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Clinician and Patient Experiences of Managing and Living with Oral and Dental Manifestations of Scleroderma: A Scoping Review

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Abstract

Oral and dental manifestations of scleroderma are extremely common, yet they are often overlooked within rheumatology and poorly understood within dentistry. Previous research has indicated the need to understand the oral and dental experiences of people living with scleroderma and those involved in their care. This scoping review aims, for the first time, to comprehensively map what is known regarding the identification and management of oral and dental manifestations of scleroderma, how these are experienced by people living with scleroderma, and to explore key characteristics of barriers and enablers to good oral and dental care in scleroderma. A scoping review was conducted using six databases (Embase, PubMed, PsychINFO, ASSIA, Scopus, and SSCI), according to the Preferred Reporting Items for Systematic Reviews and Meta-analyses – extension for Scoping Review. Grey literature were also included. Studies were eligible for inclusion if the full text and abstract were available in English, published between 2002 and 2022, and focused on the concept of oral and dental care in adults with scleroderma, either relating to identification and management, enablers and barriers to best practice, or patient experiences and wellbeing. Qualitative research which seeks to understand patients' lived experiences was a notable gap in the literature. Similarly, there was a significant lack of focus on the oral and dental manifestations of scleroderma in rheumatology.

Three key features were identified which would facilitate best practice in research and clinical contexts: the necessity of multidisciplinary care; the necessity of centralising patient experience; and the necessity of mitigating barriers to dental care. We conclude that increased awareness of scleroderma within dentistry, and streamlining referral procedures between the disciplines of dentistry and rheumatology, to enable the early identification and management of scleroderma, are crucial.

Keywords: scleroderma; scoping review; orofacial; patient experience; early identification; multidisciplinary care; rheumatology; dentistry

Background and Rationale

Despite the prevalence of the oral and dental manifestations of scleroderma (ODMS) being far more common (80% of people diagnosed) than major systemic symptoms, there is a lack of rheumatologists' awareness of the impact oral and dental problems can have and a lack of clarity regarding appropriate referral and intervention.[1] Similarly, for dental professionals, there exists a knowledge gap regarding the identification, treatment, and management of ODMS.[2] Attending to this 'awareness and referral gap' between rheumatology and dentistry and promoting early referrals and interventions to improve dental care for scleroderma patients is of critical importance for best practice in scleroderma health and dental care.

Research suggests that some scleroderma patients with early or 'mild' presentations may be identified by clinical oral and dental features prior to diagnosis.[3] Furthermore, a systematic review of the impact of ODMS on quality of life reported a statistically significant association between orofacial scleroderma symptoms and an impaired quality of life[4]. Smirani and colleagues also provided recommendations for oral care which highlighted the need for well-coordinated multidisciplinary management of oral and dental symptoms to reduce morbidity from orofacial manifestations.

Understanding how ODMS are identified and managed, and by whom, is of particular importance. One study which surveyed dentists' attitudes reported that 51% of participants were concerned that they might cause harm due to insufficient knowledge.[5] Yet, in the same study, 95.9% expressed a desire to learn more about scleroderma. This was mirrored in a study which described the current lack of awareness of scleroderma within dentistry and the value of working with dental professionals who are empathic and willing to learn.[2] These studies provide insight into the experiences of scleroderma patients, and those involved in their care. Additionally, numerous studies have indicated the need for improvements in collaborative care between dental professionals and scleroderma specialists.[6–8] However, to date, there has been no systematic or scoping review which comprehensively maps the experiences of patients or the barriers and/or enablers to delivering high quality oral and dental care to this population.

Scoping Review Questions

Research Question

How are ODMS experienced by patients and currently identified and managed by healthcare professionals?

Secondary Research Questions

What characteristics are commonly identified as barriers and enablers to receiving good oral and dental healthcare for people living with scleroderma?

Eligibility Criteria

In line with JBI recommendations[9], we have used the Participants/Concept/Context (PCC) method to identify appropriate review questions, eligibility criteria, and to guide the search strategy.

Participants

This scoping review focuses on studies that are concerned with the identification and management of oral and dental symptoms in adults living with scleroderma, in addition to exploring patient experiences and quality of life. This includes studies from the perspective of scleroderma patients and their family and carers, doctors, such as primary care

physicians and rheumatologists, and dentists. Studies were also considered if they focused on policies, guidelines, referral processes, or healthcare systems regarding health and dental care for scleroderma patients. Studies which focused only on paediatric care or symptoms of scleroderma in children were excluded due to differences in the manifestations of scleroderma and how these are managed in children.

Concept

This scoping review explores the concept of oral and dental care in scleroderma. Specifically, we examined how ODMS are identified and managed, and by whom. We also aimed to identify the characteristics of good oral and dental care for scleroderma patients. This includes, but is not limited to, exploring the perspectives of healthcare professionals, scleroderma patients, and their family and carers, regarding enablers and barriers to early interventions and previous experiences of oral and dental care.

Context

We limited literature to that published from 2002. This limit was sufficiently broad to enable us to examine publications about scleroderma over time, but is sufficiently recent to ensure the literature is relevant to the health and dental care currently experienced by people with scleroderma, as we identified no significant shifts in policy or practice during this time that would affect data analysis. Literature from any country was considered for inclusion, however only studies written in English have been included.

