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Case Report

Zygoma osteomyelitis: a case report

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ABSTRACT

Chronic osteomyelitis is inflammation of the bone along with the medullary cavity and Haversian system. Occurrence of chronic osteomyelitis in the zygoma is rare. This article is to report a case of 68 year old female diagnosed with left zygoma osteomyelitis. Computed tomography (CT) facial skeleton revealed features suggestive of chronic osteomyelitis. Surgical debridement was done with broad spectrum antibiotics coverage. Histopathological examination confirmed the diagnosis. This case describes atypical presentation of chronic osteomyelitis and its management. The article also focuses on the various aetiologies of chronic osteomyelitis and the factors influencing the extent of spread of the disease. Various treatment strategies with respect to the surgical management and medications used have also been elaborated. Other instances of chronic osteomyelitis localised to the head and neck region shed a light on the future of a higher quality and wholesome treatment protocol, whether it be bacterial, tubercular or fungal. Trauma induced and postoperative, as well as radiotherapy induced osteomyelitis require more of a protocol for prevention of further complications, osteomyelitis being one of highly morbid conditions affecting these sets of patients.

Keywords: Zygoma osteomyelitis, Debridement, Atypical presentation

INTRODUCTION

Chronic osteomyelitis in the zygoma is extremely rare.[1] Osteomyelitis is defined as inflammation of bone marrow that surrounds cortical bone.[2]

Routes of spread have been theorized to be exogenous, for example, traumatic or postsurgical, and, endogenous, that is hematogenous spread. Usual presentation of osteomyelitis in adults is subacute or chronic which most often ensues trauma to involved area of face.[3]

Osteomyelitis can also be caused by infection with tuberculosis; around 10%-15% of the extrapulmonary tuberculosis cases involve the head and neck region.[4] Tubercular osteomyelitis of the midface is unusual due to the ample blood supply and nature of flat cortical bones. Extrapulmonary TB in the zygoma as the primary site of infection is rarest.[5]

In immunocompromised patients, opportunistic infections such as *Cryptococcus neoformans* can be the etiological agent for osteomyelitis, more often disseminated infection through hematogenous route, as compared to the localised infection in immunocompetent patients by the direct inoculation of fungal spores at a site of the trauma.[6]

However, cryptococcal osteomyelitis has a very rare occurrence. Other causal agents of osteomyelitis are *Aspergillus* and *Candida* species.

Osteomyelitis of zygoma is a rare occurrence with incidence of 1.42%.[7]

There exists evidence of zygoma osteomyelitis resulting from spread of infection from ipsilateral maxilla and also, frontal sinus.[8]

Here is a case of 68 year old female presenting with pain over her left cheek since 1 year, not subsiding with antibiotic treatment.

CASE REPORT

A 68 year old female presented to the clinic with left cheek swelling since 2 years, associated with mild, dull aching pain. However, no history of dental pain was given. Furthermore, no fever, chills, weight loss, night sweats was reported.

Patient was previously diagnosed with type 2 diabetes mellitus, hypertension since 10 years and compliant with medication for the same.

On clinical examination

A solitary, diffuse lesion of size 4 cm by 4 cm (depth could not be ascertained) was noted over left malar region. Tenderness on palpation was present, no local rise in temperature, no redness was noted. On applying pressure, no purulent discharge was noted. Surface of lesion was irregular, with necrotic tissue, likely sequestrum, forming the majority.

Oral cavity examination revealed caries of left and right upper 2nd molars, however haletosis was absent.

No palpable masses noted upon examination of the neck.

Investigations

The patient was followed up with relevant investigations such as routine pre operative blood investigations, gycemic profile, cardiac evaluation and imaging studies. Chest x ray showed no pulmonary tuberculosis features. A coverage of broad spectrum antibiotics was administered as well.

