of Scleroderma at X University led by XX. Ethical approval was granted by the Faculty of Health Sciences, University of Hull and NHS HRA 15/NE/0211

Rheumatologists were recruited through a call for participants in the UK Scleroderma Study Group (UKSSG). Dentists were recruited from the British Dental Association (BDA) and the College of Dentistry (CGDent), and the call for participants was endorsed by the Oral Health Foundation. Surveys were anonymous and hosted on Jisc. All participants were provided

with an information sheet outlining the purpose of the study, what the research involved, considerations of data protection and confidentiality, and contact details.

One hundred and fifty-eight people living with a confirmed diagnosis of scleroderma completed the online survey and 13 telephone interviews were undertaken by XX. The interviews explored patients' experiences of their conditions and their interactions with professionals. All interviews were recorded, transcribed verbatim, and appropriately anonymised.

Qualitative data analysis was undertaken using thematic analysis (Braun & Clarke, 2006). This involved an inductive and flexible coding process. The resulting themes were reviewed by different team members to ensure analytic rigour. Data was synthesised by combining qualitative and quantitative data - the number of participants who contributed to the themes and codes from the analysis of free-text survey responses was noted and used as a descriptive quantitative measure (e.g. number of patients who experience oral pain) to enhance the qualitative analysis (e.g. the lived experience and impact of oral pain). An analytic/reflexive journal was used throughout data analysis and triangulation to note convergence and divergence of findings and to reflect on researcher positionality and emergent themes. A narrative synthesis of qualitative data further incorporated descriptive quantitative data – each data source was analysed individually, and triangulation occurred when the analysis of each data source was complete. The research team presented initial findings to a PPI group, and over multiple sessions, together agreed upon the format, content, and method of dissemination of the co-produced informative materials.

Respondents in this study reflected an expected autoimmune/ scleroderma profile – overwhelmingly female (96.8% - n = 153), middle-aged (61-70 years; n = 53 or 51-60 years; n = 50) and White British (87.3%; n = 137). All four countries in the UK were well represented in this group of respondents, many of whom have been living with scleroderma for a significant number of years.

As we report the data here, respondents who undertook the online survey are identified by a participant number. Interview participants are detailed as such.

The Mouth in Scleroderma

The clinical literature on ODMS demonstrates a developing understanding of the identification and management of the oral and dental problems associated with the scleroderma, the barriers associated with good oral and dental care and pointers toward best practice for professionals working with people living with the condition (Beaty, *et al*., 2021, Leader *et al*., 2014; Puzio *et al*., 2019). A recent scoping review (Authors' own, 2023) was the first paper to bring together this diverse literature focusing on how ODMS are experienced by patients. The review concluded by underlining the importance of multidisciplinary care, streamlining procedures between the disciplines of dentistry and rheumatology and, not least, the necessity of centralising the patient, and their experience, in both the process of diagnosis and ongoing treatment. It is this aspect of the condition which the scoping review identified as a significant gap in the existing literature (Authors' own, 2024). The study on which this paper is based sought to address this lacunae, purposefully foregrounding patients' experiences.

