Case Report

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# SURVIVAL OF LATERAL TOOTH DURING ORTHODONTIC TREATMENT IN PATIENTS WITH SCLERODERMA

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## ABSTRACT:

This case report describes the orthodontic treatment of a 16 year and 4 month old female patient with scleroderma. Extraoral examination showed facial asymmetry and straight profile. Intraoral examination revealed severe crowding and Angle Class II relationship. The panoramic radiograph demonstrated the root resorption of upper right central and lateral teeth on the scleroderma area. Upper first premolar teeth were extracted. At the end of the treatment, dental and skeletal Class I relationships were established. There is no difference for root resorption of central and lateral teeth after 18 months retention period.

**Keywords:** Root resorption, autoimmune diseases, skin, gingiva

## INTRODUCTION:

Scleroderma is a rare autoimmune disease with clinical heterogeneity and, its etiology is still unknown. It affects mesenchymal tissues. The most important characteristic of the disease is dermal collagen hypertrophy related skin thickening leading to decreased rigidity. Inflammatory and vascular changes also occur.[1] The fibrotic alterations in the skin might affect any organ system and, when accompanied by a systemic disease, the situation is then called systemic sclerosis. The prognosis of systemic sclerosis is aggressive. On the other hand, when involvement is limited to the skin, the disease is then called localized scleroderma (LS) with a better prognosis.[2,3]

Systemic sclerosis (generalized scleroderma) causes general alterations in collagen metabolism, resulting in disorders which affect internal organs.[4] Skin lesions are usually accompanied by these alterations that affect women within the age of 0-60 years. Early development of pulmonary, cardiac and renal complications might also be encountered.[5] The oral indications include xerostomia, microstomia, telangiectasia, increased decayed, missing and filled teeth.[6-9] The tongue may become rigid, making speech and swallowing difficult. The soft tissues around the temporomandibular joint were also affected, which results in pseudoankylosis. The loss of attached gingiva and gingival recession may also occur.[10]

LS mostly shows up in childhood. Sixty seven percent of the patients are diagnosed before 18 years of age.[3] Dermatomal distribution is followed by anomalies, mostly unilaterally, in the skin and subcutaneous tissues.[11] In LS, the

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expression "en coup de sabre" (SCS) is used to describe the lesions resembling a sword cut on the skin. The lesions on the face or head are accompanied by mesenchymal tissue anomalies.[12] Downwardly extending SCS might involve nose and upper lip. Furthermore, oral and gingival tissues might also be involved. The related areas facing the tongue could be atrophic.[13] In addition, the gingiva is affected by this condition and scar tissue is formed on the gingiva. Moreover, alveolar bone depression might appear.[14] This resorption might be considered to stem from vasculature changes and, skin thickening on the upper part of the bone together with loss of elasticity due to muscle atrophy. The periodontal ligaments expand in the tissues as a result of collagen accumulation or collagen deposition around the vessels and nerves.[15] Alveolar and craniofacial growth might be limited in areas affected by SCS.[3] In children, LS could prevent the growth of underlying muscles and bones.

In the literature, few orthodontic problems have been addressed in scleroderma cases.[16,17] Therefore, more case reports are needed to clarify this issue. The purpose of the present article is to describe the results of orthodontic treatment applied in patients with scleroderma despite anomalies involving oral and gingival tissues.

## CASE DETAIL:

A female patient with a chronological age of 16 years and 4 months applied to the Orthodontics Department of Kirikkale University, Faculty of Dentistry with a complaint of dental irregularity. When the patient was 5 years old, a lesion had shown up on the right half of her face and she had applied to the Gazi University Hospital Dermatology Department for eyebrow, eyelash and hair loss on the ipsilateral side of the lesion. When patient was 8 years old, she had been diagnosed LS as a result of the tests carried out in the same hospital. Additionally, she had also been suspected to have Parry-Romberg syndrome (PRS) by various specialists at different hospitals; however, that suspicion could not have been clarified. Hair, eyelashes and hair loss on the right side of the patient had healed spontaneously in time. A consultation was made with the specialist following her and information about the medications used by the patient was obtained. It was learned that corticosteroid pomade, topical calcipotriene, moisturizing cream, topical tacrolimus, D-penicillamine, methotrexate and psoralen ultraviolet-A (PUVA) therapies had been applied intermittently during that 8-year treatment period.

When the patient applied to our clinic, SCS called lesion on the face attracted attention. SCS caused an asymmetrical appearance on the face of the patient. She had a straight profile (Figure 1).

