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Case study

A case of chronic granulomatous craniofacial osteomyelitis

Vidhu Sharma[a], Darwin Kaushal[b,*], Poonam Elhence[c]

[a] Department of Otorhinolaryngology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India
[b] Department of Pathology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India
[c] Additional Professor, Department of Pathology, AIIMS Jodhpur

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ABSTRACT

We present the clinical image of a 56-year old male who presented with chronic multiple craniofacial discharging sinuses with lytic, sclerotic destruction of facial bones on radiology causing gross facial deformity. He remained undiagnosed for years. The diagnosis was clinched on histopathology as a chronic granulomatous osteomyelitis showing Actinomyces.

A 56 year old male presented with multiple discharging sinuses, one beneath the left lower eyelid and two on the forehead for 3.5 years following extraction of left upper last molar tooth, associated with profuse foul smelling discharge, headache, facial deformity and loss of dentition. There was no other contributing history. Examination revealed a saddle nose deformity, one actively pus discharging lesion 1.5 × 1 cm in size over the left inferolateral orbital rim causing ectropion of the left eye. Two crater-like punched out ulcers (1 cm diameter) with inverted margins and foul smelling pus discharge were present over the left lateral aspect of the forehead with bony sequestra. A provisional diagnosis of chronic granulomatous craniofacial osteomyelitis was made. Tuberculin skin test read 16 × 20 mm at 48 h; Nucleic Acid Amplification Test for *Mycobacterium tuberculosis* done on tissue from the ulcer was negative. Blood work-up (complete hemogram with peripheral smear, blood biochemistry, urine microscopic examination and viral markers) was non-contributory. He had an ESR of 58 mm (1st h), HsCRP-65.46 mg/dl, a non-reactive RPR (Rapid Plasma Reagin), positive p-ANCA (perinuclear Anti-Neutrophil Cytoplasmic Antibodies), negative c-ANCA (cytoplasmic Anti-Neutrophil Cytoplasmic Antibodies), a normal serum ACE (Angiotensin Converting Enzyme).

A contrast enhanced computed tomography scan of the face revealed lytic sclerotic destruction of the bilateral frontal bones, bilateral nasal bones, alveolar process of left maxilla, left zygomatic bone. Histopathology revealed necrotic and few viable bony trabeculae with hematopoietic elements, foci of hemorrhage, colonies of filamentous, radiating organisms resembling Actinomyces, highlighted on Grocott-Gomori Methenamine Silver stain, negative for Periodic Acid Schiff stain, Ziehl-Neelsen stain. The patient was managed with daily dressings, intravenous Amoxicillin 1gm thrice a day and supportive treatment for six weeks. After 3 weeks of treatment, he had minimal pus discharge and healthy granulation tissue (Fig. 1).

Actinomycosis is a chronic suppurative granulomatous infection caused by members of the Actinomyces genus. Of the five species

* Corresponding author.
E-mail addresses: vidhusharma88@gmail.com (V. Sharma), drdarwin.aiims@gmail.com (D. Kaushal), drpoonamelhence@gmail.com (P. Elhence).

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Fig. 1. Clinical image of the patient after three weeks of treatment showing minimal pus discharge and healthy granulation tissue.

causing human infection, the most frequently isolated species is *Actinomyces israelii* [1]. Disease manifests in cervicofacial (50%), pulmonothoracic (30%) and abdominopelvic (20%) forms. Actinomyces are non spore forming, anaerobic/microaerophilic gram positive bacilli having low pathogenicity. Thus, they generally cause disease in a setting of tissue injury, for example, following dental extraction, maxillofacial trauma and in the presence of dental caries [2].

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Conflict of interest

None declared.

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