ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at: www.ijdsir.com

Volume – 4, Issue – 2, April - 2021, Page No. : 179 - 184

Osteomyelitis of maxilla secondary to extraction in a patient with Paget 's disease: A diagnostic enigma

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Citation of this Article: Dr. Sunil Vasudev, Dr. Partha Pratim Debnath , Dr.Sahana M S , Dr.Jehan Koshy Jacob, Dr. Shaiqua Nooreen, "Osteomyelitis of maxilla secondary to extraction in a patient with Paget 's disease: A diagnostic enigma", IJDSIR- April - 2021, Vol. – 4, Issue - 2, P. No. 179 – 184.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

In craniofacial region, osteomyelitis rarely involves maxilla due to substantial collateral blood flow in the upper and middle third of the face, thin cortical plate with porous appearance and dearth of medullary tissue in the maxilla. When the condition involves the maxilla, it may lead to feared consequences like infection of the cranial cavity and brain. Aggressive management of the lesion should be planned to prevent the deadly complications associated with the cranial involvement. Due to the hyper-cementosis found frequently in Paget's disease patients, the brittleness of the affected bone, dental extractions usually become more difficult to perform, and traumatic extraction procedure in these patients may lead to the formation of osteomyelitis. Here in this report, we present a case of osteomyelitis of maxilla secondary to extraction in a patient with Paget's disease.

Keywords: Extraction, Maxilla, Osteomyelitis, Paget's disease

Introduction

Osteomyelitis can be defined as an inflammatory disorder of the bone, begins as an infection of the medullary cavity

followed by rapid involvement the haversian canal and spreads to involve the periosteum of the affected bone. French surgeon Edouard Chassaignac first described the lesion in the year 1852. In craniofacial region, it rarely involves maxilla due to substantial collateral blood flow in the upper and middle third of the face, thin cortical plate with porous appearance and dearth of medullary tissue in the maxilla. In current era of frequent and significant use of antibiotics, osteomyelitis of craniofacial bones became rare to be spotted. Paget's disease (PD) is a chronic disease of the none categorized by focal areas of excessive bone resorption secondary to bone formation. Sir James Paget in 1877, first described the disorder as "osteitis deformans".[1]When the condition involves the maxilla, it may lead to feared consequences like infection of the cranial cavity and brain. Aggressive management of the lesion should be planned to prevent the deadly complications associated with the cranial involvement. Osteomyelitis most commonly arises as a complication due to odontogenic infections in patients with altered immunity. The considerable predisposing factors for the disease include malnutrition, diabetes mellitus, anaemia, leukaemia, agranulocytosis, syphilis, patient with history of chemotherapy and radiotherapy. In India, 45.1% of the rural population diagnosed with diabetes are prone to develop the maxillary osteomyelitis. Most significant and common radiologic findings for the diagnosis of osteomyelitis include "moth-eaten" appearance, existence of "sequestrum"-dead bone and "involucrum"- in the radiographic examination could confirm the existence of osteomyelitis. Here in this report, we present a case of osteomyelitis of maxilla secondary to extraction in a patient with Paget's disease.

Case Report

A 69 years old male patient reported to the Department of Oral and maxillofacial surgery, with the complaint of pain in the right back teeth region of the upper jaw since last eighteen months. Patient gives the history of dysphagia and ear pain 5 years back, and he was diagnosed with Paget's disease and Eagle's Syndrome after which he was prescribed with a course of medications for fifteen days and names of the medications were not available. His past dental history revealed that he had visited a dentist fourteen months back due to pain and swelling in the right back teeth region of the upper jaw. History also revealed that the lone standing upper right third molar was extracted by the dentist. He did not have any previous radiographs that would specify the reason for extraction of the teeth. He gave a history of swelling and pus discharge from the operated site three days after the extraction procedure, after which the swelling gradually decreased in size over next seven days but did not subside fully. Four months post operatively he had a complaint of pain and swelling with foul discharge from the same region and consulted to an Ear, Nose and Throat surgeon who prescribed him a course of medication which were also not completely helpful to reduce the symptoms. Eight months later the patient reported to our department for the same. The pain was gradual in onset, dull aching and continuous in nature which aggravated while having food and relieved on taking medication (Tab-Diclofenac sodium 50 mg). Patient was only able chew on the left side due to the pain present in the right maxillary region since past six months. On extraoral examination facial asymmetry noted with respect to right side of the face. (Figure- 1,2) A diffuse swelling noted on the right side of the face, antero-posteriorly extending from the lateral border of the alar base up to the lateral canthal line and supero-inferiorly extending from the infra-orbital margin towards the corner of the mouth measuring approximately about 3x2 cm in diameter. The swelling was hard in consistency and no local raise of temperature noted,