In the intraoral examination revealed Class II molar and canine relationship on both right and left sides. The upper dental midline was deviated 2 mm to the

right, the lower dental midline was in line with the facial midline. Overjet was 4 mm and, overbite was 1 mm. Maxillary and mandibular dental arch crowding were 9 mm and 4.3 mm, respectively (Figure 1). The mouth opening was about 46 mm.

The panoramic radiograph demonstrated the root resorption of upper right central and lateral teeth on the scleroderma area. There was no missing tooth, including developing third molar teeth. No condylar pathology was identified (Figure 2).

According to the lateral cephalometric analysis, skeletal Class I relationship (SNA: 80°, SNB: 78°, ANB: 2°, Wits: 1 mm), hypodivergent growth model (GoGn-SN: 24.9°), proclined maxillary and mandibular incisors (1.NA: 30.6°, 1-NA: 7 mm, 1.NB: 24.5°, 1-NB: 4 mm) existed. The upper lip was 3 mm and the lower lip was 7 mm behind the E-plane. According to the McNamara analysis, the maxilla and mandible are retrognathic (Co-A: 75 mm, A-Nperp: -3 mm, Co-Gn: 106 mm, Pog-Nperp: -7 mm) (Figure 2; Table I).

Treatment Objectives

Our treatment goals for this patient were: (1) to ensure that the right upper central and lateral teeth with resorption remain in the mouth, (2) to resolve the maxillary arch crowding, (3) to obtain a balanced profile, (4) to provide normal overjet and overbite relationship, (5) to obtain dental Class I relationship (6) to improve the health of teeth, jaw and periodontal tissues (7) to get a stable occlusion, and (8) to achieve long-lasting results.

Treatment Alternatives

The treatment options considered were:

1) To apply fixed treatment with the extraction of 4 premolars in order to resolve severe maxillary crowding and dental Class II malocclusion and, to provide a normal overjet and overbite relationship.

2) To apply fixed treatment after the extraction of upper first premolars to resolve severe maxillary crowding.

3) To apply maxillary distalization with intra-oral appliances.

Treatment Plan

The second treatment option was considered to be appropriate for the patient. Due to the presence of impacted third molar in maxilla and severe maxillary crowding, distalization idea was abandoned. Upper first premolars were decided to be treated as crowding was less in the mandible and maxillary arch crowding was 9 mm. Patient and her parents was informed about treatment alternatives and, treatment was initiated by extracting the maxillary first premolars.

Treatment Progress

Maxillary first premolars were extracted to resolve crowding in the maxillary arch under local anesthesia. The first molars were banded (Ormco, Glendora, Calif) and a transpalatal bar was applied to

prevent anchorage loss during the retraction. Metal braces 0.018-in preadjusted brackets (equilibrium 2, Roth prescription; Dentaurum, Pforzheim, Germany) were bonded. Alignment and leveling are performed using 0.012, 0.016, 0.016 x 0.016-in nickel-titanium archwire in both arches. Lacebacks made from 0.010-inch ligature wire were applied on both sides for canine distalization. After the closure of the teeth space, the extraction site was stabilized with a ligation between molars. Stripping was performed on the lower incisors after leveling in order to resolve the slight crowding in the mandible.

Because the SCS lesion in the right side of the patient also affected the upper lip, the rigidity on the lip region caused patient to feel pain during retractor placement before bonding. For that reason, it was decided to apply lip moisturizer before each treatment session.

The treatment lasted 24 months in total. At the end of the treatment, no change in root resorption was observed for the upper right central and lateral teeth relative to the resorption noticed at the beginning of the treatment. Lingual retainer was applied between maxillary second premolars to contribute to the prognosis of these teeth.

Treatment Results

At the end of the treatment, upper and lower midlines were matched, Class I canine and Class II molar relationship, ideal overjet and overbite were provided, facial aesthetics were improved (ANB: 2°, overjet: 3 mm, overbite: 3 mm) (Figure 3). Resorption in the roots of upper right central and lateral teeth has not progressed (Figure 4). The cephalometric values after treatment are presented in Table 1.

At the end of the treatment, the patient was consulted to the extraction of third molars. Right lower third molar tooth is seen to be extracted on the panoramic radiograph taken in 18 month follow-up visit (Fig 6). The extraction of the impacted lower left and upper right third molar teeth were consulted to the Department of Oral and Maxillofacial Surgery again.