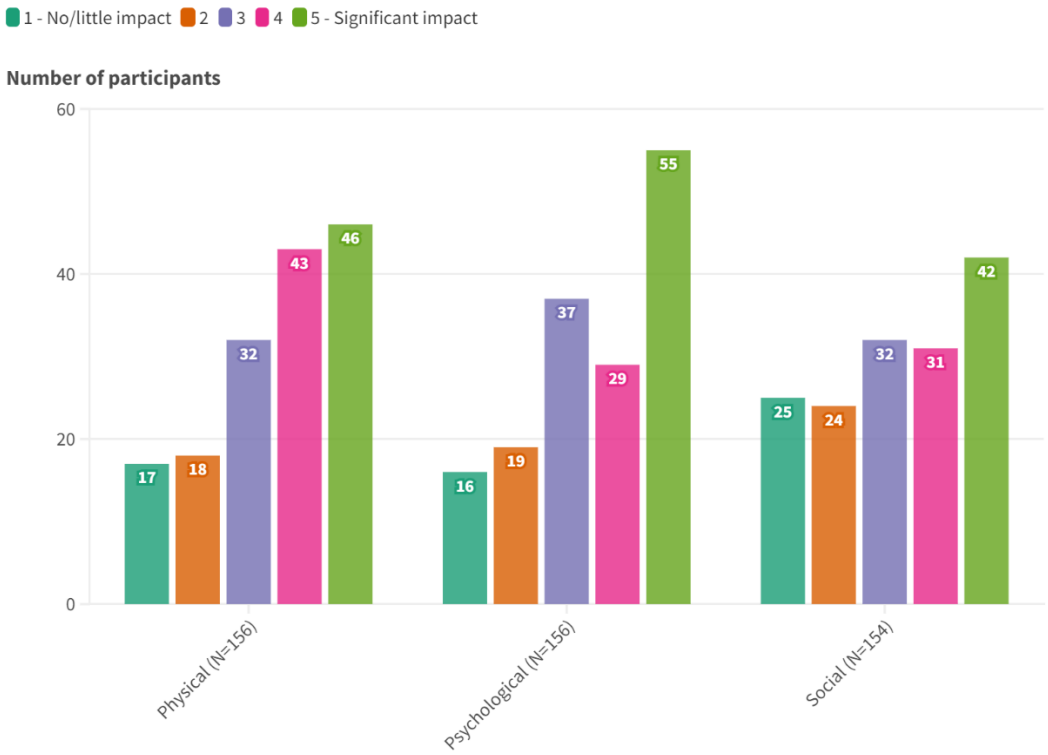
Data analysis generated a number of themes - the varied impacts of ODMS on quality of life, patients' experiences of practitioners' contrasting priorities and, finally, we focus on the varied ways, both metaphorical and very real, that that mouth can be seen to recede from view in the context of a life lived with scleroderma.

Quality of Life

Analysis of the online survey and interviews with patients suggested that whilst ODMS are poorly documented in the literature and ineffectively addressed in clinical contexts, they, nonetheless, create significant challenges for people with scleroderma. A total of 95% of participants in our survey reported problems that could be related to the symptoms of scleroderma with their mouth and/or teeth, which were identified across three related criteria – the physical, psychological, and social effects of scleroderma.

The problems respondents identified were captured in Likert scales detailing the degree to which people felt their lives were impacted where 1=very little impact and 5=significant

impact. Ratings of 4 and 5 across all domains were, as the graph details, noteworthy (Figure



1).

Figure 1: Grouped bar charts showing the physical, psychological, and social impacts of ODMS according to survey participants (Authors' own, 2024).

The physical, psychological, and social impact of ODMS

Participants also provided qualitative responses. Physical impacts included having a very sensitive or painful mouth, fatigue, restricted range of food they were able to eat, nutritional deficiencies from restricted diet and digestive symptoms of scleroderma, restricted mouth opening and changing facial appearance.

> *I can only use a small teaspoon or a baby fork to place food in my mouth. I can only put tiny pieces in. The food has to be dissolvable because I cannot position it with my tongue and I choke. Food gets stuck at side or back of mouth and I cannot move it I have to rinse it out. I mostly live on altraplen shakes. My swallowing muscles into my throat are weak and food goes into my nose and sometimes my lungs* (P061).

> *I know I need a mouth that's big enough to breathe through (my nostrils have already mostly closed due to scleroderma fibrosis) and mouth/teeth to eat with, so I worry about being able to breathe and eat* (P007).

Psychological impacts included feeling shame about the appearance of the face or mouth, depression, anxiety, worries about the future and rumination on symptoms, struggles sleeping, and feeling self-conscious when socialising, eating, or speaking.

> *I dribble and sometimes spit when I speak, I'm conscious of how I look when I smile and eat... it takes me a lot longer to eat. Pain associated with my teeth can occur when I'm out in a room with air conditioning. Or travelling in car... I'm consistently reminded everyday due to the problems that I have this horrible condition and that's depressing in itself* (P092).

> *My confidence is extremely low, since having teeth extracted, especially one of my top front, as I try not to smile or laugh now and try to bow my head or cover my mouth when I talk. My smile used to be something I was proud of, as my teeth have always been white, and something I took pride in looking after. I try not to socialise at the moment because of my lack of confidence due to teeth loss* (P105).

Social impacts included social isolation and withdrawal from social life, deep feelings of shame, discrimination, and stigma, avoiding smiling and laughing, being unable to share meals or attend events with other people, financial burden from frequent dental visits and purchasing adaptive/specialist dental equipment.