Types of Sources

This scoping review considered for inclusion both experimental and quasi-experimental study designs, including randomized controlled trials, non-randomized controlled trials, before-and-after studies, and interrupted time-series studies. In addition, analytical observational studies including prospective and retrospective cohort studies, case-control studies, and analytical cross-sectional studies were considered for inclusion. This review also considered relevant systematic reviews and descriptive observational study designs, including case series, individual case reports and descriptive cross-sectional studies.

We also considered studies focussing on qualitative data including, but not limited to, designs such as phenomenology, grounded theory, ethnography, qualitative description, action research, and feminist research.

We excluded studies which solely focused on developing and validating psychometric instruments and measures, letters to the editor, and commentaries (collectively categorised as ‘wrong study design’, figure 1).

Method

This scoping review was conducted in accordance with published JBI methodology guidance.[10]

Search Strategy

The search strategy aimed to locate both published and unpublished studies. An initial limited search of PubMed and PsycINFO was undertaken to identify relevant articles. The text words contained in the titles and abstracts of relevant articles, and the index terms that describe the articles, were used to develop a full search strategy for PubMed (see Appendix I).

The databases we searched included Embase, PubMed, PsychINFO, ASSIA, Scopus, and SSCI. We also searched ProQuest Dissertations and Theses to locate relevant unpublished studies and grey literature.

Study Selection

Following the search of all databases reported above, all identified citations were collated in EndNote.[11] The final citation list was then uploaded from EndNote to Covidence.[12] Using Covidence, two reviewers (TJM, EP) removed duplicates and independently screened citations and reviewed titles and abstracts for inclusion based on the eligibility criteria. Where any ambiguity or disagreement about inclusion arose, this was resolved by a third reviewer (LW).

The full text of potentially relevant citations was then assessed in detail against the inclusion and exclusion criteria by three independent reviewers (LW, TJM, EP). Reasons for exclusion of full texts that did not meet the inclusion criteria were recorded. The results of the search and the study inclusion process have been reported in full and presented in a Preferred Reporting Items for Systematic Reviews and Meta-analyses - extension for Scoping Review (PRISMA-ScR) flow diagram.[13] A critical appraisal of the quality of included literature is beyond the remit of this scoping review and, as such, no method of quality appraisal was undertaken.

Data Extraction

Data were extracted from articles included in the scoping review by one reviewer (TJM) and checked by a second reviewer (EP) using a data extraction tool built into Covidence. The extracted data includes specific details about the participants, concept, and context. The extraction form is provided (see Appendix II).