After 18 months of follow-up period, clinical and radiographic evaluation demonstrated no relapse in the teeth. There is no mobility in the right upper central and lateral teeth and, resorption in the roots is observed not to be progressive (Fig 5,6).

## DISCUSSION:

When the literature is examined, it is seen that there are few cases focusing on dental problems in patients with scleroderma. When faced with problems such as resorption, complication of tooth eruption and, fibrotic gingiva,[7-10] a general evaluation of the patient should be done by a medical practitioner and the patient should be examined whether there is a problem in other ectodermal tissues. More detailed information about the facial malformation in scleroderma cases and related dental effects should be obtained. Therefore, medical doctors

and dentists should cooperate. Every scleroderma patient referred to a dentist and studies about this issue could provide a better understanding of the malformation. Dentists, pedodontists and orthodontists might be the first physicians to diagnose these patients, although they primarily do not treat scleroderma patients. Thus, this case report was decided to be prepared.

Scleroderma is caused by excessive production and accumulation of collagen, which leads to chronic hardening and thickening of the skin. Scleroderma prevalence affects two out of 10,000 people.[18,19] Worldwide incidence of scleroderma is more than the estimated one, because it is often misdiagnosed and confused with other autoimmune diseases. Diversity in clinical findings and progression rate make diagnosis difficult. In addition, early diagnosis is critical so that early therapeutic protocols might improve the lifestyle of the patient, slow down the progression, and help with disease management.[15] While approaching these patients, the psychosocial effects of a chronic disease and physical deformities on the patient and family should be taken into consideration and, support be given.

The relationship between PRS and LS is controversial. PRS is defined as progressive hemifacial atrophy without cutaneous scleroderma features. However, cutaneous changes have been frequently reported in patients with PRS. The pathogeneses of LS and PRS are thought to be similar. Therefore, LS and PRS might represent the same disease process.[20,21] In our case, LS was diagnosed at the age of 8 years by taking biopsy from the lesion; nevertheless, it had also been suspected to be PRS in different hospitals without any clarification.

Pace et al.[22] reported that the patient with SCS was found to loss central teeth as a result of bone tissue depression. In our treatment, it is seen that the right upper central and lateral teeth in the region where SCS is localized, could remain in the mouth despite resorption. Clinical control examinations performed 18 months following the treatment still indicate that the resorption in the roots of these teeth has not progressed and the absence of mobility has improved the prognosis. This implies that SCS on the face of the patient also affects the oral region, resulting in a more fibrotic structure in the gingiva, so that the teeth can stay in mouth when there is almost no bone tissue support.

The most common distress during dental treatment in scleroderma patients is the physical problem caused by limited mouth opening and tongue rigidity. By doing mouth opening stretching exercises, it can be increased by about 5 mm. If this exercise is insufficient, a bilateral commissurotomy might be needed.[10] In our LS-diagnosed patient, the mouth opening was about 46 mm. The mouth area was very rigid as the SCS lesion in the right side of the patient also affected the upper lip of the patient. This caused the patient to feel pain during

placement of the retractor prior to bonding. For that reason, it is advisable to use lip moisturizer in scleroderma patients before each session during treatment.

## CONCLUSION:

Since scleroderma disease is also presented in the mouth and the teeth, the role of dentists in early diagnosis is great. Oral health is also important in the psychological rehabilitation of the patient. Despite the presence of resorption in the teeth of LS-diagnosed patient, these teeth were kept in the mouth and, 18 months following treatment, there was no recurrence and no teeth loss. It might be a sign of the success of this treatment.

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## TABLES:

Table 1. Cephalometric measurement before and after the treatment.

| | T0 | T1 |
|---|---|---|
| SNA (°) | 80° | 80° |
| SNB (°) | 78° | 78° |
| ANB (°) | 2° | 2° |
| Wits (mm) | 1 mm | 0 mm |
| Co-A (mm) | 75 mm | 75 mm |
| Co-Gn (mm) | 106 mm | 105 mm |
| NaIA (mm) | -3 mm | -3 mm |
| NaIPg (mm) | -7 mm | -6 mm |
| SL (mm) | 53 mm | 52 mm |
| SE (mm) | 12 mm | 11 mm |
| ANS- Me (mm) | 63 mm | 62 mm |
| FMA (°) | 18 ° | 18° |
| SN-GoGn (°) | 24,9° | 24,4° |
| SN- OcP (°) | 12° | 12° |
| PP- SN (°) | 7° | 8° |
| PP-MP (°) | 18,4° | 17,3° |
| Mx1 – SN (°) | 111,5° | 109,6° |
| U1 – NA (mm) | 7 mm | 5 mm |
| U1 – NA (°) | 30,6° | 29,3° |
| L1 – NB (mm) | 4 mm | 5 mm |
| L1 – NB (°) | 24,5° | 24,8° |

