

Neuralgia-inducing cavitation osteonecrosis – Fact or myth, the debate persists

ABSTRACT

Neuralgia-inducing cavitation osteonecrosis (NICO) is a debated condition characterized by cavitory lesions in the maxillary-mandibular region, often missed on conventional radiographs, and the golden standard for diagnostic measures is bone scintigraphy. It may arise secondary to trauma, such as dental extraction and endodontic treatment, and due to a low-grade chronic infection. NICO has been documented as a frequent cause of face pain involving the trigeminal nerve divisions. It may be severe, piercing pain, of short duration or even continuous pain of moderate intensity. It affects females with a greater predilection than males. A lack of awareness of the condition among health professionals is often put into the basket of atypical facial pain. Current studies describe ischemic alveolar bone marrow coagulation disorders as the cause for NICO, which may also be the result of thrombosis with or without hypofibrinolysis, which would obstruct vascular spaces impairing blood flow in the region. Treatment is decided on a case basis, depending on the clinicians' experience, on previous treatments, on the patient's general status, and more importantly, whether the site is edentulous or dentate. If surgical intervention is chosen, tissue should be sent for pathological examination. Over the years, with the advance of imaging diagnosis processes and the study and detection of genetic changes, one may also include as a cause of NICO the decreased bone marrow blood flow causing bone cavities. All of this was also associated with genetic mutations which would predispose patients to thrombophilia and hypofibrinolysis.

Keywords: Aseptic necrosis of bone, avascular osteonecrosis, ischemic necrosis, jawbone cavities, neuralgia-inducing cavitation osteonecrosis

INTRODUCTION

Neuralgia-inducing cavitation osteonecrosis (NICO) has been mentioned in literature since 1976, and definitions range from chronic neuralgia-like syndrome to an aberrant form of atypical facial neuralgia.^[1]

Clinicians have claimed that areas of bone undergo inflammation and necrosis, thereby giving rise to neuralgia-like symptoms.

The condition may or may not present on conventional two-dimensional radiographs. It is postulated that it may arise due to low-grade chronic infection.

Also called Ratner's bone cavity, a NICO was first described in dental literature by G V Black in 1920.^[2] Later, Shankland

and Bouquot studied this condition through transmission sonography in cadavers and patients.^[3,4]

Today, there are two distinct schools of thought on NICO. Some medical and dental professionals doubt that the condition exists as a disorder and consider it a controversial diagnosis. They point to data which suggest that bone cavitation defects are found routinely in cadaver samples,

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thus doubting the theory that these defects produce neuralgic pain.^[5-7]

Some researchers and clinicians alike contend that peripheral neural damage is a major etiological factor for the development of trigeminal-like pain.

On the other hand, treatment success in clinical practice based on this theory lends it some credence.

An ultrasound bone densitometer purportedly detects and precisely images porosity of the bone and may aid

medical professionals in diagnosing bone marrow edema, NICO, osteomyelitis, and periodontal pockets. However, there is no evidence-based literature on the effectiveness of these devices published in peer-reviewed medical journals.

CASE REPORT

A 55-year-old woman visited the outpatient department of our hospital seeking relief from long-standing facial pain refractory to previous therapeutic measures over the past 8 months.

