

FREQUENCY AND PATTERN OF PRESENTATION OF NEURALGIA INDUCING CAVITATIONAL OSTEONECROSIS

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ABSTRACT

Neuralgia inducing cavitation osteonecrosis is a bone disease which leads to neuralgia like pain. The aims of this study were to document the frequency and pattern of presentation of neuralgia inducing cavitation osteonecrosis and differentiate it from trigeminal neuralgia.

This case series was conducted from August 2011 to July 2012 at Punjab Dental Hospital, Lahore. Patients having facial neuralgia like pain were assessed by history, clinical & radiographic examination. From these patients, seventy patients with signs and symptoms of neuralgia inducing cavitation osteonecrosis (i.e. oro-facial neuralgic pain with intra-oral trigger zone, previous history of surgical intervention and radiographic evidence of cavitation osteonecrosis in the jaws) were included in the study for surgical debridement and curettage of necrotic bone.

Out of seventy patients, 25 were male (35.7%) and 45 were female (64.3%). Age range was of 18 to 70 years. Positive diagnosis of neuralgia inducing cavitation osteonecrosis was confirmed in 45.7% of cases. Most of the patients were from fourth and fifth decade of life. Mandibular posterior region was mostly involved (61.4%) followed by maxillary posterior region (30%). Anterior maxilla was involved (7.1%) more than anterior mandible (1.4%).

NICO most often causes sharp, shooting pain and is frequently misdiagnosed as trigeminal neuralgia. Consequently, patients are prescribed unwarranted anticonvulsant drugs and/or a variety of surgical procedures without significant pain relief. Such patients should be diagnosed properly for effective management i.e. surgical debridement of involved region.

Key Words: *Neuralgia-cavitation-osteonecrosis, mandible, maxilla, debridement, curettage.*

INTRODUCTION

Neuralgia Inducing Cavitation Osteonecrosis (NICO) is a bone disease characterized by degeneration and death of marrow and bone from a slow or abrupt decrease in marrow blood flow.¹ Its presentation varies

from asymptomatic incipient lesions to very painful cavitation lesions. The resultant bone loss is caused by a combination of poor blood supply and impaired bone healing. Prevalence rate of NICO is 1:11,000. It is more common in females. Women with age 35-60 years and teen-ager men are more affected.^{1,2}

Neuralgia inducing cavitation osteonecrosis has multi-factorial aetiology.³ Important predisposing factors include blood clotting disorders, chemo-radiotherapy for cancer, local anaesthetic injections, osteoporosis and certain autoimmune diseases. Bisphosphonates (used to prevent osteoporosis in patients undergoing cancer chemotherapy) can lead to ischemic osteonecrosis of jaw bone. There is a strong positive association between osteonecrosis and facial neuralgias.⁴⁻⁷

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Classically NICO presents as facial pain with intra-oral trigger zone in patients with previous history of tooth extraction and non-healing socket, jaw surgery, endodontic therapy or crown preparation. The pain felt may be constant or paroxysmal and is often burning and cramping, simulating trigeminal neuralgia or atypical facial pain. Initially, it may respond to anti-convulsants e.g. carbamazepine but is unresponsive to analgesics.^{8,9} It may affect any bone but with special affinity for those of face, hip and knees. The most common locations for NICO in jaws are areas of third molars, retro-molar region and maxillary tuberosity. The areas of alveolar ridge may present as non-healing extraction sites. The overlying soft tissue shows no changes. There are trigger points over involved jaw bone that produce pain when pressed or touched. Intensity of pain may or may not be related to amount of necrotic bone.^{10,11} The cavitation area is usually filled with dead bone and necrotic debris. It can be a hidden cause of neuralgia like pain. In NICO, healing process is in-effective and bone tissue breaks down faster than its repair. This leads to bone cavitation that induces neuralgia like pain.^{12,13} It is difficult to diagnose this problem as pain symptoms are often similar to other conditions e.g. atypical facial pain, trigeminal neuralgia etc. Radiographs of jaw bone may miss the cavitation and appear normal in early stages of disease but show bone cavitation in later stages.^{4,14}