overlying skin appeared normal, no tenderness was noted extra orally. On intraoral examination mouth opening and jaw movements were normal. Edentulous maxilla noted with wound dehiscence and necrotic bone on the right posterior maxillary alveolus extending up to the tuberosity measuring about 2x2.5 cm and yellowish white in colour, surrounded by erythematous buccal and palatal mucosa. (Figure-3) There was no pus discharge noted from the affected area.

Investigation

Cone beam computed tomography analysis presented the trabeculae with large rounded areas of sclerosis with surrounding demineralization giving a "cotton wool appearance" noted from the right cuspid region till the left tuberosity region. Areas of decreased density interspersed with radiolucency are noted and was surrounded by radiopaque borders extending from right bicuspid region till the tuberosity region suggestive of sequestrum and involucrum. Bucco-lingually it measured approximately 17.9mm, supero-inferiorly, 8.8 mm and antero-posteriorly 45 mm. (Figure-4,5,7)

CT Bone of skull vault, skull base, bilateral orbits, bilateral maxilla showed irregular cortical thickening. Multiple lucencies and sclerotic areas with thickened trabeculae were seen within the thickened bones - suggestive Paget's disease. (Figure-6)

Bone scintigraphy with Tc^{99} revealed evidence of active inflammation on the right maxilla, diffusely increased tracer update in the skull suggestive of osteomyelitis of maxilla and Paget's disease involving the skull. (Figure-4)

After the detailed clinical and radiological investigation, sequestrectomy of the necrotic bone was planned under general anaesthesia but the patient had refused to undergo the surgical procedure. (Figure-8)

Discussion

Osteomyelitis of the maxilla is found to be a rare condition considering the current era of frequent antibiotic usage, which can be linked with multiple systemic diseases like diabetes, autoimmune disorders, malignant disorders, malnutrition, and acquired immunodeficiency syndrome. The medications believed to promote the development of osteomyelitis are steroids, chemotherapeutic agents, and bisphosphonates.[3] Paget's disease also known as osteitis was described by Sir James Paget in 1877; it is a chronic skeletal condition categorized by abnormal and excessive bone remodelling bone.[4] Paget disease commonly affects the individuals more than 40 years and it is considered to be the second most common bone disease after osteoporosis.[5] Hughes and colleagues in 1994 quantified that the gene responsible for FEO is mapped to a locus on chromosome 18q21–22.[6] In the literature several authors described the susceptibility loci for familial Paget's disease can be identified by genome-wide search on chromosomes 5q35 (PDB3), 5q31 (PDB4), 2q36 (PDB5), 10p13 (PDB6) and 18q23 (PDB7).[7,8]

Roodman and Windle concluded in their study that, increased sensitivity of bone marrow and circulating osteoclast precursors to factors known to promote bone resorption such as 1,25 dihydroxy vitamin D and receptor activator of NF-kB ligand (RANKL); which in turn cause Increased IL-6 expression and signalling may lead to increased osteoclastic activity.[9,10]