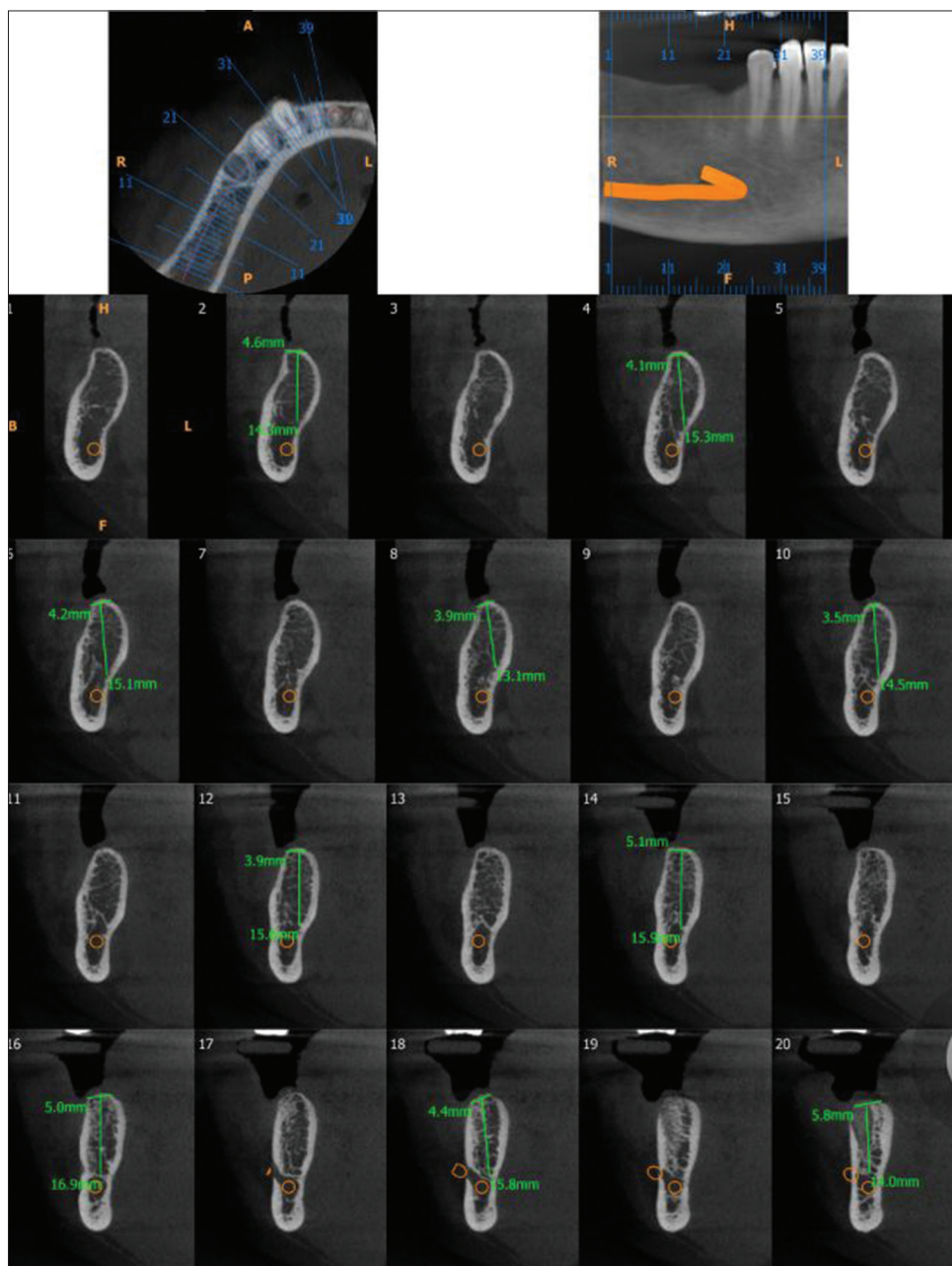


Figure 1: Cone-beam computed tomography scan cross-section view showing the bone defect

The area of concern was the mandibular right premolar region with a history of tooth extraction almost coincident with the onset of pain. A history revealed the extraction as being of a routine nonsurgical type without any incision or sutures placed.

The clinical examination revealed a healed extraction site from the soft-tissue perspective, with a slight pale-bluish hue over the tooth extracted. The site was positively tender to palpation.

Cone-beam computed tomography scans with 0.5-mm slices revealed an extraction site without osseous fill where the cortical outline of the socket could still be defined [Figure 1].

Considering the fact that she had received several therapeutic regimens by orofacial pain specialists and neurologists, she was explained the possibility of a condition called NICO generally arrived at by elimination or wastebasket diagnosis.^[8]

She consented to the plan of surgical debridement as a last resort as all other medicinal measures had not provided her any relief.

