CASE REPORT



UDC: 616.31-07/-08:616.724 DOI: 10.2298/VSP150522189B

# Oral rehabilitation of a patient with temporomandibular joint ankylosis caused by ankylosing spondylitis: A case report

Oralna rehabilitacija bolesnika sa ankilozom temporomandibularnog zgloba uzrokovanom ankilozirajućim spondilitisom

Zlata Brkić\*<sup>†</sup>, Nikola Pijevčević\*, Verica Pavlić<sup>‡</sup>, Milan Petronijević<sup>†§</sup>

\*Clinic for Dentistry, <sup>§</sup>Clinic for Rheumatology, Military Medical Academy, Belgrade, Serbia; <sup>†</sup>Faculty of Medicine of the Military Medical Academy, University of Defence, Belgrade, Serbia; <sup>‡</sup>Department of Periodontology and Oral Medicine, Faculty of Medicine, University of Banja Luka, Banja Luka, Republic of Srpska, Bosnia and Herzegovina

#### Abstract

Introduction. Ankylosing spondylitis (AS)/Morbus Bechterew is a chronic inflammatory rheumatoid disease. The temporomandibular joint (TMJ) dysfunction is involved in 4-35% of AS cases, and is correlated to the severity and extension of AS. Even though AS-caused TMJ ankylosis is exceptional, one should have high index of suspicion of TMJ ankylosis in AS for an early detection, because it is an extremely serious and disabling condition that causes problems with mastication, swallowing, digestion, speech, appearance and poor oral hygiene with heavy caries. Case report. A 54-year-old male patient sought medical attention at the Department of Periodontology and Oral Medicine, Clinic for Dentistry at the Military Medical Academy, Belgrade, Serbia, with the chief complaint of pain in the area of the upper left canine in the presence of limited mouth opening. The treatment plan consisted of upper left canine management and rehabilitation of the remaining teeth in the frontal and the premolar region in both, the upper and lower jaw. Even though molar region needed to be treated, unfortunately it was not in the treatment plan because ankylosis of TMJ made the treatment impossible. Conclusion. The patients with AS-caused TMJ ankylosis are considered a diagnostic challenge to routine dentistry. Accent should be given to early diagnosis and multidisciplinary approach in the treatment of the AS patients towards the favorable disease course and outcome.

# Key words:

spondilitis, ankylosing; temporomandibular joint; ankylosis; diagnosis; dental care for chronically ill; treatment outcome.

# Apstrakt

Uvod. Ankilozirajući spondilitis (AS)/Behterova bolest je hronično zapaljensko reumatsko oboljenje. Kod 4-35% bolesnika obolelih od AS zahvaćen je i temporomandibularni zglob (TMZ), a njegova zahvaćenost zavisi od težine samog oboljenja i dužine trajanja AS. Iako je ASuzrokovana TMZ ankiloza veoma retka, mora se obratiti posebna pažnja na njeno rano prepoznavanje kao veoma teškog oboljenja koje uzrokuje velike probleme pri mastikaciji, gutanju, govoru, izgledu i lošoj oralnoj higijeni s pojavom karijesa. Prikaz bolesnika. Muškarac, star 54 godine, potražio je medicinsku pomoć u Odeljenju za bolesti usta i zuba na Stomatološkoj klinici Vojnomedicinske Akademije u Beogradu, Srbija, zbog bola u gornjem levom očnjaku uz smanjenu mogućnost otvaranja usta. Terapijski plan se sastojao od konzervativno-endodntskog zbrinjavanja gornjeg levog očnjaka, i ostalih zuba frontalnog, kao i premolarnog područja gornje i donje vilice. Iako je i molarno područje bilo za zbrinjavanje, to je usled napredovanja oboljenja (ankiloze TMZ) bilo nemoguće sprovesti. Zaključak. Stomatološko zbrinjavanje bolesnika sa AS-uzrokovanom ankilozom TMZ smatra se pravim izazovom za rad u svakodnevnoj stomatološkoj praksi. Stoga, trebalo bi naglasiti izuzetnu važnost postavljanja rane dijagnoze oboljenja, kao i multidisciplinarni pristup u lečenju AS bolesnika u cilju unapređenja toka i ishoda samog oboljenja.

