



Temporomandibular Joint Disorder in Systemic Sclerosis: A Case Report

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Abstract

Orofacial manifestations occur frequently in rheumatic diseases and usually represent early signs of disease or of its activity that are still neglected in clinical practice. Among the autoimmune rheumatic diseases with potential for oral manifestations, rheumatoid arthritis (RA), inflammatory myopathies (IM), systemic sclerosis (SSc), systemic lupus erythematosus (SLE), relapsing polychondritis (RP) and Sjögren's syndrome (SS) can be cited. Signs and symptoms such as oral hyposalivation, xerostomia, temporomandibular joint disorders, lesions of the oral mucosa, periodontal disease, dysphagia, and dysphonia may be the first expression of these rheumatic diseases. We report a case of systemic sclerosis with temporomandibular joint involvement.

Subject Areas

Rheumatology

Keywords

Scleroderma, Systemic, Autoimmune Diseases, Temporomandibular Joint

1. Introduction

Systemic sclerosis (SSc) is an autoimmune disease characterized by inflammation and hyper-reactivity of micro- and macro-vascular circulation associated with excessive collagen deposition in tissues, with subsequent fibrosis of the skin and/or internal organs [1]. SSc has a predilection for females, with an incidence of 2 - 10/one million inhabitants in the general population [2]. In addition, there is a consensus about an increase in morbidity and mortality, with an estimated 66% survival at 10 years [3]. The oral manifestations are scarcely studied and often neglected by clinicians, although leading to major functional disability. Mi-

crostomia is the most common oral finding and develops due to collagen deposition in perioral tissues, causing limitation of mouth opening, perioral groove wrinkling, and soft palate, larynx and oral mucosa stiffness [4]. Furthermore, hyposalivation and/or dry mouth are secondary manifestations of the disease. Temporomandibular disorder (TMD) can also occur, with varying degrees of subsequent resorption of mandibular branch, coronoid process, menton and condyle [5]. It is believed that these areas are reabsorbed due to the chronic collagen deposition. Tongue cancer has a significantly increased frequency in patients with SSc that present a mouth < 30 mm [6].

The resorption of some teeth has also been reported with some frequency in these patients. There may be an abnormal increase in the frequency of decayed teeth and of a atypical tooth eruption. Apparently there is also a predisposition for the occurrence of periodontal disease (PD), due to increased plaque buildup. This problem arises from the difficulty of cleaning the mouth (caused by a smaller mouth opening) and in the use of the dental brush. This latter complication is due to sclerotic changes in fingers and hands. Furthermore, the use of systemic corticosteroids for long periods influence on reducing the periodontal inflammatory response thus makes this process a progressive and often insidious one [7].

Few studies in the literature are published concerning the temporomandibular joint (TMJ) and masticatory muscles involvement in SSc. Most descriptions are based on individual cases or small numbers of patients with inadequate controls and do not define the relationship of oral manifestations to other manifestations of the disease [8]. This case report describes temporomandibular joint disorders in 47-year-old female patient with SSc, based on clinical and radiological data.

2. Case Report

A 47-year-old women with SSc was referred to the unit of orofacial pain and temporomandibular dysfunction of the University Hospital of Piauí, Brazil, with a progressive limited mouth opening and facial pain since 2 years. A history of pain in the left pre-auricular region and masticatory myalgia more pronounced in masseter muscles with a morning stiffness of jaws was depicted. These symptoms are aggravated by function. A sound in the left TMJ was also reported. She had no history of trauma or infection of the TMJ. The patient had a medical history of systemic sclerosis diagnosed by an internal medicine specialist since 10 years and treated by methotrexate, amlodipine, cholecalciferol, domperidone and acidumfolicum. The TMJ and muscle symptoms were noted since 8 years after her diagnosis. These symptoms have slowly worsened.

Physical examination of the patient revealed that SSc affected mainly her hands with a finger movement limitation, associated with sclerodactyly complicated by digital ulcers and ulcers in lower limbs.

The clinical examination of the orofacial region revealed a masklike facies with a smooth and tight facial skin with a loss of normal animation lines and a

stiffness in masticatory muscles. The mouth opening was limited. The other mandibular movements such as propulsion and deduction are also restricted. The palpation of left TMJ region causes severe pain and a restricted condylar movement. No joint sounds and no malocclusion are depicted.

Magnetic resonance imaging (MRI), in open- and closed-mouth positions, was performed to evaluate the TMJ and to check an eventual masticatory muscles fibrosis. The MRI showed degenerative changes in the temporal eminences and mandibular condyles on the left, where marginal osteophytes are noted in the mandibular condyle and subchondral bone edema. There is no evidence of significant joint effusion. Tapered left articular disc, indicative of degenerative change, slightly subluxated medially. Normally positioned right articular disc, with normal morphology and signal intensity. During the mouth opening maneuvers, there was a reduction of the left articular disc. Hypoexcursion of the mandibular condyles with the mouth opening maneuver, smaller on the left. Visible muscle groups with normal morphology and signal intensity (**Figure 1** and **Figure 2**). Arthrocentesis of the left TMJ was performed with washing with crystalloid solution and joint infiltration of corticosteroids, referring to a significant improvement in pain in the left TMJ after the procedure. After three years, she still had no pain.



Figure 1. Magnetic resonance imaging (MRI) in open-mouth position.