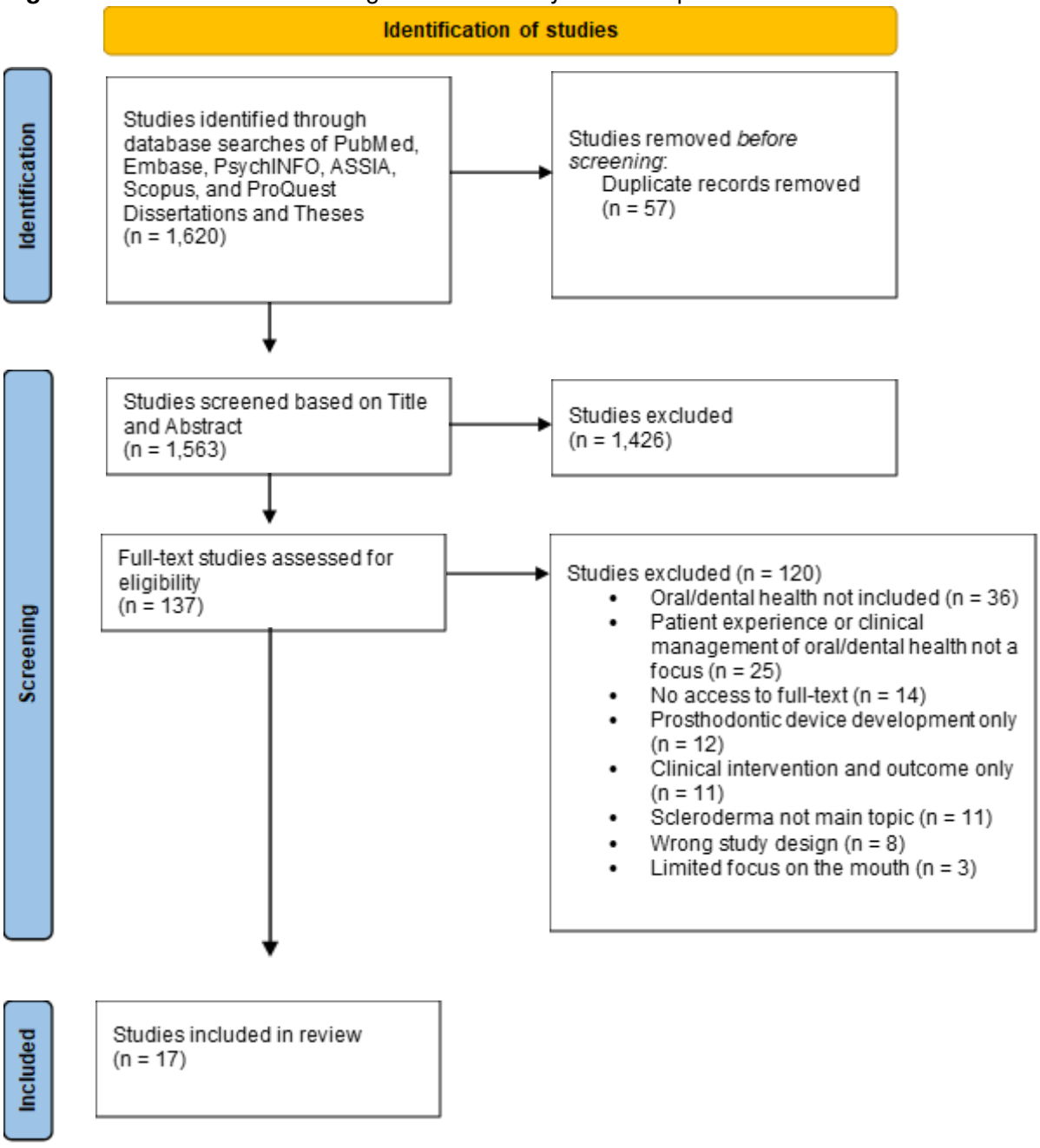
Results

Description of Included Studies

Following a comprehensive identification of relevant studies (see Figure 1), we included 17 studies which corresponded with the participants, concept, and context of this scoping review. Included study characteristics are discussed below and summarised in table 1.

We limited the search of studies to those written in English. Studies were conducted in seven countries, with the majority based in the US (41%). We searched literature published from 2002 to the date of the literature search (June 2022); included studies ranged from 2004 to 2021. The majority of studies were published between 2014 and 2021 (76%). Three-quarters were published in dentistry ($N = 13$, 76%). The majority of studies were cross-sectional ($N = 9$; 53%), with only one including a qualitative element. Where the study mentioned types of scleroderma, these typically consisted of multiple types (diffuse, limited, localised) and two included participants with overlap conditions. Only one study focused on a single type of scleroderma (limited), also the only study which incorporated a qualitative methodology. The duration participants had lived with scleroderma was typically defined as time since the onset of the first symptom of scleroderma that was not Raynaud's phenomenon; mean duration ranged from 7 years to 13.9 years. The range within studies was 1-50 years.

Figure 1: PRISMA flowchart diagram of the study selection process



Regarding the research questions related to this scoping review, eleven studies (64%) explored quality of life or psychological wellbeing in relation to oral health (as noted above, only one article addressed patients' lived experiences through qualitative methods). Ten studies (59%) discussed the identification and management of orofacial symptoms of scleroderma. Seven studies (41%) discussed barriers to good oral and dental care and four studies (23%) discussed characteristics of good oral and dental care.

Our principal aim was to explore how oral and dental symptoms of scleroderma are experienced by patients, and how they are identified and managed. We also aimed to investigate commonly identified barriers and enablers of receiving good oral and dental care. Therefore, results will be organised and discussed in the following categories 1) identification and management of orofacial symptoms; 2) patient experiences, quality of life, and

wellbeing; 3) barriers to good oral and dental care; 4) best practice in dental care for patients with scleroderma.

Table 1: Characteristics of included studies

Study information	Characteristics	Frequency (*N* = 17)	Percent
Year of Publication (range 2004 – 2021)	2004 - 2013	4	24
	2014 - 2021	13	76
Country of Publication	United States	7	41
	United Kingdom	3	17
	Canada	2	12
	France	2	12
	Italy	1	6
	Croatia	1	6
	Poland	1	6
Field of Study	Dentistry	13	76
	Rheumatology	6	35
	Oral Medicine	4	24
	Occupational therapy	2	12
	Psychology	1	6
Study Method	Cross-sectional	9	53
	Literature review	5	29
	Mixed methods	1	6
	Case report	1	6
	Text and opinion	1	6
Type of Scleroderma	Diffuse; limited	7	41
	Diffuse; limited; localised	5	29
	Diffuse; limited; overlap	2	12
	Limited	1	6
	Not mentioned	1	6
	Not applicable	1	6
Disease Duration	Mean; Range (years)	7-13.9	0.5 - 50
Total Participants	Eligible studies; Range	10	31 - 394
Concept	Orofacial Symptoms	10	59
	Dental care (clinical)	8	47
	Quality of Life	7	41
	Dental Hygiene	4	23
	Psychological wellbeing	3	17
	Healthcare systems/provider knowledge	2	12
	Patient experience	1	6

Identification and Management of Orofacial Symptoms

Ten studies discussed the identification of orofacial symptoms. Of these, six were in the context of clinical presentations and radiographic findings in a dental setting,[5,14–18] three related to symptoms identified for the purpose of the research,[19–21] and one was in the context of patients with scleroderma educating healthcare providers about the effects of orofacial symptoms.[2]