> *I don't smile. I don't like to talk to people. I avoid social situations* (P025).

> *I feel embarrassed that I choke. I feel upset I cannot sit with family and friends to eat and drink. I am frightened that I will not survive scleroderma because of my mouth* (P065).

> *I'm extremely self-conscious of my dry lips and broken teeth. I shy away from going out and having to talk to people. I have lost so much confidence that wearing a face mask (during the Covid pandemic) is actually a relief* (P072).

A number of respondents also noted the impacts ODMS had on their working and professional lives.

> *It prevents me from presenting at work, I now find myself backing away from having to speak at work... I don't care as much about how I look as I feel like a freak and there's no point getting ready like I used to...*(P092).

Participants in this study clearly articulate a sense of embodied distress in the context of ODMS which are keenly felt and experienced and which profoundly intrude on all areas of life, limiting personal, professional, social, and economic opportunities.

Contrasting priorities

Despite the evident significance of the problems presented by ODMS, many respondents noted that the mouth is afforded scant attention by clinicians. Eighty seven percent of people in the survey stated that none of the practitioners involved in their care had mentioned them or the challenges they might pose. Further, the majority of people who had been told, by clinicians, about these issues noted that their problems tended to be clinically neglected. Respondents noted that dentists knew little of scleroderma and that rheumatologists failed to engage with the problems associated with ODMS.

> *My rheumatologist has never mentioned mouth problems at all!* (P058).
>
> *My local rheumatologist didn't seem aware of it!! Don't think doctors always understand the psychological problems this causes. Never get asked* (P063).
>
> *I find it very annoying that when I tell my dentist…different members of the same practice…they say they know a bit about it, but I feel they don't know much at all and certainly not enough to be more considerate* (P076).
>
> *I told them. They had no idea what it was/is. I had to spell it for them* (P045).
>
> *No one has mentioned mouth/ dental issues in context with scleroderma. However, I know it's a thing from my own research!* (P038).

Respondents felt their concerns in this area were both trivialised and pathologised.

> *I think I would have benefited from less being blamed on phantom issues and better x-rays, less blamed on dryness and more acknowledgement of the impact that the horrible taste has on my mood …too much focus has been on joints and too much blamed on psychological issues - when it turned out I had simmering abscesses and lip dysplasia* (P107).

There are a number of entirely pragmatic reasons for this reported clinical inattention, the first of which relates to the fact that scleroderma, as previously stated, is a rare condition

and, as such, a dentist, or indeed a rheumatologist, may see very few, or no, people with the condition throughout the course of their career. This would explain a lack of knowledge about scleroderma, but not, of course, the lack of validation, understanding and appropriate clinical response.

This rarity is likely to also account for the fact that, when surveyed as to their knowledge and confidence in understanding, and treating, ODMS, 46% of rheumatologists, and the same percentage of dentists, stated they were uncertain or very uncertain (Authors' own, 2024). No practitioners from either group stated that they were fully confident in managing ODMS.

A further reason for reported clinical inattention, particularly at the point of diagnosis, is that rheumatologists are most concerned to assess, and address, the significant threats scleroderma poses to the internal organs and, potentially, to the life of the person diagnosed. As we have already noted, in scleroderma, the immune system is overactive, a situation which causes the body to attack its own healthy tissues. When the immune system is overactive in this way the resulting inflammation can cause damage to the body and, in scleroderma, continuous and excessive inflammation results in the development of scarring or fibrosis which can lead to hardening of previously healthy tissue.

All areas of the body are at risk from this process, particularly the heart, the lungs, kidneys and GI tract. This scarring and fibrosis can have a profound effect on people's quality of life and is a significant marker of mortality in scleroderma. Unsurprisingly, therefore, clinicians are most concerned to address the impact of the condition in these contexts, particularly in early and diffuse disease (at which point these issues are likely to have the most telling impact).

There are, however, many people with scleroderma who experience less aggressive disease but who live for many years with the significant, troubling, impacts of their condition, some of which are focused, in and around the mouth. Whilst they may not warrant the urgent clinical attention associated with rapidly progressing diffuse disease, they, nonetheless, require a sustained clinical response throughout the lifetime of the condition (for the rest of a person's life). This leads to a stark juxtaposition, where clinicians are focused on clinical

imperatives in which the mouth is, at best, secondary and patients are focused on quality of life, where the mouth assumes a much more central importance.

