

FUTYMA-GĄBKA, Karolina, PISKÓRZ, Magdalena & RÓŻYŁO-KALINOWSKA, Ingrid. Bilateral resorption of mandibular angle in patient with systematic sclerosis - a case report. *Journal of Education, Health and Sport*. 2023;13(1):18-22. eISSN 2391-8306. DOI <http://dx.doi.org/10.12775/JEHS.2023.13.01.002>
<https://apcz.umk.pl/JEHS/article/view/40706>
<https://zenodo.org/record/7348222>

The journal has had 40 points in Ministry of Education and Science of Poland parametric evaluation. Annex to the announcement of the Minister of Education and Science of December 21, 2021. No. 32343. Has a Journal's Unique Identifier: 201159. Scientific disciplines assigned: Physical Culture Sciences (Field of Medical sciences and health sciences); Health Sciences (Field of Medical Sciences and Health Sciences). Punkty Ministerialne z 2019 - aktualny rok 40 punktów. Załącznik do komunikatu Ministra Edukacji i Nauki z dnia 21 grudnia 2021 r. Lp. 32343. Posiada Unikatowy Identyfikator Czasopisma: 201159. Przynależność dyscypliny naukowej: Nauki o kulturze fizycznej (Dziedzina nauk medycznych i nauk o zdrowiu); Nauki o zdrowiu (Dziedzina nauk medycznych i nauk o zdrowiu). © The Authors 2022; This article is published with open access at License Open Journal Systems of Nicolaus Copernicus University in Torun, Poland Open Access. This article is distributed under the terms of the Creative Commons Attribution Noncommercial License which permits any noncommercial use, distribution, and reproduction in any medium, provided the original author (s) and source are credited. This is an open access article licensed under the terms of the Creative Commons Attribution Non commercial license Share alike. (<http://creativecommons.org/licenses/by-nc-sa/4.0/>) which permits unrestricted, non commercial use, distribution and reproduction in any medium, provided the work is properly cited. The authors declare that there is no conflict of interests regarding the publication of this paper. Received: 02.11.2022. Revised: 20.11.2022. Accepted: 22.11.2022.

Bilateral resorption of mandibular angle in patient with systematic sclerosis - a case report

Karolina Futyma-Gąbka¹, Magdalena Piskórz¹, Ingrid Różyło-Kalinowska¹

¹Department of Dental and Maxillofacial Radiodiagnostics, Medical University of Lublin, Poland, Chodźki 6 street, 20-950 Lublin

Karolina Futyma-Gąbka (corresponding Author):
karolina.futyma-gabka@umlub.pl, ORCID: 0000-0003-3415-8669

Magdalena Piskórz:
magdalena.piskorz@umlub.pl, ORCID: 0000-0003-4092-1122

Ingrid Różyło-Kalinowska:
ingrid.rozylo-kalinowska@umlub.pl, ORCID: 0000-0001-5162-1382

SUMMARY

Progressive systemic sclerosis (PSS) is chronic autoimmune disease affecting a connective tissue. The symptoms of PSS in orofacial area are: restricted mouth opening, xerostomia, facial asymmetry and problems with oral hygiene. Radiographic images can show specific features like bone resorption, especially in mandibular region or periodontal ligament space widening. The aim of this study was to present the case of 56-year-old woman with characteristic scleroderma-related changes visible on panoramic radiograph. The patient diagnosed with severe systemic sclerosis was referred by dermatologist to general dentist. The woman experienced tightening of facial skin, xerostomia and reduced mouth opening which caused problems with daily oral hygiene and dental treatment. General dentist referred the patient to the Department of Dental and Maxillofacial Radiodiagnostics of Medical University of Lublin for the panoramic X-ray. One of the main findings was bilateral resorption of mandibular angles. Localization of the bone resorption in patients with scleroderma is related to attachments of masticatory muscles. Dentists and general doctors should be aware that some of the maxillofacial manifestations of systematic scleroderma can be visible on panoramic radiographs.

KEY WORDS:

Scleroderma; Progressive systemic sclerosis; Panoramic images; Bone resorption

INTRODUCTION

Progressive systemic sclerosis (PSS) or scleroderma is a rare chronic autoimmune disease affecting a connective tissue with unclear etiology. In most cases, PSS occurs in females (30-50 years) but it can be also diagnosed in males [1]. Clinical presentation includes specific changes in musculoskeletal system, skin, blood vessels, heart or even lungs and kidneys. Recently, even neurological manifestations in scleroderma are taken into account [2]. The symptoms in orofacial area are: restricted mouth opening, mostly caused by fibrosis or remodelling of TMJ structures, xerostomia, facial asymmetry, atrophied lips and problems with oral hygiene. In the group of patients with systemic sclerosis even routine dental treatment is very challenging which influences on increasing number of missing teeth or periodontal diseases. Daily oral hygiene can be challenging not only because of microstomia but also bone or joint disability which affects the efficiency of hand and wrist movements [3, 4]. Radiographic images of patients with PSS can show specific features like bone resorption, especially in mandibular region or periodontal ligament space widening. Very rarely, patients may have an anterior open bite caused by bilateral damage of mandibular condyle. The aim of this study was to present the case of 56-year-old woman with characteristic scleroderma-related changes visible on panoramic radiograph. Due to the relatively rare occurrence of such symptoms, it can be considered a useful diagnostic clue.

