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Case Report

Dentistry Section

Cervicofacial Actinomycosis Mimicking Osseous Neoplasm: A Rare Case

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ABSTRACT

Actinomycosis is a rare chronic infectious disease caused primarily by *Actinomyces israelli*. Although they are normal inhabitants of the oral cavity, infection occurs when there is a breach in the mucosa allowing them access to the subcutaneous tissues. Poor oral hygiene, dental caries, recent dental extraction, oromaxillofacial trauma have been implicated as risk factors for actinomycosis. Cervicofacial actinomycosis is the most common form of this rare disease.

Here, we are presenting a rare case of actinomycosis involving the parotid gland and coronoid process of mandible in a young child where diagnosis was made based on histopathological findings since the classical clinical manifestations of actinomycosis, were absent in the case.

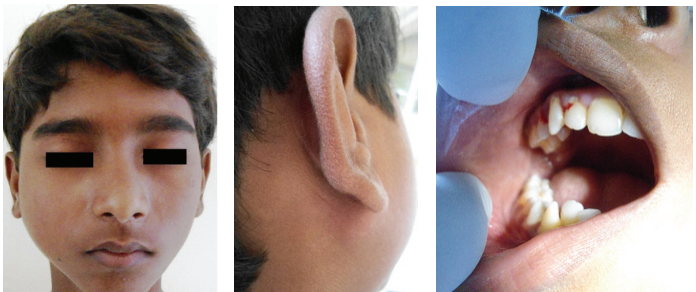
Keywords: Actinomycotic infection, Coronoid process, Histopathologic evaluation, Mandible, Parotid

CASE REPORT

A 13-year-old male child reported to the Department of Oral and Maxillofacial Pathology with the chief complaint of pain and difficulty in opening the mouth for last 6-7 months. Initially there was a small painful nodule over the right infra auricular region which gradually increased in size to attain the present dimensions. The patient did not give any history of trauma or tooth extraction. Many local physicians and dental surgeons were consulted and the patient received several courses of antibiotics and analgesics. However, he got no relief and concomitantly developed reduction in mouth opening. His past medical and family history were non contributory. He belonged to a poor socio-economic class.

Extraoral examination revealed diffuse swelling involving right middle and lower third of face and a small, localized, painless, mobile mass measuring about 2.0 x 2.0 cm. just below and behind the right ear resulting in elevation of ipsilateral ear lobule, with normal overlying skin [Table/Fig-1,2]. Nothing significant was noted on intraoral examination except for poor periodontal status and reduced mouth opening, Bleeding on probing was noted from the gingival margin of right maxillary lateral incisor and 1st premolar [Table/Fig-3]. Deviation of mandible towards left side was noted on opening the mouth.

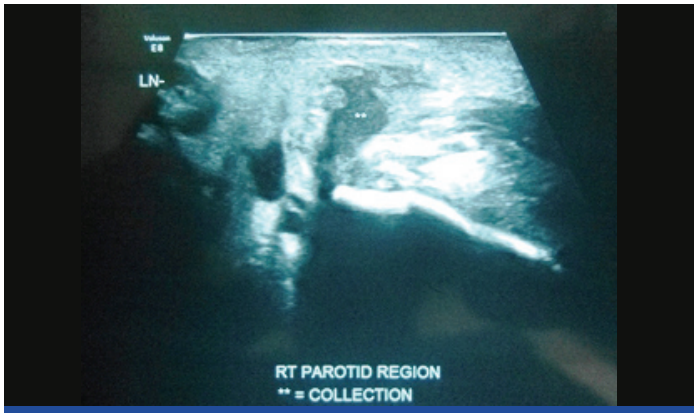
Routine haematological investigations revealed raised ESR (35 mm 1st hr.) and increased eosinophil count (13% in DLC) [Table/Fig-4]. Orthopantomogram (OPG) did not show any bony changes. No active parenchymal lesions were found in chest postero-anterior view (PA) radiograph. Ultrasonography (USG) findings revealed ill defined fluid collection with hypoechoic areas in the right parotid gland region [Table/Fig-5]. Computerised tomography (CT) scan with ionic contrast revealed heterogeneously enhancing lesion measuring 3.0 x 3.2 cm. in the right parotid region and multiple hypodense (osteolytic) areas in relation to right coronoid process of mandible [Table/Fig-6]. A provisional diagnosis of malignancy was made based on the clinical and radiological findings, our differential diagnosis included osteomyelitis, tuberculosis, intra osseous squamous cell carcinoma, Langerhans cell histiocytosis and osteogenic sarcoma.