Professional Disappearance and the Contested Mouth

We would argue, however, that some of the principal reasons for the apparent clinical inattention experienced by respondents in this study extend beyond the mouth to the politics and structures of both dentistry and medicine. In this section, therefore, we focus, on the contested histories and politics of dentistry and medicine which have resulted in shifting professional boundaries and responsibilities such that the mouth has come to assume a uniquely contested embodied space.

It was Nettleton (1988) who was, perhaps, the first to offer an analysis, of how the professionalisation and development of the dental profession early in the 20th century came to establish dentistry as a profession in its own right and, indeed, the mouth as an object of clinical concern quite separate from that which is focused on the rest of the body.

Before this time, dental problems had been managed by 'barber-surgeons', the medical and grooming experts of the Renaissance who administered a range of bodily interventions, which, in addition to shaving and haircutting, included minor surgical procedures such as the lancing of boils, and basic dental interventions, such as 'teeth pulling'. Nettleton (1988) notes that these practitioners became increasingly visible in a bid to have their expertise recognised and, in 1921, their demands were met and dentistry assumed a professional status that throughout the intervening years, has ensured that dentistry came to be perceived as an entirely separate profession from that of medicine. In the context of this paper, this, Nettleton (1988) notes, was the point at which the mouth effectively became separated from the rest of the body; a significant rupture which is emblematic of a significant professional 'disappearance' wherein the mouth and teeth have shrunk from the purview of medicine and, instead, are posited firmly within an incipient dental profession.

Our respondents' narratives would suggest that, in a determinedly 21st Century context, scleroderma offers a clear and clinically relevant example of this historical fracture. That is, that general dentistry, at least, has retained its uncompromising specialist focus, rarely shifting in terms of clinical inquiry beyond the mouth, so tends not to consider the oral and dental impacts of systemic conditions (see, also, Song, *et al*., 2013). Similarly,

rheumatologists, in the context of scleroderma, intent on maintaining an insistent focus on the internal organs and skin, tend not to stray into the historically fiercely guarded area of oral and dental care. As such, the 'ill' mouth, as exemplified in scleroderma, can itself be seen to have disappeared, in a range of contexts, from both dental and medical authority and control.

It can be argued, of course, that dentists are generalists and, given the rarity of scleroderma, may be very unlikely to come across someone living with the condition. By contrast, Special Care Dentistry (in the UK) does work specifically with people who have a range of needs over and above those that can be met by general dental practitioners. Indeed, the NHS Clinical Standards for Special Care Dentistry (2022) specifically refer to patients whose dental treatment may be severely restricted by major positioning difficulties, an inability to open the mouth or dysphagia problems (all of which may relate to people living with scleroderma). In practice, however, special care dental services are scarce (the most recent General Dental Council Registration Statistical Report (2024) states that there are only 263 special care dentists in the UK) and referral and access to the specialism is strictly limited. Thus, given the imperative for general dental professionals to identify and address the oral problems associated with scleroderma as early as possible in the disease process, whilst mouth opening is optimal, it is imperative that they are cognisant of the possible dental impacts of scleroderma to enable early identification and treatment (whether in the general surgery or via referral to Special Care Dentistry). In this context, general dental practitioners are critical gatekeepers in the illness trajectories of people living with scleroderma.

Open wide?

In the context of scleroderma, further opportunities for embodied disappearance from the professional gaze are presented by the disease process itself. For dentists, in particular, it is, we suggest, the conventions associated with the dentist's chair itself which highlight, particularly starkly, the problems associated with scleroderma. It is in the context of the dental 'check-up', we would argue, where, whilst perhaps not explicitly recognised by either dentists or patients, the nature of scleroderma itself works to fundamentally transgress the social and clinical mores followed by dentists and to which patients voluntarily submit themselves.

The dental examination is, Nettleton (1991) suggests, a unique area of clinical inspection and intervention, whereby patients present themselves for regular surveillance to ensure they are following the socially sanctioned rules of appropriate dental behaviour. It is in this context where the invocation to “open wide” is jointly understood and operationalised.