Rees in 1847 first described the osteomyelitis of maxilla.[11] Bacterial inoculation causes altered pH and changes the capillary permeability which leads to localised oedema, release of cytokines, breakdown of tissues, leukocyte recruitment, reduced O_2 tension, increased localised pressure, thrombosis of vessels, and deterioration of bone. Once the infection enters into the medullary cavity,

increased medullary pressure causes extension into the cortex by Haversian and Volkmann canals leading periosteal stripping and loss of periosteal blood supply and resultant resorption and necrosis of bone.[12] Characteristically maxillary osteomyelitis is a microbial infection, caused by various gram-positive and gram-negative microorganisms, like Staphylococcus aureus, ,Streptococci, epidermidis, Pepto coccus, Pepto streptococci, Haemolytic streptococci, Pneumococci, Escherichia coli and Bacteroides. In literature maxillary osteomyelitis secondary to invasive fungal infection by Mucor mycosis also have been reported.[13] Diabetes mellitus is considered as an immunosuppressive factor, which causes arteritis of small vessels and altered vascularity leading to reduced O2 supply to the healing site, decreased host response to infection. HbA1C of our patient indicated a poor glycemic control. The pathological changes seen in the bone of the jaws in Paget's disease form the foundation for the complications that rarely occur subsequent to other maxillofacial surgical procedures. Due to the hyper-cementosis found frequently in Paget's disease patients, the brittleness of the affected bone, dental extractions usually become more difficult to perform. While performing the extraction procedure of these teeth presents with the possibilities of traumatizing the alveolar bone and fracturing the jaw. Due to high vascularity of the affected jaw uncontrolled bleeding may be encountered after the extraction procedure. Involved maxilla and mandible by Paget's disease are considered to be more prone in developing infection by the microorganisms found in the normal oral flora. Due to the slow healing process of the extraction socket, significant number of the affected patients develop osteomyelitis secondary to local infection of the socket. With the high incidence of complications author recommends to perform oral prophylaxis prior to the extraction, so that the localized contamination of the extraction socket can be prevented. Atraumatic extraction under antibiotic coverage is highly recommended. After performing the extraction bony spicules should be removed and achieving the primary closure should be considered as the outmost important to prevent the delayed healing. While planning the surgical procedure for these patients, potential insult to the surrounding tissues must be considered adjunct to the therapeutic manoeuvre. When the condition involves the maxilla, it may lead to feared consequences like infection of the cranial cavity and brain. Aggressive management of the lesion should be planned to prevent the deadly complications associated with the cranial involvement.

Conclusion

In this case report, we present a case of osteomyelitis of maxilla secondary to extraction in a patient previously diagnosed with Paget's disease and author concludes that, the atraumatic extraction under antibiotic coverage is highly recommended to prevent the occurrence of osteomyelitis and aggressive management of the lesion involving maxilla should be planned to prevent the fatal complications associated with the cranial involvement.

