ACTA FAC MED NAISS

UDC 616.3:616.71

Case report

ACTA FAC MED NAISS 2008; 25 (2): 103-105

Milan Knezevic[1], Mario Vicente-Barrero[1]
Sergio Dominguez[1], Francisco Garcia Jimenez[1]
Juan Luis Alfonso Martin[2], Sladjana Petrovic[3]
Dragan Petrovic[4]
[1]Faculty of Medical and Health Sciences. Stomatology, Oral and Maxillofacial Surgery Department. University Hospital Insular, Las Palmas, Spain
[2]Anatomical Pathology, University Hospital Insular, Las Palmas, Spain
[3] Institute of Radiology, Clinical Centre Nis Faculty of Medicine, University of Nis, Serbia
[4] Dentistry Clinic, Faculty of Medicine University of Nis, Serbia

NEURALGIFORM OROFACIAL PAIN PRODUCED BY OSTEONECROSIS OF THE JAWS: A CASE REPORT

SUMMARY

The paper presents a case of osteonecrosis of the jaw bones with severe orofacial pain as the main symptom. The mechanism of osteonecrosis development, as well as its evolution before and after the pain is discussed, too. Suggestions for the treatment in the post-neuralgiform period are given in the text. A brief overview of the literature is presented in discussion in this article.

Key words: **osteonecrosis of jaw bones, neurogenic orofacial pain**

INTRODUCTION

In the decade from 1970s to 1980s, neurogenic pain in the jaws was associated with osteonecrosis. The term already suggested, alveolar cavitary osteopathy, soon after related to the term of its sequel: neuralgia induced by cavitary osteonecrosis (NICO: neuralgia inducing cavitational osteonecrosis).

Defined as the syndrome of chronic neurogenous orofacial pain, induced by necrosis of the bone of alveolar maxillary processus, without pus and with intraosseous cavitations of the diameter not clearly detectable by conventional radiography (1).

Case report

The patient, 43 years old, was referred to our clinic because of severe, neuralgiform pain in the mandibular region (in the edentulous zone of premolar and first molar on the right). The pain was described as paroxysmal, very intense, in intervals lasting more than 10 hours. However, he had a constant pain that lasted for several days, not very severe but persistent. Conventional radiographic assays were performed (periapical-retroalveolar radiogram and orthopantomography), which delineated mild radiolucency of the region, without any significant bone destruction *(Figure 1)*.

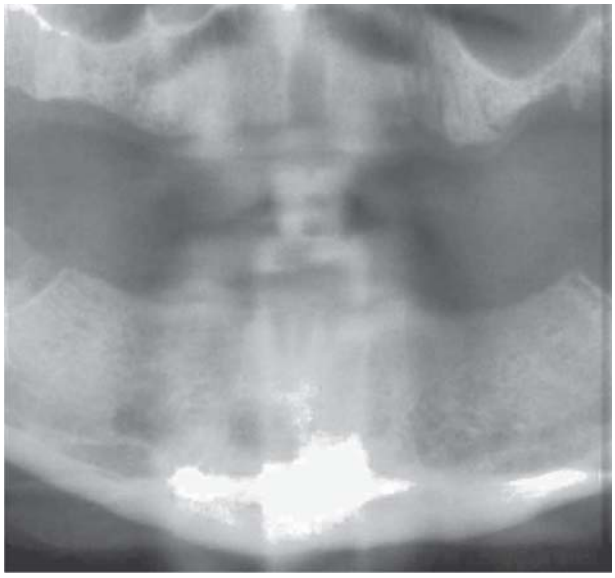

Figure 1: Radiograph of the lesion presenting mild osteolysis without significant bone destruction

Nevertheless, our decision was to perform surgical revision – curettage of the lesion – and the

Corresponding author. Dragan Petrovic • Phone: +381 63 482028 • E-mail: petrovicini@sbb.co.yu

material obtained was sent for pathohistologic analysis, which confirmed the elements of chronic-type osteonecrosis *(Figure 2)*.

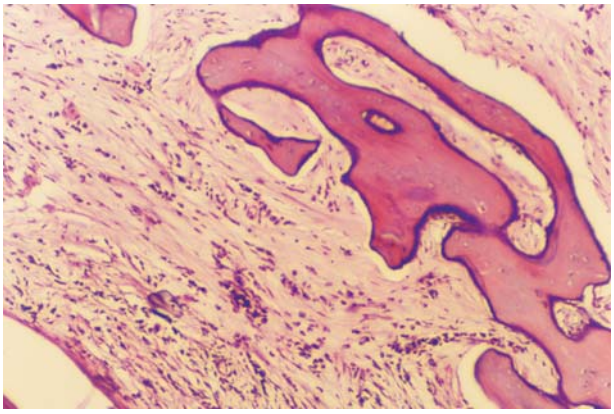

Figure 2: Histopathological finding indicating the process of chronic osteoradionecrosis

DISCUSSION

Two possible mechanisms of NICO pathogenesis could be supposed and argued based on the evidence: infectious and ischemic. The infection theory claims that the formation of intraosseous cavities is related to the presence of chronic periapical processes in the jaw bone before and after tooth extraction. However, many authors agree that this theory is not based on clear evidence (2, 3).