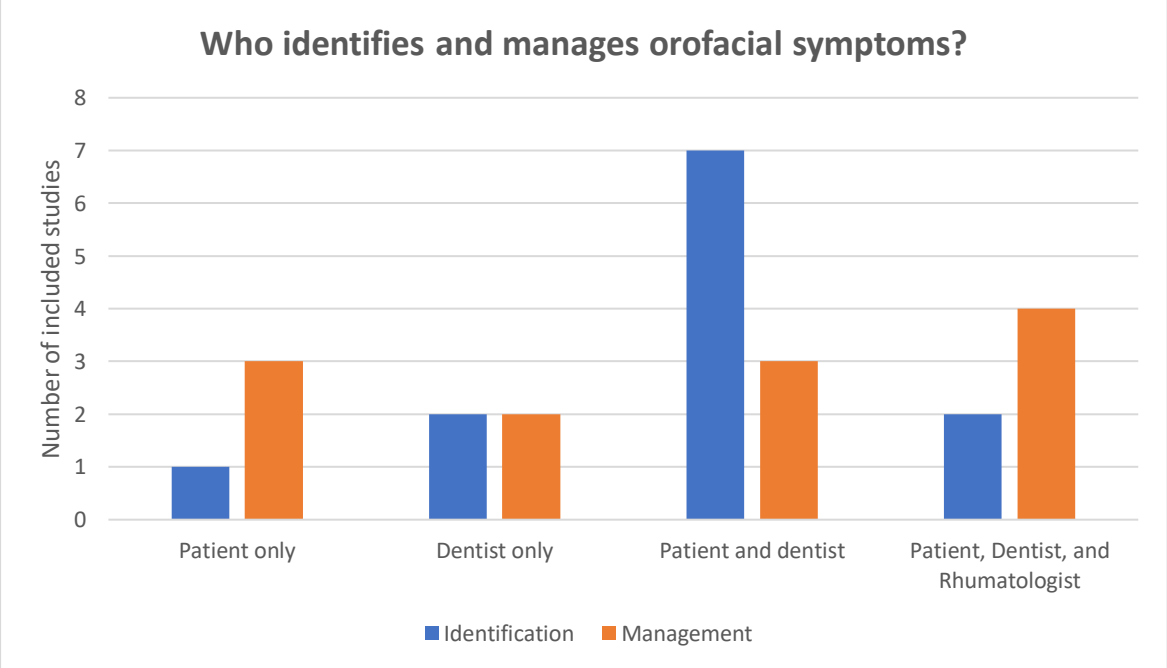
The most prevalent early indicators of scleroderma in dentistry were widening of the periodontal ligament space (PDL) and trigeminal neuropathies (TN). In five of the ten studies (50%) that discussed orofacial symptoms,[5,15,17–19] generalised widening of the PDL was

noted as being pathognomonic for scleroderma and an early diagnostic radiographic finding that is uniquely identifiable. Similarly, TN as an early symptom of scleroderma, was noted in 50% of the literature.[2,15,17–19] Therefore, it is recommended by Tolle[15] and Puzio et al.,[18] that dentists should be aware of features associated with scleroderma such as TN, Raynaud's phenomenon (RP) and Sjogren's syndrome, which may precede, or co-occur, with scleroderma. These authors recommend that dentists who observe the co-occurrence of widening of PDL, TN, RP, or Sjogren's syndrome should suspect scleroderma and make appropriate referrals to specialists, to enable early diagnosis and interventions.

Dentists are uniquely placed to be able to observe early clinical and radiographic oral changes related to scleroderma and are able to facilitate not only the treatment of orofacial symptoms but also early referral to rheumatology for diagnosis and management. Beaty et al.'s[2] exploration of the oral health experiences of 48 patients with limited scleroderma, however, revealed that, paradoxically, participants were required to actively educate their dentists about the disease.

In total, twelve studies included discussions related to who identifies, and is responsible for, orofacial symptoms and their management (see figure 2). Overwhelmingly, the identification of orofacial symptoms was seen to be a collaboration between patients and dentists, whereas their management was viewed as a multidisciplinary effort; with dentists responsible for providing dental care, and patients responsible for maintaining oral hygiene to prevent the progression of microstomia. Where rheumatologists were included, they were seen to be responsible for prescribing and managing medications and potential side effects. It was also noted that dentists should be knowledgeable about the effect of medications on the mouth (particularly causing ulcers and dryness) and also aware of methods and products that could mitigate these. The collaborative elements of identification and symptom management demonstrates the value of supportive and trusting healthcare professional and patient relationships.

Figure 2: Bar chart showing the perceived roles of patient, dentist, and rheumatologist in the identification and management of ODMS (as discussed in relevant included studies, *N* = 12)



Patient Experiences, Quality of Life, and Wellbeing

Eleven studies in this scoping review included discussions of quality of life or psychological wellbeing.[2–4,8,14,17,20,22–25] Ten studies used one or more psychometric measures of psychological wellbeing or quality of life, and most used either the Oral Health Impact Profile (OHIP, *N* = 7) or the Mouth Handicap in Systemic Sclerosis questionnaire (MHISS, *N* = 6). Only one study used qualitative methods to investigate the impact of orofacial symptoms on the lives of people living with scleroderma; this study also used both the OHIP-14 and MHISS to explore the impact of scleroderma on quality of life. Where studies included consideration of the psychological impact of scleroderma, this focused on symptoms of depression and/or anxiety (*N* = 5).

Two cross-sectional studies found that oral health-related quality of life (OHQOL) was significantly worse in people with scleroderma compared with controls.[8,20] This finding was also reported in two narrative reviews[3,17] and one systematic literature review.[4] Furthermore, low OHQOL was significantly associated with low global quality of life in two cross-sectional studies[2,24] and in three literature reviews. [3,4,17]

It is clear from the included articles that OHQOL is diminished for people with scleroderma, and that this is related to lower global quality of life. However, the relationship between disease severity and quality of life is less clear in the literature. For example, Parat et al.,[25] reported that more severe disease activity was correlated with lower OHQOL, whereas Baron et al.[24] reported no statistically significant relationship between disease severity and OHQOL. Both studies measured physician-assessed disease severity using the Medsger disease severity scale and the modified Rodnan skin score. Parat et al.,[25] however, also included a patient-assessed severity rating, using the Scleroderma Health Assessment Questionnaire (SHAQ). Therefore, this may imply that physician assessments of severity do not fully capture aspects of scleroderma that have a negative influence on OHQOL. Amin et al.'s[22] exploration of the psychological impact of facial changes in scleroderma, which demonstrated the substantial psychological impact the orofacial symptoms of scleroderma may have on an individual, provides further evidence that physician assessed disease-severity based only on organ involvement is not sufficient to understand the impacts of scleroderma on patients' lives.