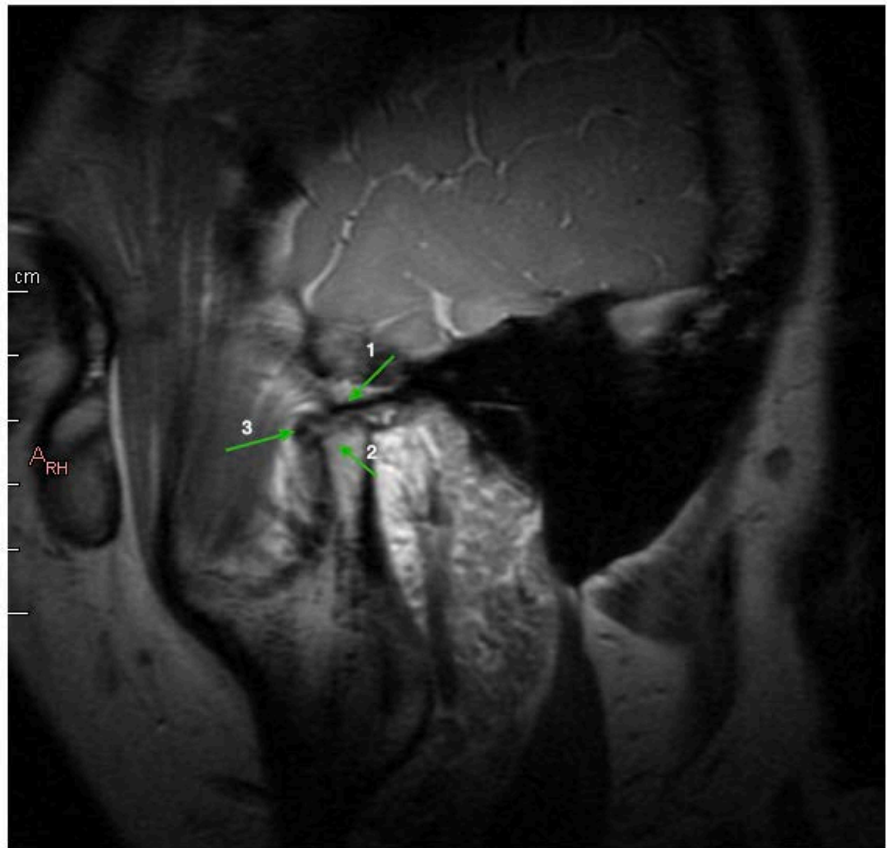


Figure 2. Image of the left temporomandibular joint in PD in the sagittal plane showing: 1) degenerative change in temporal eminence; 2) degenerative change in the head of the mandible, where are noted osteophytes and subchondral bone edema; 3) tapered articular disc, indicative of degenerative change.

3. Discussion

Systemic sclerosis (SSc) includes a variety of oral and maxillofacial manifestations. In this case, we report an internal derangement of temporomandibular joint (TMJ), disk deformations revealed in a patient with SSc.

The temporomandibular joint (TMJ) is a synovial joint and can be affected by disorders in non-articular tissues, with manifestations of muscle spasm, fibromyalgia, and myotonic dystrophy, among others. However, TMJ joint tissues may also be affected by mechanical trauma, infection, iatrogenic disorders, and gout, as well as by autoimmune rheumatic diseases such as RA and psoriasis [5]. One can observe the presence of typical inflammatory mediators of osteoarthritis, including tumor necrosis factor (TNF)- α , interleukin (IL)-1 β , IL-6 and IL8. These findings maintain correlation with the extent of the disease, *i.e.*, clinical symptoms, number of joint effusions or morphological changes [8] [9]. Some reports suggest that the findings of imaging studies are not always consistent with clinical symptoms in real-world settings and diagnoses based on Magnetic Resonance Imaging (MRI) in patients with Temporomandibular Joint Disorder (TMD) tend to more closely correlate with clinical symptom [10].

The mechanism by which TMJ involvement is seen in patients with SSc is unclear. Chebbi [11] suggested that inflammation or autoimmunity may cause the destruction of the capsular or disk attachment, resulting in internal derangement and subsequent degenerative joint diseases which result in articular surfaces changes in the condylar process and limited mouth opening. The possibility of primary articular surface resorption due to SSc is not excluded and it can result to internal derangement of TMJ which is a consequence of condylar resorption. It was suggested in the literature that the pathogenesis of primary TMJ bone lesions is due to both pressure ischemia originating from the rigidity and hardening of the overlying skin causing pressure and lack of mobility and vascular ischemia of small arterial branches of the internal maxillary artery caused by SSc [12].

A study conducted by Matarese *et al.* [13] analysed and estimated the detectable prevalence of temporomandibular joint (TMJ) symptoms and signs of dysfunction in SSc (limited and diffuse) patients compared to healthy subjects. One of the main outcomes of the study was that patients with SSc presented more symptoms and signs of TMJ dysfunction compared to healthy controls subjects. Furthermore, the statistical model used in this study has shown that in tested groups modified Rodnan skin score (MRSS) and mean disease duration values of SSc, were correlated with the worsening of maximum mouth opening, left and rightward laterotrusion, protrusion and TMJ click clinical parameters. Maximum mouth opening was also negatively influenced, to a negligible extent. This study [13] demonstrates that TMJ involvement is common in SSc patients; the findings also support the necessity for identifying TMD population based on their clinical and psychosocial impairment as an important issue to consider when assessing TMD epidemiology in this kind of patient. The present study supports the model that examination of TMJ should be encouraged in the rheumatology clinic and clinicians should be able to provide pain management and support for patients affected by SSc.

4. Conclusion

This case report reinforces the need to value the clinical complaints of patients and adequate assessment of the TMJ in patients with systemic sclerosis.

Conflicts of Interest

The authors declare no conflicts of interest.

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