Research Journal of Ear Nose and Throat
Extended Abstract
2019
Vol. 3, Iss. 1

ENT Congress 2019 : Enlarging palatal defect secondary to invasive microbial infection vs. Bisphosphonate-related maxillary osteonecrosis in a 60-year-old guamanian male patient: a diagnostic challenge- William L. Lim, MD -St. Luke's Medical Center

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INTRODUCTION: Osteonecrosis is a common disorder that is a result of collapse of architecture of the bone, determining severe anatomic alterations of the involved site.Osteonecrosis is not a primary disease entity, but rather is a final common pathway secondary to a number of conditions ultimately leading to impairment of blood supply to the bony tissue or disturbance in bone remodeling which may potentially lead to bone death. This ischemic bone disease has multifactorial etiologies such as viral, mycotic or bacterial infections, radiotherapy, cocaine abuse, immunologic diseases such as Wegener's granulomatosis and malignancies. However, recent studies prove that most cases of osteonecrosis were iatrogenic.The common sites of osteonecrosis include subarticular avascular necrosis of the femoral head, as well as osteonecrosis of the mandible or maxilla. It is of utmost importance to identify all possible etiologies rapidly to manage this possibly mutilating disease.

Bisphosphonates are generally prescribed in the prevention and treatment of resorptive bone diseases such as osteoporosis and bone metastasis associated with breast and prostate cancers. They are also recognized as an effective therapy for Paget's disease and other conditions that precipitate bone fragility, such as chronic renal disease in patients undergoing hemodialysis that may have renal osteodystrophy. These acts specifically on osteoclasts, thereby maintaining bone density and strength. Bisphosphonates act on both osteoblast and osteoclasts by promoting proliferation and differentiation of human osteoblast-like cells and inhibiting osteoclasts.

Osteonecrosis of the maxilla in patients treated with bisphosphonates is a relatively rare but well-known complication which has shown an increasing interest by dental practitioners and maxillofacial surgeons.3 It is defined as an area of exposed bone in the maxillofacial region that does not heal within 8 weeks in a patient receiving bisphosphonate medication and has not had any history of radiation to the head and neck region. Since the primary mechanism of BPs is to inhibit osteoclast function by different mechanisms, it is not surprising that the altered bone remodeling is the leading hypothesis for bisphosphonate-related osteonecrosis of the jaw development. Bacterial contamination with Actinomyces and Staphylococcus may also play a role in maintaining osteomyelitic wounds and since maxillofacial bone tissue containing bisphosphonates will resorb slowly, it is understandable that contaminated bone cannot be removed fast enough to prevent the development of chronic osteomyelitis.

We present a case of a 60-year old patient, known to have diabetes and end-stage renal disease on hemodialysis and bisphosphonate therapy, with an aggressive course of palatal necrosis despite intensive diagnostic evaluation, multidrug treatment and surgical management.

ABSTRACT: Maxillary osteonecrosis presents a diagnostic challenge due to the multifactorial nature of this pathology which can be easily missed without careful history taking and proper diagnosis. Essential diagnostic imaging includes panoramic X-ray, plain and contrast CT scans, and MRI. These help in differentiating BON from other conditions with similar pathology such as osteomyelitis, osteonecrosis, metastasis, or osteosarcoma. Microbiologic and histopathologic examinations are also important in direct-

Note: This work is partly presented at "4th World Congress on Ear, Nose and Throat Disorders" on October 14-15, 2019 Singapore

ing the proper management of bone osteonecrosis.1 The standard medical treatment for this disease include Penicillin G IV for 2-6 weeks and broad- spectrum antibiotics targeting Actinomyces and other microbial organisms which can inhabit the oral cavity. Failure of treatment encourages the need to further investigate other possible cause of osteonecrosis.

CASE REPORT: This is a case of a 60-year-old male known hypertensive, diabetic, and CKD stage 5 on regular hemodialysis and bisphosphonate therapy presenting with 2 months history of mucosal ulceration of the hard palate. Patient reports frequent traumatic brushing of teeth and hard palate, and sucking on ice cubes which often results to minimal mucosal bleeding and sloughing of palatal mucosa. No reported history of pain or dysphagia. No medical consult done or medications taken.

