

Tuberculous Osteomyelitis of the Zygoma

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Introduction:

Tuberculosis (TB) is a global disease with 32% of world population infected with the causative organism *Mycobacterium Tuberculosis*. The regions with highest incidence are the Indian subcontinent, Southeast Asia, and Africa.¹ *Mycobacterium Tuberculosis* was first isolated and described by the German bacteriologist Robert Koch in 1882.² *Mycobacterium tuberculosis* is an aerobic bacillus spread by air borne droplets. The droplets are generated by coughing sneezing or talking. After the infection is established, symptomatic individuals mostly show pulmonary manifestations of the disease. The most common expression of extrapulmonary tuberculosis is tuberculous lymphadenitis, which presents as asymptomatic enlargement of cervical or supraclavicular lymph nodes.³ Extra pulmonary tuberculosis involving head and neck is rare {excluding tuberculous lymphadenitis}. 80-90-% of patients with head and neck manifestations of TB exhibit no signs of pulmonary disease.⁴ Tuberculous osteomyelitis has been reported to involve the mandible.⁵ Zygomatic bone Osteomyelitis also is an extremely rare occurrence. Adekeye et al published a review of 4 cases of Osteomyelitis of the jaws and reported the incidence of malar bone Osteomyelitis to be only 1.42%.⁶ A rare case of tuberculous osteomyelitis of the zygoma is reported in this article.

ABSTRACT:

Extra pulmonary tuberculosis of midfacial bones is extremely rare. Diagnosis and treatment of tuberculosis can be a diagnostic challenge, especially the extra pulmonary Tuberculosis of head and neck region because of the rare incidence and lack of symptoms. This article presents a rare case of tuberculosis presenting as a space infection and subsequent investigation confirming osteomyelitis of the zygoma. The diagnosis of tuberculosis was confirmed by FNAC of supraclavicular lymph nodes that developed only after the establishment of osteomyelitis of zygoma. The case illustrates the tricky nature of the disease and the rare development of osteomyelitis of zygoma.

Key words: Tuberculosis, Osteomyelitis, Zygoma

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Case report:

A 29 year old reported to the Department of Oral and Maxillofacial Surgery, Army College of Dental Sciences (ACDS) with complaints of fever and gradually increasing swelling on the right side of face for one and half months. He had limited mouth opening. Patient gave a history of fever prior to development of swelling. He was treated by local physiotherapy and heat application for the resolution of swelling but was not helpful. He was prescribed with ciprofloxacin, erythromycin, cefotaxime and amikacin by various practitioners but with no signs of resolution of the swelling. The patient reported to ACDS when there were no signs of improvement. No history of trauma or previous infection was reported by the patient. Routine blood investigations were normal. The hemoglobin was 12.9 g/dl, total leukocyte count (TLC) was 10,100 cells/mm³ and ELISA test was negative. The erythrocyte sedimentation rate (ESR) was 32 mm/hr at the 1st hour and increased to 58 mm/hr at the 2nd hour. Sputum for AFB was negative. Radiological investigation was done. A submentovertex view revealed an ill defined osteolytic lesion in the right Zygomatic bone. An ultrasound, CT scan and FNAC was done. Ultrasound showed 6-7mm of collection over right zygomatic prominence. FNAC of the facial swelling was suggestive of nonspecific abscess. Microscopic examination of cellular smear showed numerous degenerated polymorphs with few macrophages and lymphocytes in a hemorrhagic background. No evidence of granuloma or atypia was seen. CT scan suggested an abscess overlying the right zygoma with probable Osteomyelitis (Fig 1 and 2). On clinical examination, a 4x 4cm fluctuant swelling beneath the right zygomatic arch and in front of the parotid was noted. Intraoral examination and panoramic radiograph revealed no intraoral source of infection. Aspiration was done intraorally and 1ml fluid was aspirated. Under local anesthesia the abscess was drained and pus was sent for culture and sensitivity. Patient was prescribed Metronidazole, analgesics and a mouth wash. Culture and Sensitivity showed no growth. Gram stain was positive for cocci in clusters. Incision and drainage was repeated as there was residual swelling. Culture and sensitivity showed no growth and Gram stain showed cocci in chains. On the 5th day patient complained of discharge from the mouth. On examination discharge was seen in the region of lower right third molar along with pericoronal inflammatory changes. Lower right third molar and