[Table/Fig-1]: Extraoral photograph showing diffuse swelling involving right middle and lower third of face **[Table/Fig-2]:** Extraoral photograph showing small, localized swelling over right parotid region below the ear **[Table/Fig-3]:** Intraoral photograph showing no abnormal features except poor periodontal status

Hb%	12.2gm%
ESR	35 mm in 1st hr.
TLC	8500 / cubic mm.
DLC Neutrophils Lymphocytes Monocytes Eosinophils Basophils	 62% 23% 02% 13% 00%
RBC	Normocytic normochromic
Platelates	Adequate
BT	1 min 15 sec
CT	3 min 10 sec

[Table/Fig-4]: Haemogram of the patient

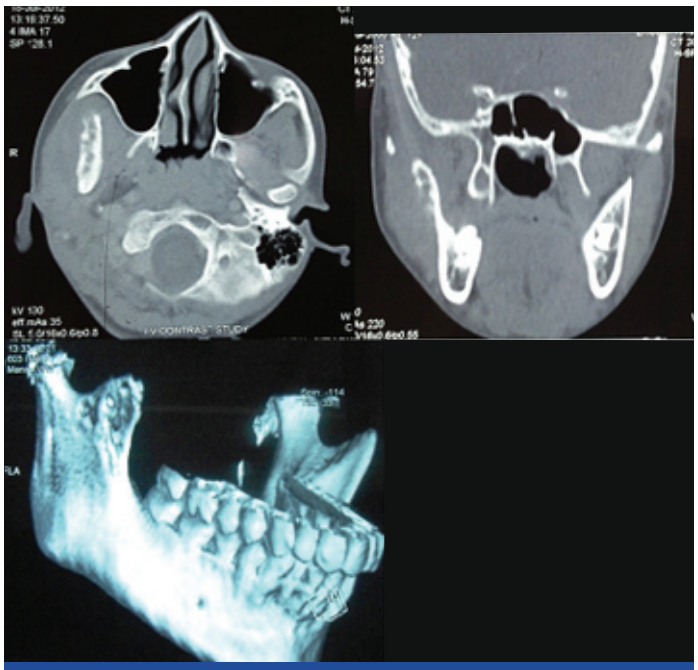


[Table/Fig-5]: Ultrasonography findings showing ill defined fluid collection with hypoechoic areas in the right parotid gland region

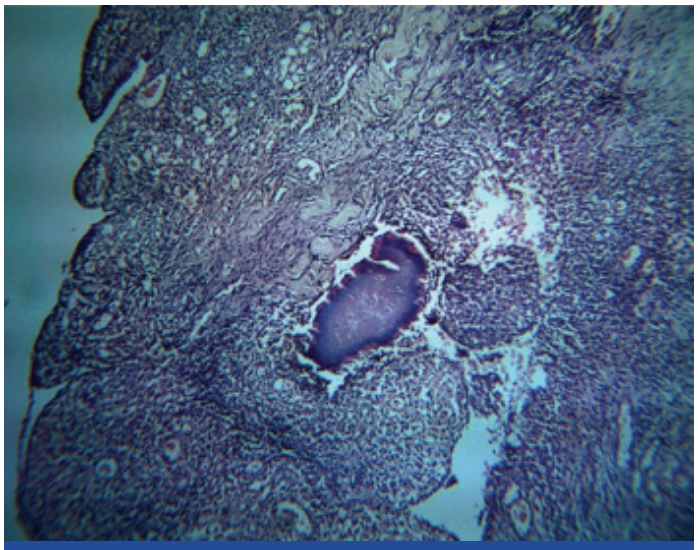
The patient was referred to the Department of Oral & Maxillofacial Surgery for incisional biopsy which was performed from the representative site and the specimen was sent for histopathological evaluation.