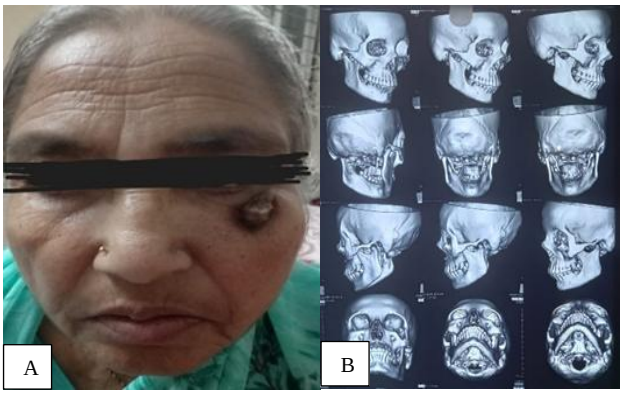

Figure 1 (A and B): Pre operative picture showing left malar swelling and CT facial skeleton (3D).

CT facial skeleton-features suggestive of chronic osteomyelitis: irregularity of cortex of left zygoma with sinus tract noted extending to skin surface.

Surgical intervention

Surgical debridement of bony sequestrum and the necrotic soft tissue surrounding the cavity with a margin of 1 cm of visibly healthy soft tissue was done, followed by curettage of the cavity created in the cortex of zygoma. Thorough irrigation of cavity was given with normal saline and betadine solution.

Stay suture placement was done with nonabsorbable Ethilon 3-0 suture material. Broad spectrum antibiotics were administered intravenous for 5 days, followed by oral route of same on discharge.

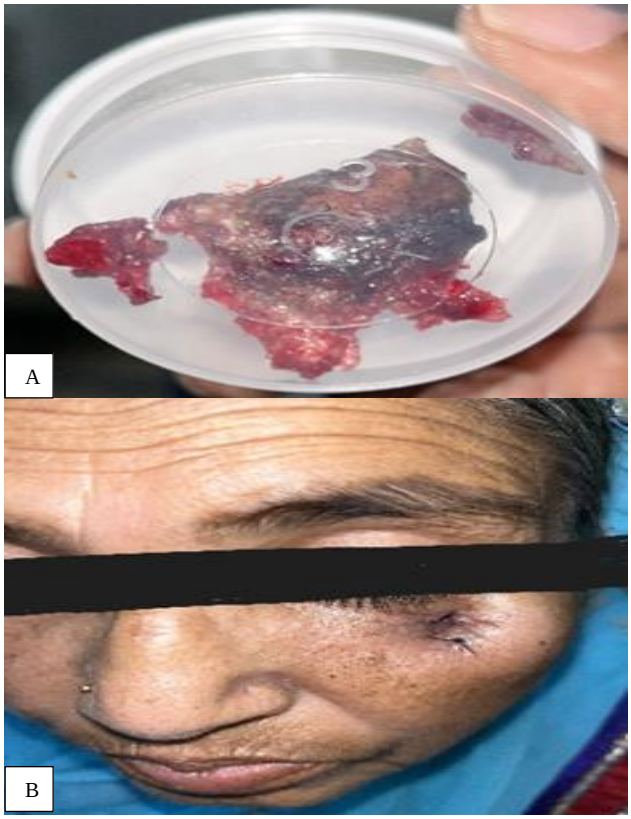

Figure 2 (A and B): Specimen excised along with bony spicule and post operative day 5.

The bony sequestrum removed and surrounding zygomatic bone debrided. Histopathological examination confirmed the diagnosis of chronic osteomyelitis. CBNAAT of the specimen was negative for *Mycobacterium tuberculosis*. The specimen was also sent for culture which showed no fungal filaments.

Patient was followed up on weekly basis and suture removal was done 1 week later. No discharge was noted and wound healing process followed a healthy and complete course.

DISCUSSION

The case highlights the non-traumatic and endogenous etiology of chronic osteomyelitis of an uncommon site of facial skeleton, that is, zygoma. Chronic osteomyelitis in this case could largely be attributed to immunocompromised host defense owing to poorly controlled type 2 diabetes mellitus and older age.