#### Ključne reči:

spondilitis, ankilozirajući; temporomandibularni zglob; ankiloza; dijagnoza; zubi, nega i lečenje hronično obolelih; lečenje, ishod.

Correspondence to: Zlata Brkić, Clinic for Stomatology, Military Medical Academy, Crnotravska 17, 11 000 Belgrade, Serbia. E-mail: <u>zlatavanja@open.telekom.rs</u>

### Introduction

Ankylosing spondylitis (AS)/Morbus Bechterew is a chronic inflammatory rheumatoid disease <sup>1</sup>. It occurs in 0.1–0.2% of adult population, mostly in the second or third decade of life, and 3–5 times more often in males <sup>1–4</sup>. AS causes progressive synovial changes that eventually involve all of the axial joints (mainly joints in the spine/"bamboo spine" and the sacroiliac joint in the pelvis), but peripheral joint involvement may also be an important feature <sup>5–7</sup>. The disease course, although highly variable, will progress to severe disability of the temporomandibular joint (TMJ) in almost one third of AS patients <sup>1–4</sup>. TMJ disability is correlated to the severity and extension of AS <sup>1</sup>.

Involvement of the TMJ appears to present with increasing pain on eating, often associated with progressive limitation of mouth opening, stiffness and gross restriction of jaw movement, accompanied by radiographic evidence of joint degeneration (loss of joint space, osteophytes, surface erosion and ankylosis). The available literature suggests that AScaused ankylosis of the TMJ is exceptional <sup>1-3</sup>. Even though exceptional, one should think about its possibility as it is a serious and disabling condition that causes problems with mastication, swallowing, digestion, speech, appearance and poor oral hygiene with resultant rampant caries <sup>1-3</sup>. For these patients access to routine dentistry is limited and they are considered a diagnostic challenge for the dentist <sup>4</sup>. The aim of this study was to present caries management in a patient with AScaused TMJ ankylosis and to draw attention to early detection and multidisciplinary approach in the treatment of AS patients towards to the favorable disease course and outcome.

# **Case report**

A 54-year-old male patient sought medical attention at the Department of Periodontology and Oral Medicine, Clinic for Dentistry, Military Medical Academy, Belgrade, Serbia, due to pain in the area of the upper left canine in the presence of limited mouth opening.

Clinical dental examination revealed painful and limited mouth opening. His feeding was characterized by the inability to masticate food, limiting intake of liquids or semisolids. The TMJ range of motion was limited and associated with preauricular pain, while maximum unassisted interincisal mouth opening was 15 mm (Figure 1). Assisted (passive) mouth opening did not increase the interincisal distance. No clicking sounds on opening, closing and lateral movements of the TMJ were noticed. The patient had no history of trauma or infection of the TMJ. Due to the degenerative changes in TMJ, the patient had occlusal changes (protruded mandible) and anterior open bite.

The detailed medical history of the patient revealed that the patient due to AS was for 25 years under the appropriate treatment in the Institute of Rheumatology of Serbia (IRS). The IRS gave a detailed description of the patient's condition along with the laboratory findings (presence of HLA-B27 antigen) and conventional radiographic findings of spine and sacroiliac joint in the pelvis (Figures 2 and 3).



Fig. 1 – Maximum unassisted interincisal mouth opening (15 mm) on the first visit to the clinic.



Fig. 2 – Lumbar spine radiograph shows syndesmophytes bridging the vertebral bodies with the classic "bamboo spine" appearance.



Fig. 3 – Conventional radiograph of the pelvis shows bilateral sacroiliitis.

After clinical (anamnesis and physical examination) and imaging evaluations, the diagnosis was bilateral TMJ ankylosis secondary to AS.

Intraorally, teeth were severely damaged. Due to poor oral hygiene habits almost all present teeth, apart from the painful left canine, were affected by extensive multiple caries and periodontal disease (Figure 4). The treatment plan in accordance with the possibilities of opening the mouth was made. The plan consisted of the upper left canine management and rehabilitation of the remaining teeth in the frontal and the premolar region in both, the upper and lower jaw. Even though molar region needed to be treated, it was



Fig. 4 - Digital panoramic radiograph of the patient before the upper left canine treatment.

not in the treatment plan because TMJ ankylosis made the treatment impossible, even for tooth extraction.