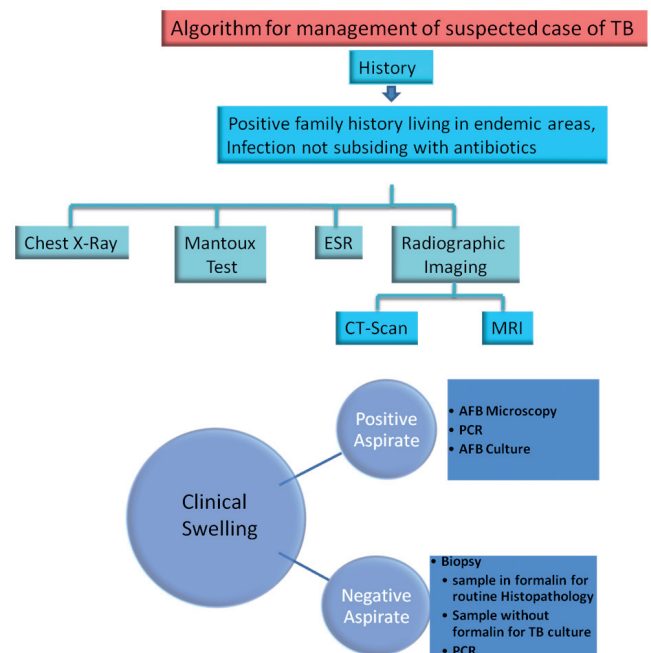
upper right third molar were extracted and exploration of the pterygomandibular and submassetric spaces was done to drain the spaces. Patient was prescribed with oral Augmentin 625 mg TDS for a week. Postoperative trismus was managed by physiotherapy. The mouth opening had improved with resolution of swelling, fever and headache. After 10 days, the patient returned with a complaint of salty taste. A CT scan was done for the second time. The CT scan revealed osteomyelitis of the right zygomatic arch and anterior wall of the maxillary sinus, with inflammatory changes in the masticator space (fig.3-5). The aspirate from the lesion was sent for PCR to rule out tuberculosis for which the result was negative. Chest radiograph was unremarkable. All medications were stopped and Patient kept under review. Subsequently, he developed a supraclavicular swelling after intervening period of 3 weeks. FNAC of the swelling showed many epithelioid granulomas in a necroinflammatory and caseous background showing polymorphs, histiocytes and few lymph plasmacytes. Special stain for AFB [Fig 6] showed fast staining bacilli ultimately confirming the diagnosis of tuberculous lymphadenitis (cold abscess). The patient was referred to a pulmonologist for medical treatment of tuberculosis and was put on a standard regimen of anti tubercular drugs (Isoniazid, Rifampicin, Ethambutol and Pyrazinamide for 2 months and Isoniazid and rifampicin for 4 months). Patient reported improved symptoms and weight gain after beginning the treatment. At 1 year follow up a post operative CT scan was done showing marked signs of improvement in the Osteomyelitis of Zygomatic bone [Fig 7, 8].

Discussion:

Tuberculosis is usually classified as pulmonary or extra pulmonary. Tuberculosis of the bones and joints is responsible for about 10% of extra pulmonary cases.³ Tuberculous osteomyelitis of the zygoma is extremely rare,^{8,9,10} and presents as a swelling or discharging sinus over the zygoma.^{8,9,11} As compared to the above, Osteomyelitis of the mandible is a common condition with established causes and treatment, whereas that of the middle third of the face is rare and commonly arises from odontogenic sources.⁷ The key to the diagnosis of tuberculosis is a high index of suspicion. Diagnosis of tuberculosis is by AFB microscopy of expectorated sputum or tissue sections, mycobacterial culture or detection and amplification of DNA {real time PCR}.³