Four cross-sectional studies investigated the relationship between specific orofacial symptoms and quality of life. Baron et al.[20] report that people with scleroderma had more oral abnormalities and worse OHQOL compared with a healthy control group. However, the extent to which these oral abnormalities contributed to a diminished quality of life in the sample of people with scleroderma was not investigated. Beaty et al.,[2] found that xerostomia was the symptom most frequently associated with reduced OHQOL, as measured by OHIP-14 and MHISS. The challenges associated with xerostomia also meant that daily oral hygiene practices were challenging. Additionally, feeling self-conscious, embarrassed, or anxious about their oral health was commonly reported to have a negative effect on quality of life by participants in this study. Beaty et al.'s[2] findings provide support for the results of Amin et al.'s[22] research which demonstrated that 'noticeability' and 'severity' of facial differences had a significantly negative effect on self-esteem and anxiety. Similarly, Abdouh et al.,[8] revealed that 50% of participants with scleroderma reported difficulties in smiling without embarrassment, in comparison to just 6% of participants without scleroderma.

The mutuality of the relationship between psychological wellbeing and oral health in people with scleroderma was highlighted in two cross-sectional studies[8,23] and two literature reviews.[4,14] Yuen et al.[23] explored factors associated with oral hygiene in 178 people with scleroderma and found that clinically significant symptoms of depression reduced the

likelihood of brushing teeth twice daily by 71%. Furthermore, Abdouh et al.[8] discovered that 58% of scleroderma patients had moderate to extreme dental anxiety, and had significantly worse OHQOL in comparison to a control group. These studies demonstrate that symptoms of depression and anxiety are associated with OHQOL in people with scleroderma, and, as noted by Smirani et al.,[4] this can cause a spiral of reduced oral hygiene behaviours and worsening depression and self-image, which can cause patients to neglect their oral health. Challenges in maintaining oral hygiene and psychological wellbeing can be compounded by orofacial features of scleroderma, such as xerostomia, microstomia, and reduced manual dexterity. Leader[14] states that factors such as scleroderma-related facial changes that restrict expressiveness and movement can be distressing as it may cause patients' appearance to no longer match their self-image.

Barriers to Good Oral and Dental Care

Seven of the studies in this scoping review discussed factors which may act as barriers to good oral and dental care for people with scleroderma (see table 2 for a summary).[2,5,8,17,21,23,25] As discussed in the previous section, symptoms of depression[23] and increased levels of dental anxiety[8] can also constitute barriers to good dental care.

Socioeconomic status (SES) may also present barriers to good oral and dental care both in terms of access to dental professionals and oral self-care. Yuen et al.,[21] found that low income was related to not flossing and not having visited a dentist within the past 12 months. Similarly, Parat et al.'s[25] exploration of the relationship between SES and OHQOL for scleroderma patients found that lower SES was associated with worse OHQOL. Moreover, Beaty et al.'s[2] examination of the oral health experiences of people with limited scleroderma revealed that the financial burden of dental care is often a prohibitive factor. These financial difficulties were further compounded because participants reported having to purchase a range of oral hygiene aids and products which were not always helpful.

Two studies highlighted that scleroderma related physical limitations can also be a barrier to oral and dental care.[2,23] Restricted manual dexterity, in particular, was cited in both studies as a prohibitive factor in the maintenance of good oral hygiene. An additional six studies mentioned the necessity of using adaptive devices to maintain oral hygiene.[3,14,15,18,20,21] These adaptions were, however, discussed in the context of the management of symptoms, rather than conceptualised as a barrier to oral and dental care.

Three studies also mentioned physical barriers to dental care.[2,5,17] These barriers were related to the clinical environment, healthcare systems, and healthcare provider attitudes. All three articles discussed patient-reported difficulties of finding a dentist knowledgeable about the condition and willing to treat people with scleroderma. Leader et al.[5] surveyed dentists about their knowledge of scleroderma, and, whilst 71% of dentists felt prepared to treat a patient with scleroderma, 51% were worried they would cause harm by not fully understanding how to provide care, and only 51% felt confident that their staff would be able to assist a patient with scleroderma. 95.9% of dentists in the study were interested in learning more about scleroderma. This is important because Beaty et al.[2] highlighted that patients expressed a need for more understanding and knowledge from their dentists, and that they would purposefully seek out those who were willing to learn about the condition. Jung et al.[17] and Beaty et al.[2] also discuss environmental barriers patients may encounter while trying to access dental care. Patients in both studies reported pain during dental procedures, often because the equipment was too large or the patient found holding their mouth open too fatiguing. Further barriers may be experienced due to the dentist's response to difficulties encountered in the course of providing dental care. For example, one patient in Beaty et al.'s[2] study reported "a new dentist who screamed at me because I couldn't open

my mouth large enough". Such difficulties were commonly reported in this study, demonstrating that previous negative experiences, or the expectation of struggling to access dental care, can be a significant barrier to seeking dental care.