One of the key physical impacts of scleroderma, however, is the tightening and hardening of the soft tissues, which can be a particular problem around the face and mouth. This means that, for some people, the oral cavity shrinks (microstomia), making it particularly difficult for dentists to access the mouth and teeth. Standard dental instruments do not fit and maintaining an open mouth for any length of time can prove to be extremely challenging and painful.

> *Dentist not heard of scleroderma or the effects, don't understand that you can't open your mouth* (P003).

The closed, compromised, oral cavity associated with scleroderma is, therefore, anathema to dentists who are not able, giving the challenges associated with microstomia, to perform the expected functions of dental intervention. These include the ‘check-up’ itself (the prime seat of dental control and intervention) and the more complex interventions that may be required by people living with conditions such as scleroderma.

The taken-for-granted expectations, therefore, of both dentists and patients, that the patient will be willing, and able, to ‘open wide, are inadvertently challenged by this condition - it transgresses the taken-for-granted boundaries of professional intervention, making the shared expectations of how the mouth is managed, corrected and controlled (Nettleton, 1988; p. 164) difficult, if not impossible, to meet. In addition to their potential second-class dental citizen status, therefore, people living with scleroderma, can also be perceived to inadvertently contravene what it means to be a good (for which read, compliant) dental subject.

> *The dentist I had gone to for a great many years never accepted my illness and mouth problems as being one and the same* (P004).

Dentist doesn't have a clue about the disease and doesn't want to. When I mentioned it they nearly struck me off their list as they are NHS and do not want complex or awkward cases (P007).

Feel strongly that rheumatology don't consider oral health as important as other symptoms. After a tooth broke off during lock down I felt very vulnerable.... (P010).

On my last treatment when I pointed out problems with my mouth pending caused by the scleroderma they [dentist] remained mute. I am unsure if they even believed me I thought they took me more that I was anxious and made my symptoms up (P084).

The mouth itself, of course, also assumes a liminal embodied space where the boundaries constituting the inside and outside of the body are poorly defined but fiercely guarded. Theorising this further, we draw on Crotty (1993:109) who, in the context of her work on eating and nutrition, wrote that it is the act of swallowing which "divides nutrition's "two cultures", the post-swallowing world of biology, physiology, biochemistry and pathology, and the pre-swallowing domain of behaviour, culture, society and experience". This observation was proffered to underline Crotty's (1993) argument that the field of nutrition gives scant attention to the ways in which people's social contexts influence their food choices and eating behaviours.

It is the pre and post-swallowing worlds which are of particular interest in the context of our own work, as respondents' experiences clearly articulate the fact that rheumatologists' principle focus remains insistently on the post-swallowing world (indeed, much patient, and practitioner, facing information about scleroderma notes that the problems associated with scleroderma start in the oesophagus and can affect any part of the body thereafter). As such, the pre-swallowing world of the mouth and teeth – the social contexts in which people navigate and experience the very real oral and dental problems associated with scleroderma – are of less interest to clinicians, but, as our data suggest, of critical concern to patients. Thus, respondents' narratives demonstrate that this can result in a categorical mismatch between patients' experiences and concerns and an appropriate clinical interest and response.

Another, closely associated, reason why systemic problems associated with the mouth may be neglected by rheumatologists, in particular, relates to how the mouth has come to sit,

often uneasily, on the historically contested margins of the 'ill' body. Thus, Nettleton (1988) argued that, whereas dental problems often included diseased tissues, people who experience them are not ordinarily regarded as being *ill.* This is, she noted, because problems with dental health are not perceived as misfortune in the same way as other illnesses, but, rather, are often perceived to be failures of individual responsibility.

People who experience problematic dental health are, therefore, judged to have inadequately carried out the routinised, and socially sanctioned, practices people are expected to undertake in the context of a properly maintained and it is the dental profession that is customarily charged with policing these practices (Nettleton, 1991). People living with Scleroderma, therefore, may be inappropriately dentally, clinically, and socially, 'read' as inadequate 'dental citizens' when, in fact, they are often quite the opposite. This was the case for many of our respondents:

> *You just automatically think it's your fault; when you first have these problems with your teeth, I think cos it's drilled into to you as a child, as an adult you must clean your teeth and floss, and the first thought is 'oh my goodness, I'm doing this all wrong'* (IP11).

> *I worry that people will notice (…) nearly everybody else I know has lovely teeth, I don't know anybody with as horrifying teeth as mine. (…) I probably am worried about what they're, what they're thinking, what they're assuming I do to my teeth…* (IP11).