CASE REPORT

Fifty six year old female, diagnosed with severe systemic sclerosis was referred by dermatologist to the Department of Conservative Dentistry with Endodontics, Medical University in Lublin for assessment of dental status and exclusion of potential sources of inflammation. Future treatment plan included the use of immunosuppressants. General symptoms comprised dyspnoea, hoarseness, chronic cough and swelling of the cubital joint area. The patient experienced tightening of facial skin, xerostomia and reduced mouth opening which caused problems with daily oral hygiene and dental treatment. General dentist referred the patient to the Department of Dental and Maxillofacial Radiodiagnostics of Medical University of Lublin for the panoramic X-ray. The examination revealed impacted upper right canine, periodontal ligament widening in apical area of the upper right first premolar and lower molars, several teeth after conservative and endodontic treatment and extensive bilateral resorption of mandibular angles (Fig 1). In addition, alveolar bone atrophy in the maxilla and mandible was evident. No significant inflammatory lesions were found. Despite difficulties with wide mouth opening, dentists were able to continue and finish dental treatment in this case.



Figure 1. Panoramic radiograph of the patient with systemic sclerosis showing bilateral resorption of mandibular angle.

DISCUSSION

Systemic sclerosis is a disease that influences many areas and structures in human body. Musculoskeletal system is one of the most commonly affected location. Resorption of the bone can be visible in radiographic projections. One of the most characteristic radiological features is resorption of distal parts of ulna and radius and phalanges of the hands [5]. Another skeletal structures with signs of resorption are ribs, cervical spine and zygomatic arch [6]. In the maxillofacial region one of the potentially resorbed bones due to long-term disease is mandible. The most common radiographic findings of scleroderma potentially visible on panoramic radiographs are shown in Table 1.

Table 1. Radiological features of scleroderma visible on panoramic radiographs.

<p>Bone osteolysis: - mandible angle (mostly bilateral)</p> <ul style="list-style-type: none"> - condyle - coronoid process - zygomatic arch - borders of ascending ramus
<p>Widening of periodontal ligament, especially in posterior teeth region</p>

According to Haers and Sailer [7] resorptive changes are found mostly between five up to seven years after first diagnosis of systemic sclerosis. In the other hand, Marmary et al. [8] in the study performed in 1981, resulted that there is no clinical correlation between signs of mandibular resorption and the duration of the disease. The most frequently affected part of mandible is angle, followed by condyle, coronoid process and digastric region. Localization of the resorption is related to attachments of masticatory muscles: lateral pterygoid, temporal and masseter muscle [9]. The stage of resorptive changes can be assessed on panoramic radiographs from blurring of bone outlines to severe concavity of the mandibular angles [1]. Bilateral damage of the mandibular angle is more

common than unilateral [6, 10] which is correlated with findings in our case. There are studies involving patients with symmetrical resorption of condyles and authors claim that these cases are more prone to have TMJ disorders and anterior open bite [6, 7, 11]. Regarding dental manifestations of PSS most of them are correlated with limited oral hygiene or reduced saliva production. Apart from dental caries higher risk of periodontal disease is significant. On panoramic radiographs it is possible to observe a periodontal ligament widening (PDL) but it should not be considered as a characteristic symptom of scleroderma [12]. Some authors have reported that periodontal ligament space in the posterior teeth can be wider in patients with PSS than in general population [8, 13]. Mehra [14] explains this phenomenon through increased collagen synthesis, Auluck [14] observed that periodontal widening can be caused by tautness of the masticatory muscles which leads to higher occlusal forces affecting molars and premolars. However, there is a study which showed that there was only one case from examined patients with PDL changes that had more advanced ligament space widening in posterior teeth in comparison to anterior region [12].

CONCLUSIONS

PSS is a multi-symptomatic disease and could lead to some difficulties with proper dental treatment. Some of the oral manifestations of systemic scleroderma can be visible on panoramic radiographs. Mandible resorption is one of the most characteristic radiological findings, the most common affected areas are mandibular angles.

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