Table 2: Summary of barriers and enablers of good oral and dental care for people with scleroderma

Barriers	Enablers
SES/financial burden preventing regular dental appointments and the procurement of adaptive aids to maintain dental hygiene	Good communication and multidisciplinary working between healthcare and dental care professionals
Depression and anxiety as a result of previous negative dental experiences, anticipation of pain or stigma, and the psychological effect of living with scleroderma	Dental providers should have compassion, empathy, and patience
Physical restrictions (e.g., microstomia, digital ulcers, etc.) preventing daily maintenance of dental hygiene and limiting oral access for dentists	Dentists should provide accommodations during appointments (e.g., short appointments, more breaks, warmer room)
Lack of understanding among dental professionals of scleroderma and orofacial effects	Increased professional knowledge of scleroderma in dentistry, and/or willing to learn more

Best Practice in Dental Care for Patients with Scleroderma

Four studies discussed factors which enable good oral and dental care (Scardina & Messina, 2004; Leader, 2007; Tolle, 2008; Yuen et al., 2014b).[14,15,19,21] See Table 2 for a summary of enablers of good oral and dental care for people with scleroderma.

The four studies highlighted the importance of awareness of scleroderma among dental professionals. Scardina & Messina,[19] Leader,[14] and Tolle[15] report that, in order to provide best practice in dental care for people with scleroderma, dental professionals should be able to identify orofacial symptoms of scleroderma to enable early diagnosis and treatment. Additionally, Tolle[15] and Yuen et al.,[21] state that dental professionals should take manual dexterity into account when advising the patient on maintaining oral hygiene, and should be able to recommend adaptive devices to assist the patient if necessary, such as an oscillating-rotating-pulsing electric toothbrush or alternatives to flossing. Furthermore, Tolle[15] adds that dental professionals should be aware of the psychological effects of living with scleroderma, including increased pain and lower self-esteem. In their summary of characteristics necessary in dental professionals to provide high quality care to people with scleroderma, Leader[14] reports that all dentists should have sensitivity, patience, and ingenuity.

Only one study offered practical recommendations for dental professionals. Tolle[15] advised that dentists should prepare appropriate devices and be aware of signs of patient discomfort during dental treatment; short appointments with breaks if needed were also recommended. Additionally, the author recommends the room temperature should be higher than usual, or blankets offered to patients who may have Raynaud's phenomenon or digital ulcers.

Two studies highlighted the role of multidisciplinary working. Yuen et al.,[21] state that dentists may need to refer patients with restricted manual dexterity to occupational therapists for assistance with maintaining oral hygiene. Additionally, Leader[14] highlights the importance of maintaining good communication between dental and medical professionals when coordinating dental treatment. Although four studies of the 17 included in this scoping review identified rheumatologists as important in managing oral and dental care (see figure 2), it was only Leader[14] who discussed the importance of rheumatologists in facilitating good oral and dental care for people with scleroderma.

Discussion

This scoping review presents, for the first time, an in-depth synthesis of literature from the last 20 years relating to the identification, management, and experiences of ODMS. Four key themes were identified: 1) identification and management of orofacial symptoms; 2) patient experiences, quality of life, and wellbeing; 3) barriers to good oral and dental care; 4) best practice in dental care for patients with scleroderma. The following discussion details three implications for research priorities and clinical practice which emerged from the four key themes. At the end of this section we summarise these implications, and discuss the limitations of this study.

The Necessity of Multidisciplinary Care

A central finding of this scoping review was that dental professionals are uniquely situated to identify scleroderma by the prodromal or early manifestations of orofacial symptoms. Therefore, they are also able to facilitate early referral to rheumatology. Some early features, such as widening of the PDL, are seen as pathognomic to scleroderma, however other features, such as TN or Sjogren's syndrome, can be associated with a multitude of other conditions. Awareness of such features is particularly important in relation to Sjogren's syndrome and sicca symptoms; Sjogren's occurs secondary to scleroderma in approximately 30% of patients, however, approximately 75% of patients experience sicca symptoms[26] Additionally, Mouthon et al.[27] propose that sicca symptoms (e.g. mouth dryness) may be a direct symptom of scleroderma. Therefore, it is crucial that dentists have a good working knowledge of these features and their possible implications. It is not feasible for all dental professionals to have a detailed knowledge of each rare condition they may encounter; accordingly, it is important that dentists have the ability to quickly access clear guidelines relating to scleroderma. Our results suggest that these guidelines should enable dental professionals to understand the constellation of symptoms that would necessitate referral to a rheumatologist, and should elucidate how to make appropriate and timely referrals based on presenting symptoms.

Despite 35% (N = 6) of studies in this scoping review indicating that rheumatologists play an important role in the identification and management of orofacial manifestations of scleroderma, particularly in relation to diagnosis and treatment, only one identified multidisciplinary working between rheumatologists and dentists as a key factor in facilitating good oral and dental care. Furthermore, the majority of studies (76%) were published in the field of dentistry, indicating a gap in the literature regarding the clinical management of orofacial symptoms of scleroderma in the context of rheumatology. This supports previous claims that oral and dental symptoms of scleroderma are typically neglected in a rheumatology context.[1]

Dental professionals may be the first clinician scleroderma patients will consult to discuss their oral and dental concerns (although they may lack scleroderma-specific knowledge), which demonstrates the necessity for multidisciplinary working and increased

communication between dentists and rheumatologists. This collaborative working should include the sharing of information, advice and guidance, co-creation of treatment plans, and referrals to occupational health and psychological services to help patients maintain dental hygiene and improve QoL. We believe this would ensure that dentists are able to provide safe, high-quality dental care for scleroderma patients, and would emphasise the importance of attending to patients' oral and dental needs early in the process of diagnosis and treatment.