> *I'm conscious people may think my dental hygiene is poor* (P068)

Our data demonstrate that people living with scleroderma transgress and unsettle the taken for granted expectations and understandings of what constitutes good dental hygiene and care in that, whilst they evidently *do* undertake oral and dental maintenance as expected and required, the illness processes associated with scleroderma fundamentally undermine their efforts situating, in the process, the mouth at the centre of many people's illness experience. In so doing, some people living with scleroderma inadvertently experience a type of iatrogenic stigma (Sartorius, 2002; White, 2007) that would, ordinarily, be reserved only for those who evidently transgress these social (and moral) conventions and expectations.

The dys-appearing mouth

We have, thus far, suggested a number of reasons for the evident mismatch between patients' and clinicians' experiences of, and responses to, ODMS and we now turn to some of the impacts of this disparity.

As we have noted, respondents' narratives suggest that their concerns, and experiences, in the context of their oral and dental problems are not seen, or recognised, by clinicians, such that these issues are invisible to one-half of the patient/clinician dyad. A concept, then, that unifies these ideas is that of 'dis/dysappearance' in both literal and metaphorical contexts. In the context of scleroderma, in particular, there are multiple ways these varied disappearances and absences can occur and be experienced.

First, as noted earlier, there is the very real (physical) disappearance of the mouth and oral cavity which occurs as a result of the disease process in scleroderma, where, as previously outlined, the mouth and oral cavity can shrink, the lips recede and, particularly in diffuse scleroderma, the soft tissues of the face harden, resulting in a mask like facial appearance.

> *My lips have disappeared and my mouth is shrinking* (P013).
>
> *I'm very aware my top lip is disappearing* (P038).

These physical changes, whilst perhaps less immediately visible to other people, can have a profound effect in the context of the embodied experience of the illness.

> *I feel very sad that my smile has gone and people find it hard to interpret my meaning because of lack of facial expression and my speech difficulties (also due to scleroderma)* (P061).

These literal disappearances are accompanied by less visible, but equally impactful, metaphorical absences and, in this context, it is helpful to turn to the work of Leder (1990) who argued that, whilst human bodies are indisputable proof of our physical existence, they, nonetheless, spend significant amounts of time in what might be termed the corporeal background, such that we tend to be unaware of much of the body when it is not in use.

Leder (1990) referred to this as a corporeal absence, which can include any part of the body, including the internal organs which, ordinarily, operate beyond the boundaries of awareness. This mode is characterised by "a productive unawareness of the functioning of one's own body" (Wilson, 2012:7). So, for the most part, the mouth and teeth exist in this same somatic background as taken for granted, largely unproblematic, aspects of everyday life.

Illness, disfigurement, psychological distress, and social pressures, however, tend to shift the body into the foreground, where it can assert itself in dysfunctional ways. Then, the body loses its previously taken-for-granted background status, instead assuming an insistent presence wherein the previously taken-for-granted body is compelled into an active consciousness. Leder (1990) refers to this as 'dysappearance'.

> *It's only when you realise like brushing your teeth is really difficult because it's just so painful that you start to think about it more…*(IP04).

Leder's (1990) work foregrounds the body as a site of self-assertion and a vehicle to communicate identity, but also a site prone to dysfunction and dys-appearance, with the potential to cause a "cleavage between body and self" (Leder, 1990: 77). In response, Leder (1990) illustrated how narratives of dys-appearance can be accompanied by those of restitution; an urge to take action and alleviate the intrusive distress of the dys-appeared body by returning it to the corporeal background.

Respondents' efforts to address the dys-appearing mouth in this context took different forms. A frequently mentioned psychological and social impact of ODMS was the urge to hide the mouth.

> *I don't smile anymore and I put my hand over my mouth a lot* (P113).
>
> *I hold my hand to my mouth as I'm embarrassed* (P141).
>
> *I'm reluctant to laugh or to smile because so much is revealed, so I tend not to* (IP08).