The theory of ischemia states that pain in NICO could be attributed to the changes in intraosseous fluid dynamics due to ischemia and inflammatory mediators, rather than to the changes because of local nerve injury.

In the study of Bouquet et al. (1), similarity of the generation of NICO after cavitary alveolar osteopathy of the jaws on one side and avascular necrosis of the femoral head (where fat necrosis and medullary fibrosis – with limited neutrophil and histiocyte numbers – are present).

Another analysis, presented by Grupo and Gluck (4, 5) comprised patients with severe orofacial pain and intraosseous cavitary osteopathy, where thrombocytophyllic alteration was assessed.

The authors concluded that thrombo-cytophyllic alteration can be a significant risk factor for cavitary alveolar osteopathy and femoral head osteonecrosis.

It has been accepted that osteonecrosis of the jaws can be the consequence of thrombosis which causes the obstruction of alveolar spaces and compromised blood flow (6, 7).

In these cases, reduced intramedullar pressure is induced, which produces hypoxia and cell death.

Toxins, immunoglobulins and inflammatory mediators, released from the sites of ischemia and necrosis, strongly stimulate nerve endings, provoking thus neuralgiform pain *(Figure 3)*.

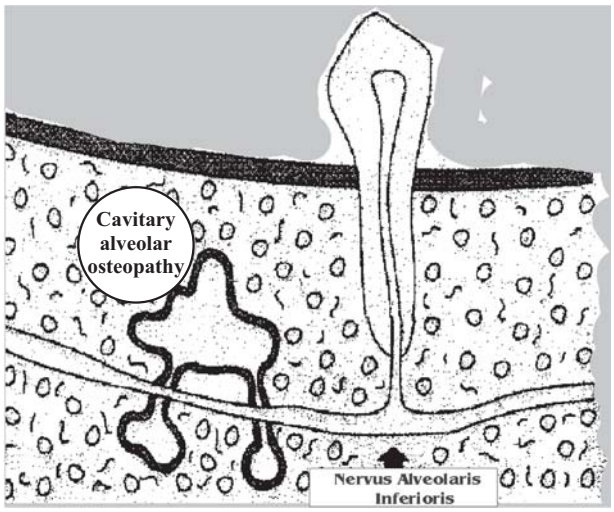

Figure 3: Graphical presentation of cavitary alveolar osteoradionecrosis which could account for the pain in the region of n. alveolaris inferiores

Published papers indicate that cavitary alveolar osteopathy is more common in women than in men. Ratner et al. (8) found female to male proportion of 4:1; Roberts et al. (9) the proportion of 2,4:1, and Bouquet et al. (1) reported the proportion of 3:1; all of them reported the age group of 40-60 years. The principal, striking symptom in patients with cavitary alveolar osteopathy is severe pain in the mouth cavity, dentoalveolar region and face as a whole. This pain was located in the innervation area of the trigeminal nerve; it was very strong, sudden and uncontrollable (paroxysmal), though, occasionally, continual (8, 10). It was located in the portion of the jaw bone where a tooth had been previously extracted. It can be bilateral and involve either maxilla or mandible.

The pain does not occur during sleep and can be completely or partially supressed by the application (infiltration) of local analgesic into the pain-involved region. Clinical exploration of peripheral parts of the cranial nerves does not reveal any changes (8-10).

Pain-involved regions, as noticed by inspection, were edentulous.

There are several possible ways to record osseous alterations in the jaws, though the methods that have been utilized up to the present offer only a partial solution: conventional radiographic study (periapical intraoral, extraoral panoramic, and lateral mandibular), computerized tomography, where minor osteolytic zones may be noticed in the bones, nuclear magnetic resonance and computerized

tomography with simplified emission of technetium 99 protons (11).

The latter assay would be a diagnosic method of choice in the diagnosis of intraosseous infarctions and ischemia-related changes, identifying, as a rule, "hot spots" which demarcate reduced vascularization zones (12).

Bouquet and Roberts (3) described radiographic findings as radiotransparent lesions similar to soap bubbles, poorly delineated, and also radio-opaque forms of cotton swabs, or the absence of normal ossification with the presence of lamina dura in the extraction zone.