Differential diagnosis of NICO includes trigeminal neuralgia, atypical facial pain, phantom toothache, causalgia, burning mouth syndrome and TMJ/myofascial pain and dysfunction syndrome.¹⁵

In NICO, hydroxyapatite crystals of jaw bone split with loss of calcium and phosphate. The original solid bone softens and leads to increased permeability for X-rays and a corresponding brightening of X-rays by loss of calcium and phosphate.¹⁶ Glueck et al hypothesized that T-786C mutation of endothelial nitric oxide synthase gene affecting nitric oxide production is associated with NICO and may open therapeutic approaches to treatment of NICO by provision of L-arginine, the amino-acid precursor of nitric oxide.¹⁷

The treatment of NICO is surgical debridement/curettage.¹⁸ The cure rate (free of pain for at least 5 years) is more than 70%. NICO has a strong tendency to recur and to develop in other jaw bone sites.¹⁹ Sciubba and Zuniga stated that NICO should be challenged because of poor evidence. Many oral and maxillofacial surgeons do not currently endorse the treatment.^{4,20,21}

There are a lot of studies about trigeminal neuralgia both in national and international literature but a

few articles about NICO in international literature. No work has been done on this condition in Pakistan. The objective of this study was to document the frequency and pattern of presentation of NICO and differentiate it from trigeminal neuralgia.

METHODOLOGY

This case series was conducted at the Department of Oral & Maxillofacial Surgery, de, Montmorency College of Dentistry / Punjab Dental Hospital, Lahore from August 2011 to July 2012.

Seventy patients presenting with complaint of neuralgia like pain with intra-oral trigger zone, previous history of tooth extraction or other surgical procedure in the jaws and radiographic evidence of jaw bone cavitation were included in the study, irrespective of age and gender, and surgical debridement was performed to confirm the diagnosis of NICO.

Patients suffering from facial pain with extra-oral trigger zone, facial pain due to some other maxillofacial problem or with some systemic disease e.g. diabetes mellitus were excluded from the study. Pre-operative assessment was done on the basis of patient's history, clinical examination and radiographs i.e. peri-apical and OPG views.

For each patient, a number of variables were recorded including demographic details, presenting complaint, site of occurrence of pain, trigger zone, edentulous area of previously extracted tooth/ teeth and radiographic evidence of jaw bone cavitation.

From each patient, written informed consent was taken and risk benefit ratio was explained. Patients with signs and symptoms of NICO were selected for surgical debridement. A follow-up of two months was given to each patient at one week, two weeks, one month and two months timings to assess relief from pain by surgical debridement/curettage.

All the data were collected and entered in SPSS version 16 and analyzed through its statistical package. Frequency distributions for all variables were worked out and results were analyzed accordingly.

RESULTS

A total of 87 patients were assessed clinically and radio-graphically and underwent surgical debridement/curettage for suspected NICO. Positive diagnosis of NICO was confirmed in 45.7% cases (n=70). Rest of

the cases did not respond to surgical debridement/ curettage. Seven-teen patients did not report for follow up and were excluded from the study.

Total number of patients included in this study were 70, with 25 males (35.7%) and 45 females (64.3%). The age range was 18-70 years (mean age of 46.06 years). Most patients were from fourth decade of life followed by fifth decade. Chief complaint of all patients was neuralgia like pain with intra-oral trigger zone. Right side was affected more than left side and mandible was more commonly involved than maxilla. In 68 patients pain was unilateral while in 2 patients pain was bilateral. Both of these two patients were with complaint in maxillary anterior region.

On radiographic examination, bone cavitations were found in 56 patients both on peri-apical and OPG views (Table 1). In most patients, cavitation area was in the form of an ill-defined radiolucency present in the area from where tooth extraction was done in the past (Fig 2, 1b).