The act of hiding the mouth can be viewed as a symbolic silencing of the dys-appearing mouth, which, both metaphorically and literally creates a barrier between the contested internal and external worlds of the mouth and an attempt, perhaps, to forcibly return the

mouth to its rightful state of corporeal invisibility. Achieving this enables the short-term comfort of a body and self temporarily in alignment, but perhaps at the cost of placing a barrier between the internal world of the individual and their embodied reality, and indeed, between the individual and others. Thus, the dys-appeared mouth not only causes a "cleavage between body and self," but also between body, self, and others (including professionals) and it should be stressed that the alleviation of dys-appearance, to the participants in our study, was elusive; highly sought, yet rarely achieved.

Our respondents' narratives clearly articulate that, whilst problems with the mouth and teeth come to assume an indisputably 'front of stage' importance, this embodied prominence is neither recognised, understood, or validated by clinicians. The final disappearance to which we refer here, then, and one which has proved to be perhaps the most frustrating for patients, is that which occurs in clinical settings where, despite the prominence given to ODMS by the patient, these issues are effectively 'disappeared' from patient/ practitioner interactions in both rheumatology clinics and dental surgeries.

When the mouth becomes invisible, the relentless intrusion of the problem, from the perspective of patients, surpasses clinical concerns and, for some, becomes an object of existential threat (*"I am frightened that I will not survive scleroderma because of my mouth"* P061). As such, one of the key objectives in our work is to help both patients and practitioners generate more holistic clinical conversations that take account of the real-world experiences of patients whilst maintaining a clear clinical overview of the impacts scleroderma can have in all areas of a person's life. To this end, our collaboration with the project Patient Public Involvement group has resulted in patient and clinician-facing awareness-raising materials which have been endorsed by the Oral Health Foundation. Animated videos (Authors' own, 2024) offer targeted information for patients and dentists about ODMS and a patient information card offer a simple, but, hopefully, effective, way for people living with scleroderma to foreground their own clinical concerns in their interactions with dentists. Further, in purposefully foregrounding the mouth at the centre of patients' experiences of scleroderma, this study has underlined the evident need for better interdisciplinary care pathways and improved referral mechanisms between rheumatology and dentistry, ensuring that the mouth is given more purposeful clinical attention at the point of diagnosis and beyond. There is also an evident need for dental and rheumatology

training which recognises the importance of ODMS. Further, our respondents' narratives and the study's co-production activities with people living with scleroderma strongly indicate that routine inclusion of the mouth in clinical consultation is the easiest, but probably most effective, way for both patients and professionals to recognize and address ODMS through the patient journey – a simple question - do you have problems with your mouth and / or teeth puts the issues firmly on the clinical agenda whilst simultaneously addressing patients' priorities.

Conclusion

This project sought to explore ODMS generating, in the process, important theoretical insights into the experience of living with scleroderma, particularly in the context of the layers of historical, professional, clinical and personal disappearance and dysfunction we have outlined and that, we would argue, may have significant resonance in the context of other long-term conditions.

The paper has explored how patients' and clinicians' perspectives and responses to these issues appear to be fundamentally mismatched, a situation which results in a range of profound, and keenly felt, absences. This exploration has offered a welcome opportunity to explore a range of ideas that help explain how these absences occur and are navigated by both patients and professionals. More importantly, however, the project, and its findings, have also offered insights into strategies that might help support people living with scleroderma, and the clinicians involved in their care, to reposition the mouth in the 'pre-swallowing' world of the patient, where, as we have noted, embodied boundaries are difficult, but not impossible, to appreciate, regulate, and manage.

The unambiguous message in the context of scleroderma and, we would argue, other conditions in which there is a complex interplay of priorities and expectations between patient and practitioner, however, is that far greater significance and attention should (and could) be given to the patient's own account of their condition whether or not this fully aligns with the practitioner's perspective. Thus, whilst scleroderma is an indisputably complex condition, the answers to at least some of the challenges faced by patients are remarkably simple, yet they evidently remain profoundly difficult to engage.

Limitations

This study has explored a largely ignored issue in the diagnosis and treatment of scleroderma, foregrounding the experiences of people experiencing ODMS. The sample of respondents, however, was largely limited to White British Women and may have been subject to self-selection bias, as those experiencing severe ODMS may have been more likely to participate. In addition, the online survey would have been subject to the well-rehearsed limitations of this approach, namely that the population to which the survey was directed was challenging to describe and people with no/limited digital access would not have been able to participate. In addition, the inability to clarify/follow up on questions may have led to analytical inconsistency.

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