Haematoxylin and eosin stained sections revealed the presence of connective tissue stroma containing an ovoid mass surrounded by numerous polymorphonuclear leukocytes and lymphocytes with peripheral fibrosis. The peripheral portion of the mass stained eosinophilic while the core stained basophilic [Table/Fig-7,8]. Under

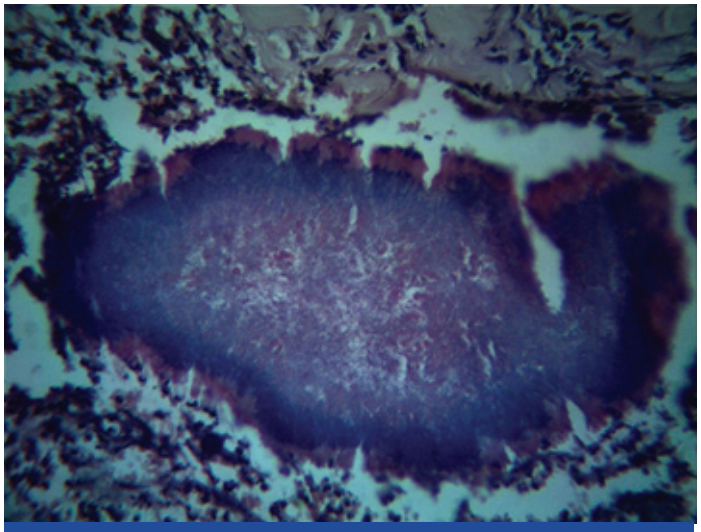
high power (40x, 100x) club shaped filaments forming a radiating rosette pattern were observed. These features were very much corroborative to actinomycosis [Table/Fig-9]. Thereafter, the patient was referred to the Department of General Medicine for further treatment where parenterally Cefuroxime 500 mg twice daily along with Metronidazole 200 mg thrice daily were prescribed and the patient was referred back to the Department of Oral & Maxillofacial Surgery for surgical treatment. After receiving the consent from the patient's parents, a superficial part of parotid gland, ramus, condyle, coronoid process of the involved side of mandible were resected because of the extensive bony involvement. Reconstructive plate was placed to correct the facial asymmetry [Table/Fig-10]. Immediate post surgical period was uneventful [Table/Fig-11]. We could not follow up the case as the patient did not report back.



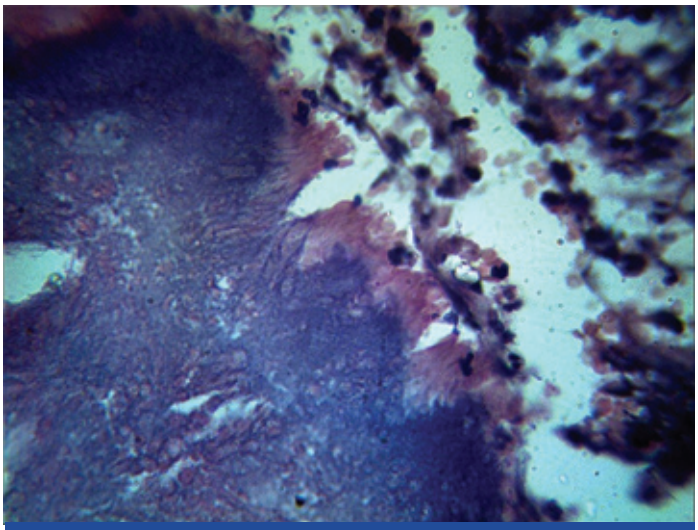
[Table/Fig-6]: Computed Tomographic views showing multiple osteolytic areas in relation to right coronoid process of mandible and the radiographic changes in parotid gland



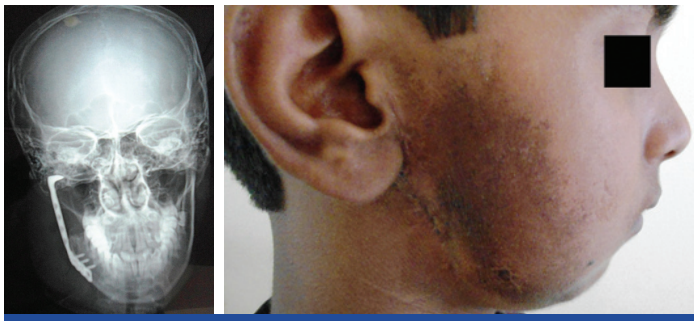
[Table/Fig-7]: Low power photomicrograph (H&E, 10x) showing an ovoid mass in the connective tissue stroma



[Table/Fig-8]: High power photomicrograph (H&E, 40x) showing actinomycotic colony surrounded by chronic inflammatory cells



[Table/Fig-9]: Oil immersion high power photomicrograph (H&E, 100x) showing club shaped filaments of actinomyces forming a radiating rosette pattern.