The hematogenous spread of infection is suspected, by bacteremia from distant foci of infection which could not be exclusively demarcated in this case, but postulated to be from oral or dental focus.

Pulmonary involvement was excluded by absence of typical symptoms such as productive cough, evening rise of temperature and loss of weight; laboratory tests such as chest X ray and CBNAAT were done to rule out pulmonary tuberculosis. CBNAAT of the specimen obtained was also done, in order to rule out a case of extrapulmonary tuberculosis of the zygoma.

Such an occurrence without any previous surgery or trauma to face is quite rare and hence, needs to be studied more.

Special emphasis on the treatment of dental caries and oral health in general is thus required in cases with non-traumatic etiology of chronic osteomyelitis of the facial skeleton.

This patient had a localized infection of the zygoma owing to her immunocompetence. Factors attributing to prevention of dissemination of infection were good glycemic control and timely visits to the physician.

Rai et al reported a case of maxillary osteomyelitis spreading to frontal sinus, lateral nasal wall, medial wall and floor of orbit as well as zygoma. This was managed with excision, curettage and debridement. Disease from maxillary sinus and maxilla was managed with sequestromy through a Weber Ferguson incision. A bicoronal approach was used to perform sequestromy of frontal sinus followed by adequate irrigation. Reconstruction with a temporalis myocutaneous flap was done and oroantral communication was managed with an obturator.[8]

Yadav et al reported a case of bilateral zygomatic fungal osteomyelitis which the treated successfully with sequestromy using a lateral rhinotomy bilaterally using the batwing incision and extended Dieffenbach's approach. Medical management was with liposomal amphotericin and oral antifungals on discharge.[9] This carries with it additional requirements such as monitoring electrolyte imbalance and renal function.

Hence, choice of the preparation of amphotericin and patient selection are vital aspects of fungal infections of head and neck.

Matsuki et al presented the first documented case of cryptococcal osteomyelitis involving the zygoma which they managed conservatively with oral fluconazole in an effort to preserve the facial aesthetic contributed significantly by the zygomatic bones.[5]

Bhola et al were the first to report a case of primary tuberculosis of the zygoma and illustrated the management of the patient with debridement and curettage as well as antitubercular regimen of 9-12 months. This strategy turned out to be successful with no recurrence.[6]

Borle et al contributed to the research with a case of zygomatic osteomyelitis caused by nonunion of fractured segment secondary to improper immobilization by intraosseous wiring.[1]

Osteomyelitis of head and neck can occur as complication of radiotherapy with a predilection to cervical spine with poor prognosis and significant morbidity, following high dose radiation.[10]

Postoperative chronic osteomyelitis and radiation induced chronic osteomyelitis may be prevented by better treatment protocols and precautionary measures. This may be done in order to provide strict asepsis and adequate antibiotic cover for postoperative causes, and safer delivery of radiation in radiation induced osteomyelitis.

Other treatment modalities apart from medical management as well as surgical debridement and curettage include hyperbaric oxygen therapy for refractory cases. However, disadvantages of this treatment modality such as high cost and extended time period of effective treatment hinder the wider usage.

CONCLUSION

This case of chronic osteomyelitis has shown the atypical presentation with respect to site, that is, osteomyelitis of the zygoma, of a nontraumatic and localized nature. The localized nature may be attributed to immunocompetence whereas, the nontraumatic aspect is yet to be understood completely. Hence, the most plausible etiology could be localized infection through the dental caries. Wide local excision with a margin of 1 cm followed by curettage and thorough irrigation with closure along with intravenous broad spectrum antibiotics resulted in a positive outcome with relief of symptoms, adequate clearance and no recurrence yet. A universal protocol for management of chronic osteomyelitis of head and neck is yet to be devised. This study shows a successful treatment modality and along with other research in this particular field, an optimum treatment protocol can be established.

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