The Necessity of Centralising Patient Experience in Research and Clinical Practice

The results of this scoping review suggest that we cannot understand the full extent of the psychological impact of living with orofacial manifestations of scleroderma through physician-assessed disease severity alone. Limiting understanding to difficulties arising only from functional disability or disease severity neglects to consider the psychological and emotional impact of the daily challenges of living with scleroderma. This not only concerns the 'noticeability' or 'severity' of facial changes, but also the extent to which people living with scleroderma are able to confidently engage in their social, economic, and recreational lives.

The considerable psychosocial burden of living with scleroderma, and its effects on oral health, has clear implications for research and clinical practice. We found that the lived experiences of people with scleroderma were under-represented in the literature.[2] Therefore, prioritising the inclusion of patient experience into scleroderma research is crucial. To enable clinicians to understand the extent of functional and psychosocial impacts, patient-reported outcome measures (PROMs) could be utilised in routine clinical practice. The most commonly used PROMs in the included studies were OHIP[28] and MHISS[27]; these measures are highly correlated and psychometrically sound. OHIP is not specific to scleroderma, and therefore may better capture the experiences of individuals with overlap/comorbid conditions, and it has a more explicit focus on psychosocial impact in comparison to MHISS. Additionally, it has a short version which is easier to administer in routine clinical settings (OHIP-14[29]), and a version for edentulous patients (OHIP-EDENT[30]). However, clinicians should keep in mind that MHISS may be better suited to indicate functional change and/or to examine treatment efficacy, due to it being designed specifically for use with scleroderma patients[31].

Moreover, it is particularly important for people living with scleroderma to receive appropriate emotional and social support; some individuals feel demoralised by the progressive nature of scleroderma, while others become socially isolated and depressed, all of which may cause them to neglect their oral health. This provides further evidence that a patient-centred multidisciplinary approach to care would be beneficial, not only for patient wellbeing and OHQOL, but also for adherence to medical treatment, oral hygiene practices, and physical therapy such as stretching the oral aperture. This approach may also encourage regular attendance at dental services. Further, due to the embarrassment and stigma that people living with scleroderma may experience, a compassionate and active approach to addressing these concerns in both rheumatology and dental clinics may reduce the hidden nature of oral health challenges.

The Necessity of Mitigating Barriers to Dental Care to Facilitate Best Practice

Although the limited knowledge and stigmatising attitudes of dental professionals were often mentioned as barriers to accessing dental care for people with scleroderma, only one study explored this barrier from the perspective of dentists.[5] Despite just over half of dentists in this study feeling worried about causing harm due to not understanding the condition, it is positive that almost every dentist surveyed (96%) was interested in learning more about scleroderma. Similar results have been found in studies that focus on the knowledge of

dentists in relation to other autoimmune conditions. For example, Song et al.[32] reported that dentists desired more information about the relationship between oral health and systemic conditions, and discussed the difficulties of providing dental treatment for patients with medical conditions where no clinical dental guidelines exist.

The knowledge and attitudes of UK dentists in relation to treating people with scleroderma represents a critical gap in the literature; no research that focuses on UK samples has explored dentists' knowledge of scleroderma nor has it explored patient experiences of dental care. Though the results seen in US samples are promising, further research should examine the views and knowledge of UK dentists.

Additionally, it is important to recognise that in addition to the specific barriers that people living with scleroderma may face in the context of dental care, global barriers to healthcare compound these. The World Health Organisation (WHO) recently published a Global Oral Health Status Report, which emphasises that oral diseases are socially, culturally, and geographically bound, resulting in oral health inequities that largely affect marginalised and vulnerable groups. Barriers which include poor access to dental services, financial burden of dental care, socioeconomic and social circumstances, among others, could be mitigated through the inclusion of oral healthcare in universal health coverage programmes.[33]

Limitations

Although this was a broad and comprehensive scoping review, the study design has some limitations. By restricting the language to English only, we may have excluded relevant literature, and over-represented literature from countries for whom English is a first language, or those that typically publish in English. Additionally, studies which did not explicitly include 'scleroderma', 'localised scleroderma' or 'systemic scleroderma' in the title or abstract may have been missed. Finally, additional factors exist which may affect ODMS, but did not appear in sufficient frequency or detail in the included studies to allow full consideration. These include, but are not limited to, diet, probiotics, autoantibody pattern, medication and side effects, tobacco and alcohol use, and environmental setting.

Research Suggestions

This scoping review found a lack of literature regarding oral health in scleroderma in the context of rheumatology, as such, increased research on this issue by rheumatologists is recommended. Future research could explore the role of rheumatologists in facilitating the identification and management of orodental problems; these are significant issues for patients and we hope this scoping review highlights the necessity of increased attention from rheumatologists regarding ODMS. We suggest that researchers should seek to establish current patient demand for comprehensive multidisciplinary services, and their preference for the provision of scleroderma-specific information about oral and dental manifestations. Additionally, researchers should aim to develop training and educational resources for dental professionals to aid in the identification and management of ODMS. These should incorporate consideration of the psychosocial impact of ODMS to improve the dental experiences of people living with scleroderma. Multiple studies in this review noted that frequent dental visits are essential, however there was no consensus on the specific frequency of these appointments; an exploration of the effect of dental visit frequency on ODMS would be informative.