She was started on an antibiotic 24 h preoperatively (amoxicillin 500 mg with clavulanate potassium 125 mg, to be taken twice daily) for a total of 5 days.

A 30 s preoperative mouth rinse with chlorhexidine was advised before the procedure.

On the day of surgery, the site was anesthetized using lignocaine 2% with adrenaline 1:200,000 for an inferior alveolar, lingual, and long buccal nerve block.

A paracrestal incision was planned with a lingual bias so as not to close the incision on unsupported bone [Figure 2].

Mucoperiosteal flaps raised showed an area of soft necrotic bone with granulation tissue tags adherent to the periosteum.

Thorough debridement was performed and the site irrigated with sterile saline [Figures 3 and 4].

Closure was done with 5-0 polypropylene suture (Prolene, Ethicon).

The patient was reviewed on day 3 and day 7 postsurgery and was doing well with complete resolution of constant pain in the region, with the only discomfort present being from the surgical intervention.



Figure 2: Preoperative view showing a bluish tinge over the area of interest

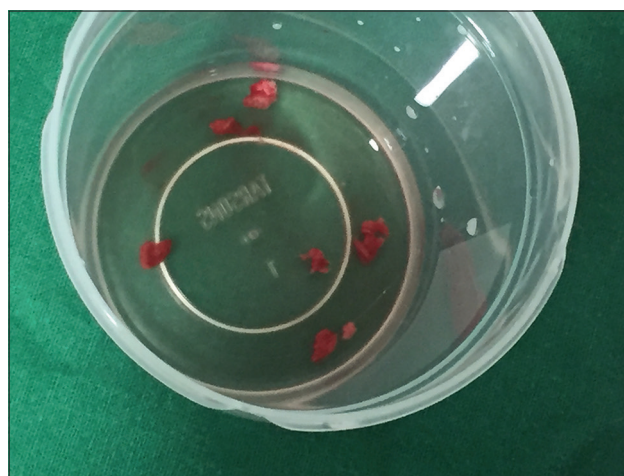


Figure 3: Specimen collected from debriding the defect



Figure 4: The region postdebridement

DISCUSSION

Ischemic osteonecrosis has been reported in orthopedic literature as not being a true bone disease. However, more a

result of systemic and local disorders or events that ultimately lead to ischemia and infarction of bone marrow, bone, or both.^[9] Clinicians have postulated that osteonecrosis can occur in the maxilla and mandible as a consequence of trauma and infection leading to NICO.

Bouquot and McMahon^[10] reported that a patient with NICO may have concomitant pulpal or periodontal infections, may have undergone tooth extractions, endodontic procedures, or periodontal surgeries, or may have experienced trauma to the facial region.

Some clinicians have used terms such as bone cavities^[11] to describe conditions associated with bone destruction without any signs of inflammation.

Bouquot *et al.*^[11] introduced the term “NICO” to describe low-grade osteomyelitis of the jaws, characterized by bone cavities and associated with orofacial neuralgia. More recently, researchers have described NICO as nonsuppurative osteomyelitis secondary to bone marrow ischemia and associated with hereditary coagulopathies, acquired coagulopathies, or both.

Several authors expanded this description to include autoimmune concepts, antibodies, thrombophilia (that is, an increased tendency to form blood clots), and hypofibrinolysis (that is, a decreased ability to dissolve clots as they form).^[6]

The overall prevalence of NICO is unknown, but it has been reported to be sexually dimorphic with a 3:1 female predilection.^[4]

CONCLUSION

This clinical case shows how a condition though debated in literature can present in the clinical scenario as being resistant to routine conservative therapeutic measures.

Surgical debridement, although considered as a last measure after all other diagnoses and therapeutic measures have been evaluated, may offer relief to patients who present with such conditions, though no such affirmative conclusion can be drawn.

A larger number of cases are required though to determine the absolute efficacy of intervention in such cases; thus, we

limit the findings of this study to the present evidence and literature available.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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