The treatment of upper left canine consisted of necrotic pulp removal by chemomechanical instrumentation of the root canal. Mechanical treatment was carried out by a hand NiTi file with the passive step-back technique. Root canal drying was done with paper points and it was temporary filled, with aqueous suspension Ca(OH)<sub>2</sub> with pH value 12 (Calcipro, VOCO Germany); after that, the tooth was closed with temporary cement (Provis, Favodent, Austria). Control exam was scheduled in two weeks. After two weeks, since patient had no clinical and subjective symptoms, the definitive root canal filling was done with the paste based on ZOE (Endomethasone N, Septodont, France) and gutta-percha points (Beutelrock, VdW dental, Germany) using cold lateral compaction of gutta-percha (Figure 5).



Fig. 5 – Digital panoramic radiograph of the patient after upper left canine treatment.

For all teeth in the frontal region of the upper and lower jaw, calculus deposits were removed by an ultrasonic scaler.

#### Discussion

AS is a chronic idiopathic rheumatic disease of multifactorial etiology, associated with the combination of

infection (such as *Klebsiella*), autoimmunity and strong genetic factors <sup>1-3</sup>. AS is diagnosed through a combination of clinical history, examination, radiological imaging and determination of HLA genotype (the antigen HLA-B27 in the blood is present in more than 90% of patients with AS). To date, the diagnosis of AS is usually made with an average 5-10 years of delay <sup>1-5</sup>.

The available literature suggests that TMJ involvement is found in 4–35% of AS patients, usually at a relatively late stage, after 10–30 years of presence <sup>5, 6</sup>. Therefore, evaluating TMJ function is strongly recommended in patients who have the long history of AS.

Having all this in mind, it is obvious that the accent should be put on the early diagnosis of AS. For the early AS diagnosis we should be informed about age of patients, symptoms and signs of initial or early manifestation, the significance of laboratory findings and radiological changes, especially on sacroiliac joints. When the early diagnosis of AS is set, the clinician should be suspicious of possible TMJ involvement and make a routine clinical examination in order to detect the condition and possibly present rare complications of TMJ. If involved, clinical findings in the TMJ are pain, swelling, movement impairment, crepitation and malocclusion of the teeth (in advanced stages)<sup>7</sup>.

#### Conclusion

The early diagnosis of AS will give a chance to the reumatologist to make a multidisciplinary approach by team work (such as maxillofacial surgeon, dentist, psychiatrist, physiotherapist, speach therapist...) towards the most favorable disease course and outcome. The patient should be adviced to accept recommendations concerning his behaviour regarding, among others, dental health (periodical dental health examinations) before the TMJ ankylosis occur. The patient should know that all the teeth should be rehabilitated, not extracted. Even if TMJ ankylosis occurs in later stages, the patient can have a surgical treatment of ankylosis, and in that case all his teeth will have an important role in the future coordination of mandibular movement after the operation.

# REFERENCES

- 1. Wenneberg B, Kopp S. Subjective symptoms from the stomatognathic system in ankylosing spondylitis. Acta Odontol Scand 1982; 40(4): 215–22.
- Arora P, Amarnath J, Ravindra SV, Rallan M. Temporomandibular joint involvement in ankylosing spondylitis. BMJ Case Rep 2013; 2013. pii: bcr2013009386.
- 3. Locher MC, Felder M, Sailer HF. Involvement of the temporomandibular joints in ankylosing spondylitis (Bechterew's disease). J Craniofac Surg 1996; 2013: 205–13.
- Dachowski MT, Dolan EA, Angelillo JC. Ankylosing spondylitis associated with temporomandibular joint ankylosis: Report of a case. J Craniomandib Disord 1990; 4(1): 52–7.
- 5. *Mastrović Z.* Ankylosing spondylitis as a social and medical problem. Reumatizam 1975; 22(5): 169–76.
- Mau W, Zeidler H, Mau R, Majewski A, Freyschmidt J, Deicher H. Outcome of possible ankylosing spondylitis in a 10 years' follow-up study. Clin Rheumatol 1983; 22(Suppl 2): 137–43.
- Vibhute PJ, Bhola N, Borle RM. TMJ Ankylosis: Multidisciplinary Approach of Treatment for Dentofacial Enhancement: A Case Report. Case Rep Dent 2011; 2011: 187580.

Received on May 22, 2015. Revised on November 16, 2015. Accepted on November 25, 2015. Online First September, 2016