1 month prior, patient noticed a more irregular texture of the left side of the hard palate upon contact with the tongue especially on gargling and ingestion of food with minimal bleeding. Patient at this point can still tolerate regular food without discomfort. However, during the same period, patient started to notice bitter-tasting discharge from the traumatized area of the hard palate associated with foul-smelling whitish discharge from the left nostril and frontal headache. No other associated nasal symptoms such as nasal obstruction, facial fullness, or hyposmia. No episode of fever has been reported. Patient then consulted at a local hospital, wherein he was admitted and managed as a case of necrotizing fungal sinusitis and was treated with Amphotericin B. Biopsy was done in the same institution revealing only benign squamous mucosa with granulation and ulceration. Despite complete medical treatment, patient had persistent symptoms which prompted consult in our institution for further evaluation and management.

During admission, oral cavity CT scan without contrast was done with findings suggestive of an infectious process. Blood glucose was continuously monitored and hemodialysis was continued. Flexible nasopharyngolaryngoscopy was done revealing ulcerating lesion on the left side of the hard palate suggestive of newgrowth. Biopsy of the said ulcerating lesion was performed with results leaning towards an acute or chronic inflammatory process with no evidence of malignancy. Patient was started on Coamoxiclav 1.2g IV every 12 hours post op. KOH for fungus was unremarkable while culture study showed positive result to Streptococcus salivarius only sensitive to Vancomycin. Coamoxiclav was discontinued after completion of 7 days treatment and patient was referred to Infectious Disease service. At this point, a repeat biopsy was recommended with additional microbiological studies and patient was started on Vancomycin 1.25g IV for 1 dose and was later on shifted to Tazobactam + Piperacillin 2.25g IV every 8 hours. Endoscopic sinus surgery with biopsy was done with intraoperative findings of purulent nasal discharge, friable mucosa and bone fragment. Repeat microbiological studies were negative for KOH and Mycobacterium species but revealed few growth of Klebsiella pneumoniae on culture. WBC was within normal value. Piptazobactam was shifted to Ceftriaxone 2g IV every 24 hours. Final histopath of the nasopharynx and sinonasal tissue again revealed chronic inflammatory process with microscopic characteristics suggestive of Actinomycosis. Patient was then started on Penicillin G 2 million units every 6 hours. On the 38th hospital day, a repeat flexible nasopharyngoscopy with palatal biopsy was performed revealing resolution of palatal ulceration with noted decrease in size of palatal defect but with progression of bony necrosis of the left side of the hard palate. revealed the again revealed benign results. On further history taking, patient was noted to be on bisphosphonate therapy. After review of its association with maxillary

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osteonecrosis, patient was advised to discontinue the medication resulting to mucosal healing upon repeat flexible endoscopy. Patient was then sent home with antibiotic medications and was noted to have improved healing of the soft palate on follow up.

Case Report: Several reports regarding palatal osteonecrosis have been reviewed and studied. By far, established associations with this disease are postanesthetic necrosis, nasomaxillary malignancies, embolization, and necrotizing fungal infection.5 We are presented with a case of a 60 years old male with progressive palatal defect despite series of diagnostic tests, repeated biopsy, aggressive medical and surgical treatment. Failure of resolution despite the use of standard medical treatment, broad-spectrum antibiotics and surgical debridement prompted the need to further investigate on other possible related factors. Finally, with meticulous further investigation, control of metabolic disease, and sequential monitoring, the patient eventually responded to the treatment with gradual mucosal healing of the hard palate.

Keywords:Osteonecrosis, bisphosphonate-related osteonecrosis of the jaw, microbacterial, oral cavity

CONCLUSION AND RECOMMENDATIONS: Maxillary osteonecrosis remains to be difficult to manage due to its multifactorial etiology. The usual pathogenic organisms found in the oral cavity can later on evolve into opportunistic pathogens aggravating its condition especially for patients with metabolic and immune disorders. This, added with other related factors which can be undiagnosed due to its rare association to the disease entity places a challenge in properly addressing the problem. Even with aggressive treatments and repeated diagnostic test, the prognosis remains to be poor causing debilitating consequences which can later on affect both the function and psychological state of a patient. Diagnostic imaging, microbiologic studies, biopsy and endoscopy remains to be the standard in diagnosing maxillary osteonecrosis. However, complete history taking remains to be critical in increasing the chance of successful treatment particularly in cases where standard management shows no resolution.

Results: The patient in this particular case presented with osteonecrosis of the maxilla. Patient was placed on various antibiotic treatments which include broad-spectrum antibiotics and Penicillin G IV for 6 weeks, and surgical debridement with no resolution of bone necrosis. Bisphosphonate therapy was later on discontinued resulting to healing of palatal mucosa.

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