Diagnosis of extra pulmonary tuberculosis is difficult and requires persistence in sending samples for examination.¹² In this case report a 29 year old male patient was treated for antibioma on the right maxillary right Zygomatic region by various practitioners for a period of 2 months. Although tuberculosis was considered in the differential diagnosis, and investigated by chest x ray, pus culture, FNAC, CT and real time PCR, the diagnosis was only confirmed when there was cervical lymphadenopathy. FNAC of the right cervical lymph nodes and special staining for AFB was the main confirmative diagnostic aids for tuberculosis. The presentation is usually confusing at times. Raised ESR, positive mantoux test and histopathology are the main key diagnostic aids, but at times all these may be negative thus creating a doubt in the provisional diagnosis. The most common radiological presentation is a lytic lesion [13]. In our case there was radiolytic lesion in the submento vertex view. Ultrasound confirmed 6-7cc of collection over the right Zygomatic prominence. Taking into the consideration of various diagnostic results a doubt was created within the mind whether the patient had to be treated for an antibioma or a nonspecific abscess. Moreover extrapulmonary tuberculosis involves inaccessible sites where in it is difficult to ascertain the extent of damage caused by the bacilli. In our case initially the swelling showed no positive clinical and histological symptoms for tuberculosis, but when the abscess was aspirated intraorally followed by the extraction of teeth a non healing intraoral sinus developed with right cervical lymphadenopathy. FNAC and AFB staining confirmed the diagnosis of Tuberculosis. The recent advances that have been developed for diagnosing tuberculosis are rapid culturing techniques and nucleic acid amplification tests. Rapid culturing techniques are based on radioactivity (BACTEC 460-TB), fluorescence (BACTEC MGIT 960), phage based tests and inverted microscopy leading to faster culture and sensitive reports¹⁴ of the aspirated sample. The nucleic acid amplification test has been limited to nonrespiratory specimens.¹⁵ These tests are valuable diagnostic aids when all the other tests are negative. In our case these tests might have confirmed the diagnosis of tuberculosis at initial stages itself but because of the high cost, limited availability of these tests in India and seeing the economical status of the patient it was not performed in this patient. Also it would have saved a lot of time and resources and earlier initiation of ATT if

the pus from abscess was subjected to AFB staining and culture. Treatment of tuberculous osteomyelitis is mainly medical. This case report illustrates the problems encountered in diagnosing extra pulmonary tuberculosis. Also it cannot be ruled out that intraoral surgical intervention lead to formation of an intraoral sinus and cervical lymphadenopathy which helped to arrive at a favorable diagnosis. An algorithm for management of tuberculosis is shown below.



Conclusion:

Tuberculous Osteomyelitis of zygoma is a rare condition. The nature of presentation of the disease with negative results for most of the laboratory and radiologic tests creates a doubt in the mind of the treating practitioner to arrive at a provisional diagnosis. At times it can be noticed that when the lesion is disturbed it might start reacting to the intervention and show some specific signs of the disease which can aid in the confirmative diagnosis as was observed in our case. This doesn't mean that all the cases have to be intervened in such a fashion; it is coincidental in this case that we could arrive at the diagnosis with a minor surgery that was performed in the patient. Most of the times tuberculosis is treated by chemotherapy with surgery being required. Early suspicion and appropriate diagnostic workup is imperative in diagnosing and treating extrapulmonary Tuberculosis.

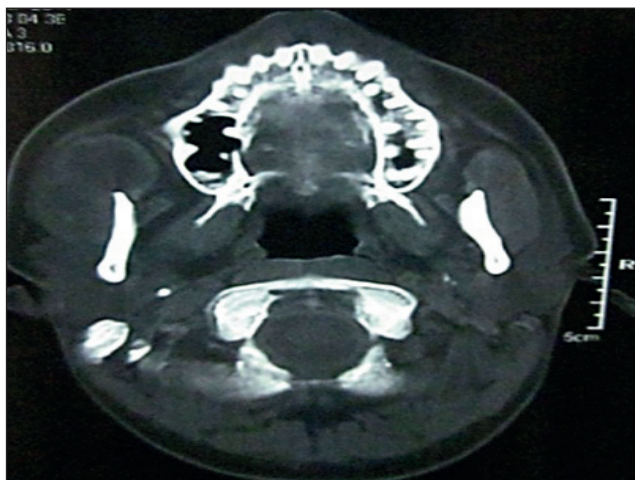


Fig. 1: CT scan showing well localized abscess lateral to the ramus of mandible, beneath the right zygoma.