Out of 70 patients, pain was relieved in 32 patients (45.7%) after surgical debridement. So, a positive diagnosis of NICO was established in this population (Fig 3). Among these 32 patients, pain was relieved after single surgery in 26 cases within one to two weeks of follow up. In four patients, pain was relieved after

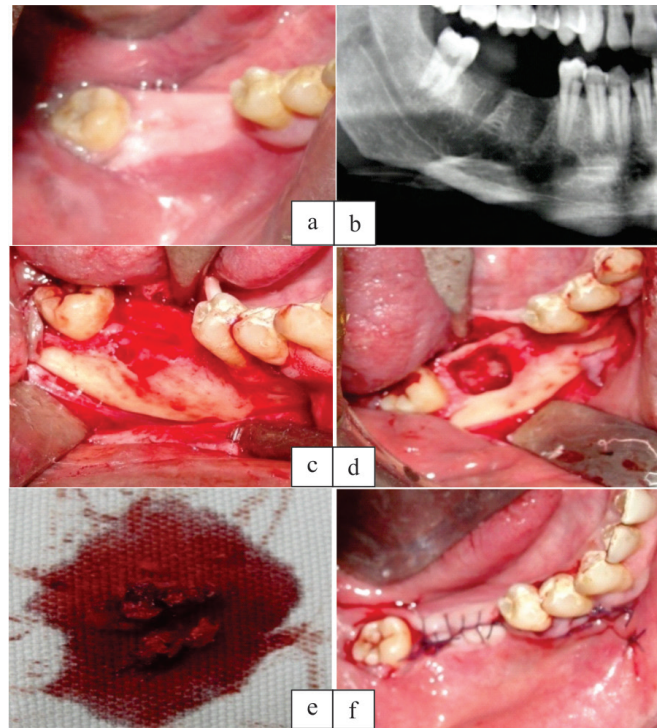


Fig 1-a: Non dentate painful area with healed soft tissue. b.Non healing extraction socket on radiograph. c.Non healing extraction site exposed. d.Bone cavitation after debridement. e.Necrotic debris/bone removed. f.Final sutured wound.

TABLE 1: RADIOGRAPHIC EVIDENCE OF BONE CAVITATION IN NICO

Cavitation	Male	Female	Total	Per-
				centage
Present	22	34	56	80.00
Absent	3	11	14	20.00
Total	25	45	70	100.00

TABLE.2: SITE DISTRIBUTION OF NICO

Site	NICO		Total
	Yes	No	
Maxillary anterior region	3	2	5
Right maxillary posterior region	5	7	12
Left maxillary posterior region	5	4	9
Mandibular anterior region	1	—	1
Right mandibular posterior region	10	17	27
Left mandibular posterior region	8	8	16
Total	32	38	70

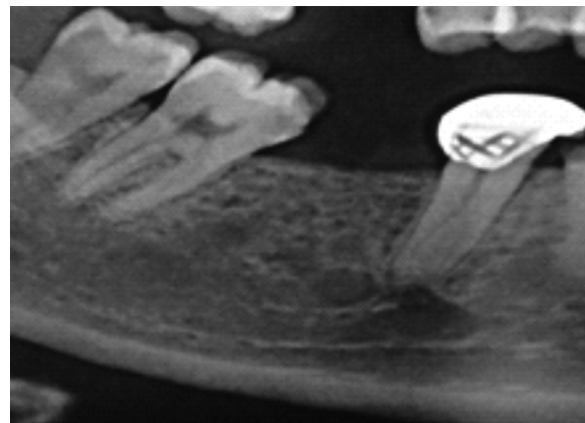


Fig 2: Radiograph showing bone cavitation in right mandibular 2nd premolar & first molar region.

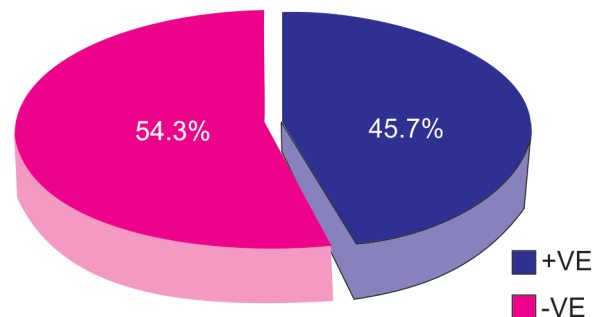


Fig 3: Frequency of NICO

one month of follow up. In two patients, pain was still present after surgical debridement but it reduced in intensity and totally disappeared after second surgery within two months of follow up. Site distribution of NICO is shown in Table 2.