The treatment consists of complete resection of appropriate bone tissue in the pain zone, identifying it under the action of local analgesia.

After the removal of the suspected bone fragment, curettage of the bone marrow is performed. Ratner et al. (8) defined the criteria of a successful treatment in view of reduced intensity and pain duration.

Of the patients treated by Ratner (8), 90% had alleviated pain, which oscillated in the period from two months to nine years.

Bouquot and Christian (13) published the results of their retrospective analysis of 242 patients treated between 1971 and 1988; the analysis was based on a questionnaire with 16 questions on pain characteristics. The pain, on average, disappeared in the period of 4,6 years (in the range from 6 months to 18 years).

REFERENCES

1. Bouquot JE, McMahon RE: Ischemic alveolar osteonecrosis in 2023 patients with chronic facial pain. J Orofacial Pain 1997;11-18.

2. Adams WR, Spolnik WR, Bouquot JE :Maxillofacial osteonecrosis in a patient with muItiple “idiopathic” facial pains a case report. J Oral Pathol Med 1999;28: 123-32

3. Bouquot JE, Roberts AM, Person P, Christian J: Neuralgia inducing Cavitional osteonecrosis (NICO) Osteoielitis in 224 jawbone samples from patients with facial neuralgia. Oral Surg 1992;73: 307-19.

4. Glueck CJ, McMahon RE. Bouquot JE : Thrombophilia, hypofibrinolysis and alveolar osteonecrosis of the jaws. Oral Surg 1996 ; 81:557-66.

5. Grupo R, Glueck CJ, McMahun RE, Buuquot JE, Becker A, Tracy T: Anticardiolipin antibodies, trombophilia, and hypofibrinolysis: Pathophysiology of osteonecrosis of the jaws. J Lab Clin Med 1996; 127: 481-9.

6. Zuniga JR: Challenging the neuralgia-inducing cavitional osteonecrosis concept. J Oral Maxillofac Surg 2000; 58:1021-8.

7. Friedmann RB: Osteoradionecrosis causes and prevention. NCI Monogr 1990; 9: 145-149.

8. Rattner EJ, Person P, Kleinman OJ, Shklar G, Sucransky SS. Jawbone cavities and trigeminal and facial neuralgias. Oral Surg 1979;48: 3-20.

9. Roberts AM, Person P, Chandran NB, Chandran NB, Hori JM, Elkins W : Further observations on dental parameters of trigeminal an atypical facial neuralgia. Oral Surg 1984; 58: 121-9.

10. Penarrocha MA, Penarrocha M, Bagan JV: Estudio clinico-evolutivo de 50 pacientes con neuralgias trigeminales tipicas. Av Odonto stomatol 1997; 13:127-35.

11. Marx RE. Osteoradionecrosis: a new concept of its Pathophysiology. J Oral Maxillofac Surg 1983; 41: 283-288.

12. Denncci OJ, Dionde RI, Chen CC, Meehan SC. SPECT bone scanning in the diagnosis of chronic idiopathic jaw pain. J Dent Res 1997;79: 237.

13. Bouquot JE, Christian: Long term eflects of jaw bone curettage on the pain of facial neuralgia. J Oral Maxillofac Surg 1995;53: 387-97.

NEUROGENI OROFACIJALNI BOL IZAZVAN OSTEONEKROZOM VILICA: PRIKAZ SLUČAJA

Milan Knežević[1], Mario Vicente-Barrero[1], Sergio Dominguez[1], Francisco Garcia Jimenez[1], Juan Luis Alfonso Martin[2], Slađana Petrović[3], Dragan Petrović[4]

[1]Fakultet medicinskih i zdravstvenih nauka, Odeljenje za stomatološku, oralnu i maksilofacijalnu hirurgiju, Univerzitetska bolnica Insular, Las Palmas, Španija
[2]Anatomska patologija, Univerzitetska bolnica Insular, Las Palmas, Španija
[3]Institut za radiologiju, Klinički centar Niš, Medicinski fakultet Univerziteta u Nišu, Srbija
[4]Stomatološka klinika, Medicinski fakultet Univerziteta u Nišu, Srbija

SAŽETAK

U ovom radu prikazujemo slučaj osteonekroze viličnih kostiju sa neurogenim orofacijalnim bolom kao glavnim simptomom. Prikazan je mehanizam nastanka osteonekroze i njena evolucija do i posle pojave bola. Predlog za lečenje u postneuralgiformnom periodu je naveden u tekstu. Kratak pregled literature zauzima deo diskusije u ovom članku.

Ključne reči: **osteonekroza vilica, neuralgiformni orofacijalni bol**