| | | |
|---|---|---|
| IMPA (°) | 101° | 101° |
| Interincisal angle (°) | 122,4° | 123,7° |
| Overjet (mm) | 4 mm | 3 mm |
| Overbite (mm) | 1 mm | 3mm |
| Lower lip E- line (mm) | -7 mm | -8 mm |
| Upper lip E- line (mm) | -3 mm | -5 mm |
| Nasolabial angle (°) | 84,4° | 78,2° |
| Convexity (°) | 177,7° | 179,9° |

T0: Before treatment, T1: After treatment.

## FIGURES:

Figure 1. Initial facial and intraoral photographs.

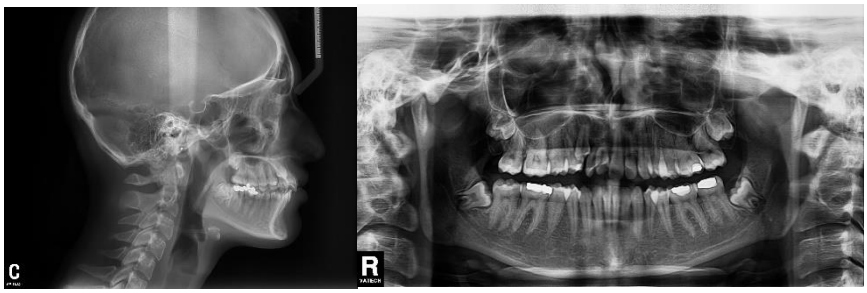

Figure 2. Initial lateral cephalometric radiograph, panoramic radiograph.

Figure 3. Final facial and intraoral photographs.

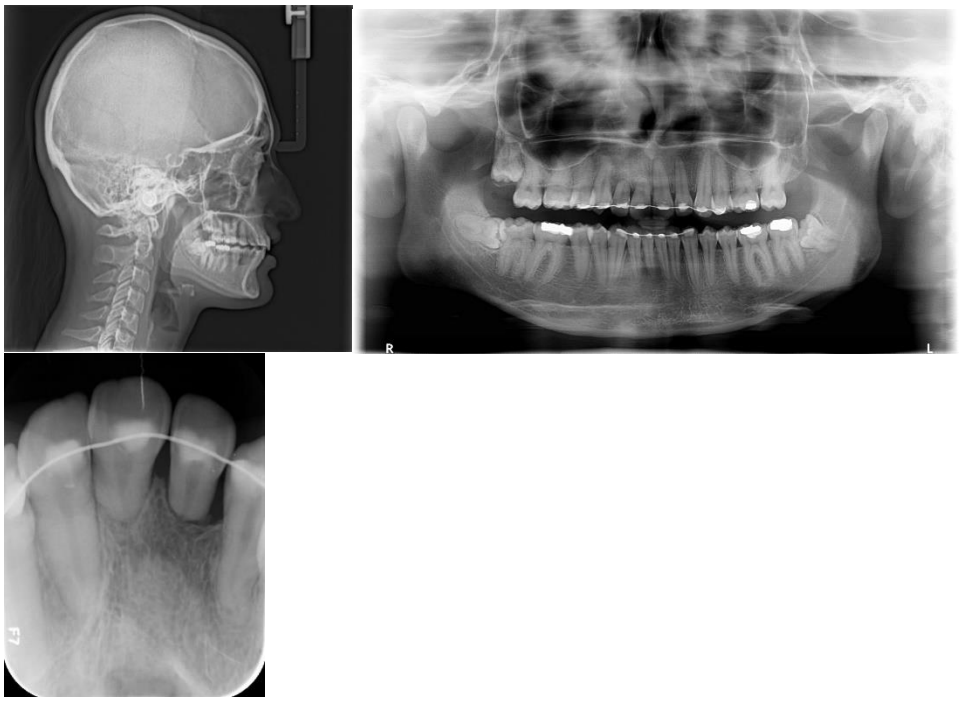

Figure 4. Final lateral cephalometric radiograph, panoramic radiograph and periapical radiograph.

Figure 5. Facial and intraoral photographs 1.5 years after treatment.


Figure 6. Panoramic radiograph 1.5 years after treatment.