It would also be interesting to explore the financial impact of dental care in UK scleroderma populations, given the significant morbidity related to oral health in scleroderma and the necessity of regular dental care, treatment, and additional adaptive devices.

Similarly, given the observed relationship between low SES and poor OHQOL, it would be prudent to examine this in low- and middle-income countries, which may have limited healthcare infrastructure and related oral health resources for patients and clinicians.

Clinical Implications

As is it likely the majority of scleroderma patients will receive diagnosis and/or treatment from a rheumatologist, it would be prudent to explore the integration of routine inquiries into patients' dental health and orofacial concerns at the point of diagnosis. As rheumatologists will have a more detailed working knowledge of scleroderma, it is crucial that they are fully aware of ODMS and the detrimental effects this has on QoL and wellbeing. Incorporating discussion of ODMS into routine clinical practice may enable early intervention regarding dental hygiene and treatment, and similarly may promote communication and collaboration between rheumatologists and dental practitioners.

To facilitate best practice in dental care for people with scleroderma, it is critical that accessible and practical information for dental professionals working with this population is easily available. Therefore, we recommend that developing guidelines for dental professionals working with this population should be a priority, with improving early diagnosis and interventions through streamlining referral processes, both from, and to, rheumatology.

Most importantly, a patient-centred, multidisciplinary care approach is recommended. This would facilitate increased communication between all clinicians involved in coordinating the care of people living with scleroderma. Furthermore, establishing and maintaining a supportive and collaborative relationship between clinicians, dental professionals, and patients would also aid in the management of orofacial symptoms. This may also mitigate some barriers that scleroderma patients commonly experience, such as dental anxiety, shame and stigma, and previous negative experiences of dental care. Through understanding patients' lived experiences, rheumatologists and dentists alike would gain a better understanding of the oral and dental problems that people living with scleroderma experience, and thus, may promote creative and compassionate interventions for dealing with these issues.

Table 3: Summary of recommendations for dentists, rheumatologists, and all clinicians to achieve best practice in oral and dental care in scleroderma

Dentists	**Rheumatologists**
• Seek personalised information from patients and their healthcare team	• Routine inquiries into oral and dental health at point of diagnosis
• Provide accommodations in appointments and plan for limitations (e.g., smaller instruments, longer appointment times)	• Referrals to dental services to facilitate early intervention
All Dental and Healthcare Clinicians	
• Awareness of ODMS, the related negative psychosocial impacts, and barriers to care (e.g. stigma, functional limitations, mental health)	
• Communication and collaboration (requests for advice and guidance, co-created treatment plans, sharing knowledge and information)	
• Consider referral to physical therapy, occupational health, psychological services for assisting patients in maintaining dental hygiene	
• Empathy, patience, flexibility, willingness to learn	

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Conflicts of Interest

There are no conflicts of interest in this project.

Author Contributions

Tyler J. Mills: Conceptualisation, Methodology, Investigation, Writing – original draft, Visualisation; **Elizabeth Price:** Conceptualisation, Funding, Methodology, Investigation, Writing – review and editing; **Vishal R. Aggarwal:** Conceptualisation, Methodology, Writing – review and editing; **Francesco Del Galdo:** Conceptualisation, Writing - review and editing **Liz Walker:** Conceptualisation, Funding, Supervision, Methodology, Investigation, Writing – review and editing.

Data Access statement

For the purpose of open access, the authors have applied a Creative Commons Attribution (CC BY) licence to any Author Accepted Manuscript version arising from this submission

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Appendices

Appendix I: Search strategy: PubMed

Search	Query	Results Retrieved
#1	("scleroderma, systemic"[MeSH Terms]) OR ("scleroderma, localized"[MeSH Terms]) OR (scleroderma [Text Word])	29,131
#2	(Dentist* [all fields]) OR (dental care [MeSH Terms]) OR (special AND care AND dentist* [all fields]) OR ("oral health"[MeSH Terms] OR oral health [Text Word])	458,665
#3	("health services"[MeSH Terms]) OR ("referral and consultation"[MeSH Terms]) OR ("rheumatology"[MeSH Terms]) OR ("dental health services"[MeSH Terms]) OR ("health services"[MeSH Terms]) OR ("referral and consultation"[MeSH Terms]) OR ("rheumatology"[MeSH Terms]) OR ("dental health services"[MeSH Terms])	2,388,375
#4	#1 AND (#2 OR #3)	1135
#5	Filter: English language only, years 2002 - 2022	931

Appendix II: Data extraction instrument

Title	
Date Published	
Journal	
Lead author contact details	
Country in which the study was conducted	
Field of study	
Aim of study	
Concept of study	
Study design	
Study funding sources	
Population description	
Inclusion criteria	
Exclusion criteria	
Method of recruitment	
Total number of participants	
Type of scleroderma	
Disease duration	
How are oral and dental symptoms identified?	
How are oral and dental symptoms managed?	
Who is expected to provide care and management for these?	
How are these symptoms experienced by patients?	
Barriers to good oral and dental care	
Characteristics of good oral and dental care	
What tools were used to measure QoL?	
What is the impact of oral health on QoL?	
What is the relationship between QoL and ssc symptoms?	