[Table/Fig-10]: PA view of skull showing reconstructive plate positioned over the involved area. **[Table/Fig-11]:** Postoperative extra-oral photograph of the patient after two weeks of surgery

DISCUSSION

Actinomycosis is a chronic, suppurative and granulomatous disease [1-3]. It is derived from a Greek word where "aktino means radiating appearance of sulfur granules and "mykos" means a fungal disease [4]. The first case in humans was reported by Von Langenbeck in 1845 and attributed to a fungus. In 1891, Wolff and Israel first isolated the micro organism from humans by culturing it anaerobically and growing it at body temperature. In 1960s, Waksman concluded that *Actinomyces* was not a fungus but a gram-positive bacteria [2,4].

In humans, the disease is caused primarily by A. *Israelii* and less frequently by A. *viscosus*, A. *propionica*, A. *naeslundii*, and A. *Odontolyticus* [3,5-10]. These micro-organisms are gram-positive, non-acid fast, anaerobic or microaerophilic filamentous branched bacteria which are very difficult to culture [2-7,9]. They are normal inhabitants of the oral cavity and are found in the periodontal pockets, gingival crevices, carious teeth, dental plaque, calculus or tonsillar crypts [5-9]. They are usually harmless but become virulent when there is breach in the mucosa and presence of copathogens [1,3,5-7].

Based on the site affected, Cope in 1938 classified actinomycosis clinically into cervicofacial (50%), pulmonothoracic (30%) and abdominopelvic (20%) [2,3,5,6]. Cervicofacial actinomycosis is the most common form of this rare disease [2-7,9]. Most cases are odontogenic in origin and occur predominantly in immunocompetent individuals where the frequently affected sites are parotids, submandibular glands and mandible [2-4,6,7].

It was earlier called "lumpy jaw" due to wooden, hard, board like lesion which had a lumpy appearance when the disease involved the jaws [4,6-8,10]. A retrospective study done in the University of Cologne reported the incidence of actinomycotic infection affecting the mandible (53.6%), cheek (16.4%), chin (13.3%), submaxillary ramus and angle (10.7%), maxilla (5.7%) and TMJ (0.3%) [4,9,11]. Various studies have reported mandible to be involved commonly than maxilla (4:1). [10]

Actinomycotic infection is commonly found between 4th to 6th decade of life and very rare in infants and children [6,12]. Males are affected more than females (4:1) [10,13]. The common initial signs and symptoms of infection such as fever, sudden onset of pain, swelling, erythema, and oedema may be absent [2]. Classically it manifests as a slowly growing, painless, indurated mass with multiple drainging sinus tracts on the skin or oral mucosa and occasionally presence of thick, yellow exudate with characteristic sulfur granules. Later due to involvement of masticatory muscles, pain and trismus develop [1-5,7,9,10]. It usually spreads into adjacent soft tissues without regard for tissue planes and is characterized by contiguous spread. Lymphadenopathy may develop later due to secondary infection [1-4,6,7,10]. Dissemination of the disease to distant organs like brain, lungs and digestive tract has been reported [4,7].

Imaging techniques like CT and USG show non-specific findings and are non-contributory to positive diagnosis. They are only helpful in delineating the surgical margins [2,5,7]. Hence, till today, histopathological evaluation of biopsy sample and culture of bacteria from the lesion are considered gold standard for diagnosis [3-5,7,9]. Still the disease is difficult to diagnose as it commonly mimics various infectious and non infectious conditions like tuberculosis, nocardiosis, fungal infection, chronic granulomatous lesion and malignancy [1-4,6,7,9]. Hence, it is considered as "the most misdiagnosed disease" and is listed as a "rare disease" by the Office of Rare Disease (ORD) of the National Institute of Health (NIH) [6,7].