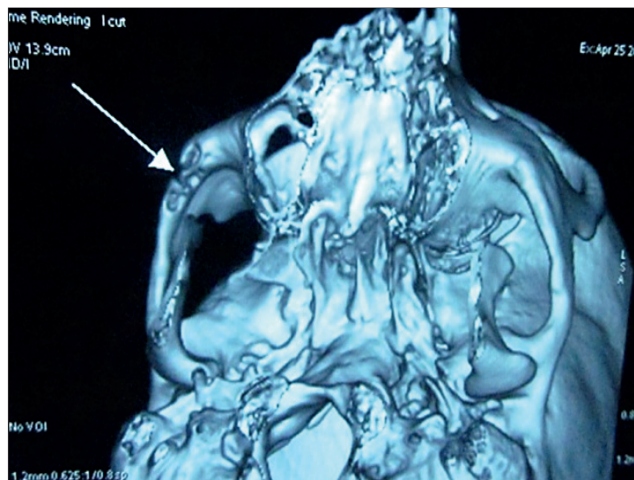


Fig. 4: 3D CT scan showing the inferior view of the right zygomatic arch.



Fig. 2: CT showing erosion of the right zygoma.

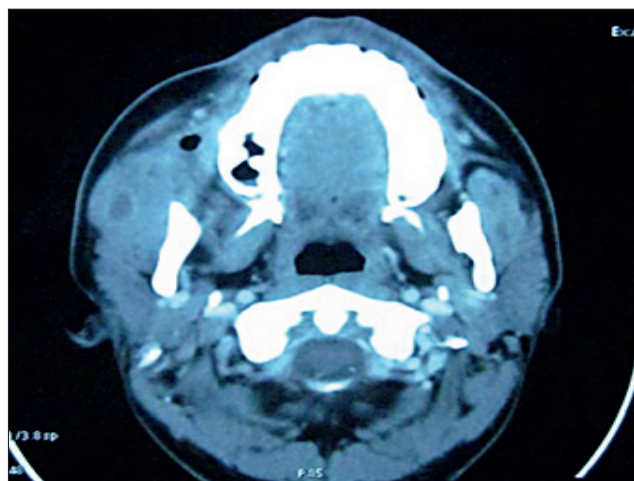


Fig. 5: 3D CT scan showing an abscess within the right masseter muscle and inflammatory changes in the surrounding masticator space



Fig. 3: 3D CT showing osteomyelitis of the right zygomatic arch.

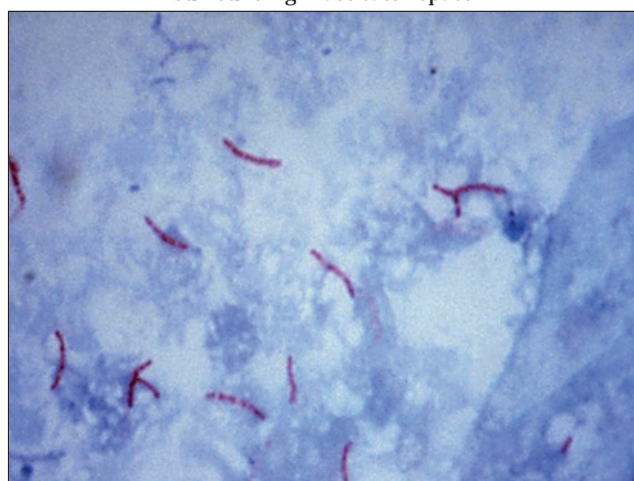


Fig 6: Cytology report

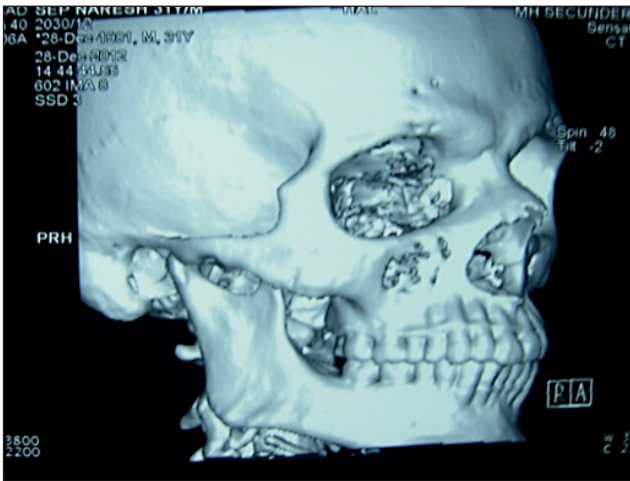


Fig 7: Post operative CT scan after 1 year.



Fig 8: Pre and Post Operative Extraoral Comparison

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