In 54.3% patients (n=38) pain was not relieved after surgical debridement even up to three months of follow-up. These patients were advised medication and glycerol injection for their pain management.

DISCUSSION

NICO is a maxillofacial version of ischemic osteonecrosis with well established clinical, radiological and histopathologic criteria. However, this issue has been taken up first time in Pakistan. NICO frequently mimics trigeminal neuralgia and often causes doubt in diagnosis. Trigeminal neuralgia is slightly more common in females with slight predilection for right side and highest incidence in sixth decade of life. The problem is episodic and may go into remission for several months and returns with similar symptoms. The reported incidence is 4 per 100,000. There is severe sharp, shooting, lancinating, electric shock like pain initiated by facial movements e.g. talking, chewing, brushing, shaving or touching a specific area known as trigger point. Each pain duration lasts for seconds to minutes, often worse in the morning, and is so sharp that it makes the patient wince or flinch (*tic douloureux*).²²

NICO is a bone disease and has strong association with non-healing extraction socket, bone cavitation and previous surgical procedure while typical trigeminal neuralgia is idiopathic in nature. The pain caused by NICO is slowly progressive and may vary in intensity, location and character. This pain is supposed to be due to intra-osseous fluid dynamics and inflammatory mediators rather than damaged nerves.²³ Like trigeminal neuralgia, NICO is also more common in females with slight predilection for right side. But it is more frequent in fourth and fifth decade of life.²⁴

NICO occurs mostly in mandibular molar region and edentulous alveolar ridge areas while trigeminal neuralgia occurs along nerve distribution. It is associated with old extraction sockets and intra-oral trigger zone while trigeminal neuralgia is mostly associated with extra-oral trigger zone. The pain is also less intense than trigeminal neuralgia. Without a confirmed clinical diagnosis, aggressive and invasive procedures are not warranted. The general dental surgeons must have knowledge to diagnose such pain conditions or

should refer such patients to oral and maxillofacial surgeon for appropriate therapy.²⁵

In OPD of Punjab Dental Hospital, almost 10-12 patients of neuralgia like pain are seen weekly which are treated by anti-convulsant drugs or surgical procedures including glycerol injection and peripheral neurectomy. The observed frequency of patients with neuralgic pain is high and raises a suspicion about diagnosis. Therefore, it is vital to accurately diagnose patients with neuralgic pain and differentiate neuralgia inducing cavitation osteonecrosis from trigeminal neuralgia. In this way, patients can be saved from un-necessary medications and invasive surgical procedures and thus be treated by simple curettage.

Women have lower pain threshold and tolerance than men. In this study, there was more female predilection, which was similar to the findings encountered in other studies.^{26,27} The more frequently involved population in the present study was from fourth, fifth and sixth decade which was also similar to the findings in other studies.²⁷

In the current study, pain was relieved in 45.7% (n=32) patients after surgical debridement. In 54.3% patients (n=38) pain was not relieved after surgical debridement even up to two months of follow up period. So, the effectiveness of surgical debridement in alleviating NICO pain was less in the present study than in international studies.^{27,28} This could be due to following reasons:

1. There are no standard diagnostic criteria for NICO and its differentiation from trigeminal neuralgia.
2. There is variable response to pain and patients may confuse the signs and symptoms and may pose problem in clinical diagnosis of NICO.
3. There is a long list of predisposing factors for NICO which vary in intensity in different patients, population and regions. So, the study conducted on Western population may have different results than the studies done on Pakistani population.
4. The patient's response with regard to bone remodeling and reperfusion of curetted jaw bone area could be different depending on age, sex and systemic condition. This may also alter the final results.

The problem of NICO should be viewed in the same way as any other disorder of oral cavity. To date, no uniform diagnostic or management criteria have been

defined except surgical debridement/curettage. Considerable additional research is needed to determine the strength and type of association between NICO and neuralgia like pain. There is also need to clearly define the criteria of presentation, diagnosis and treatment of NICO.²⁹

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