Management of cervicofacial actinomycosis includes both antibiotics and surgery. Mild forms are treated with antibiotics alone but severe and complicated cases require high doses of antibiotics for prolonged period along with surgical debridement [2,3,5-7,9]. It has a very good prognosis and low mortality rate if diagnosed and treated early. Long term follow up is necessary since recurrence is common [3,9].

In the reported case, there was involvement of both hard (coronoid process) and soft tissue (parotid) by actinomyces. The involvement of the mandible was probably by direct extension from either a periodontal pocket or a soft tissue focus from the parotid [4]. The classical clinical manifestations of cervicofacial actinomycosis were absent in our case, Ultrasonography revealed ill defined fluid collection with hypoechoic areas in the right parotid gland region suggestive of parotid abscess. Computed tomography showed multiple osteolytic foci which were suggestive of an aggressive osseous malignancy. However, on histopathological examination we found an ovoid mass in the connective tissue stroma containing club shaped filaments forming a radiating rosette pattern, which were inconsistent with the imaging interpretation and diagnostic of "actinomycosis" [3,4,8,9]. Bony erosion of the coronoid region noted in CT scan demonstrates the tendency for actinomycotic infections to spread via direct extension without regard for anatomical barriers [3,6]. The patient was treated with antibiotics and surgical debridement. Thus, a very rare case of actinomycosis was diagnosed in the light of histopathological examination despite the absence of classical clinical features.

CONCLUSION

The diagnosis of actinomycosis has always posed a challenge to the clinicians. It should be considered in the differential diagnosis in case of any unusual destruction in the oral tissues even in the absence of typical clinical presentation and occasionally histopathological findings based on biopsy may be the only way to diagnose this rare disease.

REFERENCES

[1] Bala S, Narwal A, Gupta V, Duhan J, Goel P. Actinomycotic Osteomyelitis of Mandible Masquerading Periapical Pathology. *J Oral Health Comm Dent*. 2011;5(2):97-9.
[2] Lancella A, Abbate G, Foscolo AM, Dosdegani R. Two unusual presentations of cervicofacial actinomycosis and review of the literature. *Acta Otorhinolaryngol Ital*. 2008;28:89-93.
[3] Heo SH, Shin SS, Kim JW, Lim HS, Seon HJ, Jung SI, et al. Imaging of Actinomycosis in Various Organs: A Comprehensive Review. *RadioGraphics*. 2014;34(1):19–33.
[4] Bochev V, Angelova I, Tsankov N. Cervicofacial actinomycosis - report of two cases. *Acta Dermatoven APA*. 2003;12(3):105-08.
[5] Volante M, Contucci AM, Fantoni M, Ricci R, Galli J. Cervicofacial actinomycosis: still a difficult differential diagnosis. *Acta Otorhinolaryngol Ital*. 2005;25(2):116–19.
[6] Rodan R. Unusual Presentation of Actinomycosis Misdiagnosed as Severe Periodontal Destruction. *JRMS*. 2012;19(1):53-6.
[7] Valour F, Sénéchal A, Dupieux C, Karsenty J, Lustig S, Breton P, et al. Actinomycosis: etiology, clinical features, diagnosis, treatment, and management. *Infection and Drug Resistance*. 2014;7:183–97.
[8] Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 3rd ed. India: Elsevier Publishers; 2009. Pp. 203-205.
[9] Vorasubin N, Wu AW, Day C, Suh JD. Invasive Sinonasal Actinomycosis: Case Report and Literature Review. *Laryngoscope.* 2013;123:334-38.
[10] Rathnaprabhu V, Rajesh R, Sunitha M. Intraoral actinomycoticlesion: a case report. *J Indian Soc Pedod Prev Dent*. 2003;21(4):144-46.
[11] Schaaal KP, Beaman BL. Clinical significance of actinomycetes. In: Goodfellow M, Mordarski M, Williams ST, eds. The biology of actinomycetes. New York: Academic Press; 1983. Pp.389-424.
[12] Miller M, Haddad AJ. Cervicofacial actinomycosis. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998;85:496-508.
[13] Lerner PI. Actinomyces and arachinia in infectious diseases. Nonsiewicz MJ, Philadelphia: W.B.Saunders Co; 1992; 1626-1632.

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