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Legend Figure

Figure1: Profile

Figure 2: Bird's Eye View

Figure 3: Intra Oral Picture Showing The Affected Region

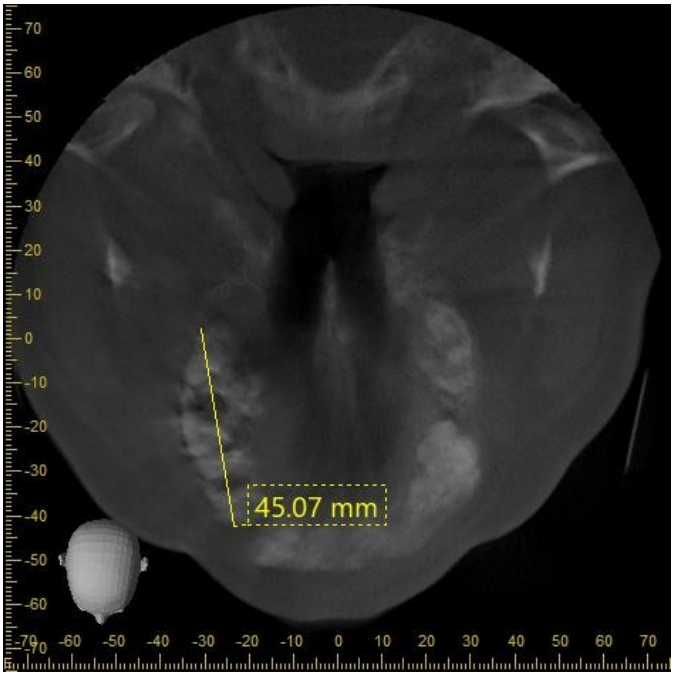

Figure 4: CBCT Axial View

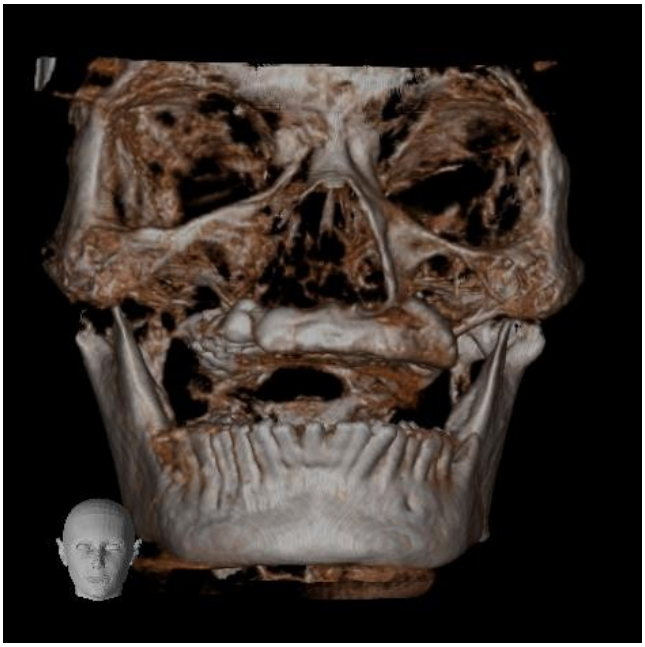

Figure 5: CBCT 3D reconstruction

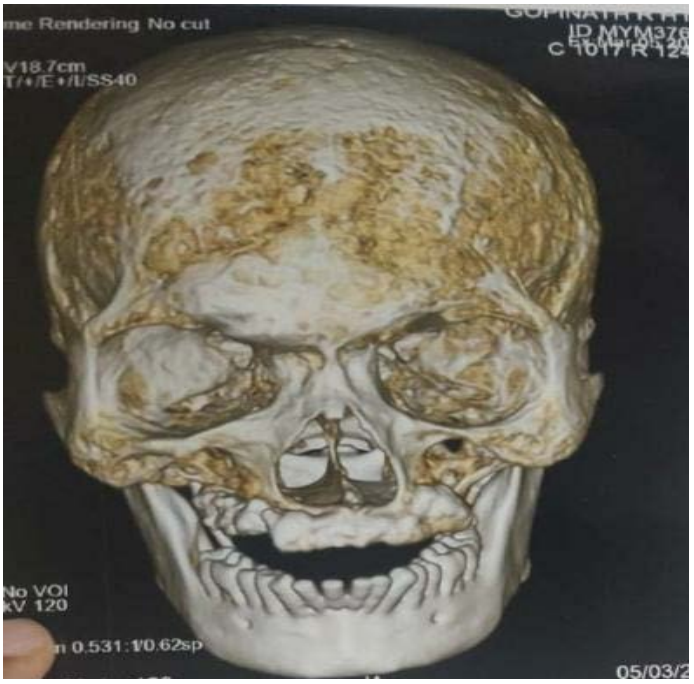

Figure 6: CT 3D Reconstruction

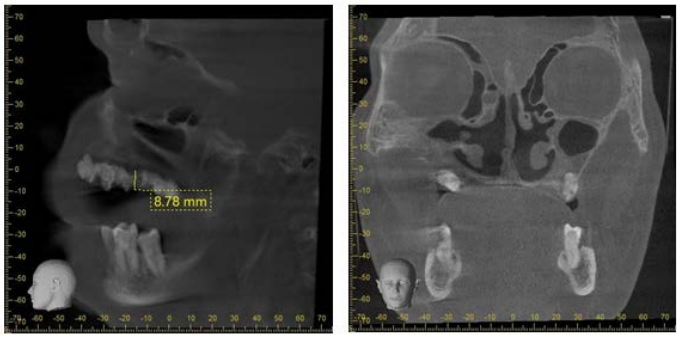

Figure 7: CBCT Lateral And Coronal View

Figure 8: Bone Scintygraphy