

Tonsillar Actinomycosis with Multiple Epidermoid Cysts: A Rare Case Report

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Abstract: This study reports a case of multiple epidermoid cysts of the tonsil with actinomycosis in a 56-years-old female patient presented to our ENT outpatient department with discomfort in the throat and presence of a few whitish masses over her right tonsil which was her major concern. She underwent a tonsillectomy followed by a course of antibiotic for 3 weeks the diagnosis was confirmed by histology. Our results indicate that actinomycetes may play an active role in the etiology of epidermoid cyst of tonsillar tissue. The clinical importance of recognising this organism lies in the fact that definitive treatment requires a long course of penicillin group antibiotics even after surgical excision. To the best of our knowledge, this is the first case report in English-indexed literature. **Keywords:** Palatine tonsil, multiple cysts, epidermoid cyst, Actinomyces

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I. Introduction

The palatine tonsil is a mass of lymphoid tissue situated in the lateral wall of oropharynx, where it lies within the tonsillar fossa. They play an extremely remarkable role in the antimicrobial defense of the body. Actinomycosis are filamentous branched bacteria, living as commensal organisms in the oral cavity, becoming invasive when, through a mucosal lesion, they gain access to the subcutaneous tissue. Actinomycosis can present in a variety of forms and may mimic other infections or even neoplasm. The clinical significance of presence of actinomyces in tonsils and its role in pathogenesis of tonsillitis is a topic of debate for long.¹ A varieties of cysts can arise in the tonsil with tonsillar retention cyst being the most common. Epidermoid cyst, lymphoepithelial cyst, hydatid cyst are other rare causes of tonsillar cyst.² Intraoral epidermoid cysts involve 0.01% of the population.³ The incidence of epidermoid cysts in head and neck has been found to be 1.6-6.9%.⁴ Various studies have been postulated in its pathogenesis, that include the inclusion of ectodermal tissues during embryogenesis, metaplasia in response to prolonged irritation.

II. Case Report

A 56 year old woman presented to our ENT outpatient department with discomfort in the throat for 6 months. She incidentally noticed few whitish masses over her right tonsil which was her major concern. There was no dysphagia or dyspnea. On examination she had enlargement of her right tonsil with multiple whitish cysts over tonsillar surface. Right tonsil was larger in size than the left. There was no cervical lymphadenopathy. Blood and urine examination were within normal limit. Patient was assessed for HIV status and was found seronegative. We planned tonsillectomy as asymmetrical adult tonsils with normal mucosa in the absence of cervical lymphadenopathy are associated with 7% risk of malignancy.⁶ Under general anesthesia coblation tonsillectomy was done. The procedure was uneventful. Specimen was sent for histopathology. Patient was discharged on the 2nd post-operative day. Follow up was uneventful. Histologic evaluation of the specimens was done on hematoxylin and eosin-stained slide that showed -tonsil with a cyst lined by stratified squamous epithelium showing ulceration with formation of keratinous cyst. There are colonies of Actinomycosis amongst the hyperplastic lymphoid follicle along with dense acute inflammation. A diagnosis of Tonsillar Actinomycosis with infected epidermoid cyst of the tonsil was made. The uneventful healing of the fossa was noted by one week. The patient was started on a combination of amoxicillin (500mg) with clavulanic

acid (125mg) for 3 weeks.⁷The patient made complete recovery and, moreover, follow-up revealed no recurrence of the infection and/or cyst.



Fig 1: View of the asymmetrical hypertrophy of the right tonsil with multiple cysts

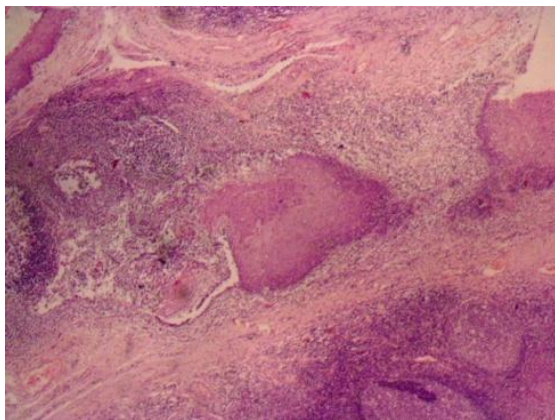


Fig 2: Cystic cavity within the tonsillar tissue lined with keratinized epithelium. H/E x40

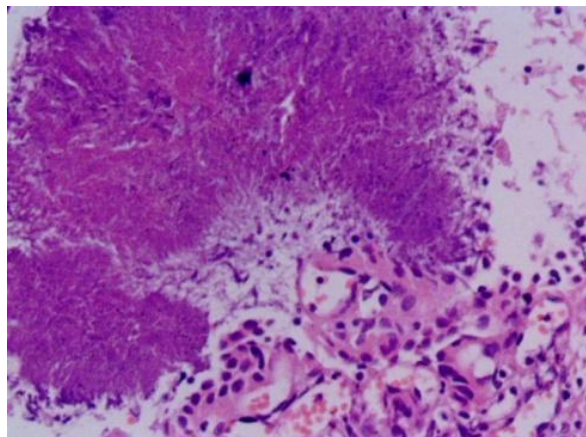


Fig 3: High power view showing actinomyces colony with "sunray appearance" (H/E, 100x)

III. Discussion

Chronic tonsillitis most often affects children, but can be seen in adults. Many studies have stated that chronic inflammation is present in both tonsillitis and tonsillar hypertrophy. The differential diagnosis to be considered for tonsillar hypertrophy includes tonsillar tumors, tumors of parapharyngeal space, infectious etiology and inclusion cysts. Cervicofacial involvement is the most common manifestation of actinomycosis, accounting for 50 percent of all cases.⁸ The pathogenesis is related to its ability to act as an intracellular parasite and thus resist phagocytosis as well as its tendency to spread without respect for established tissue plains or anatomic barriers. Actinomyces are gram-positive, non-acid fast, anaerobic or microaerophilic filamentous

branched bacteria which are very difficult to grow in culture, with < 30% of cultures being positive. These bacteria are all normal commensals of the human oral cavity. For the diagnosis of actinomycosis to be established, typical findings on histological examination of the tissue with an outer zone of granulation tissue and a central zone of necrosis containing many granules that represent microcolonies of actinomyces should be present.⁹ Van Lierop et al found no tissue reaction due to actinomyces colonies and hence reported no correlation between tonsillar actinomycosis and recurrent tonsillitis.¹⁰ Contrary to this, Aydin et al analyzed 1820 tonsillectomy specimens and reported that cryptitis was a common histopathologic indicator of tonsillar actinomycosis.¹¹

Histologically there are three types of dermoid cysts i.e epidermoid cyst, true dermoid cyst and teratoid. Roser, in 1859, described the term epidermoid cyst. Epidermoid is a type of dermoid cyst, commonly seen in face, neck, chest or trunk; usually occur just beneath the skin.¹² The occurrence of epidermoid cysts in the mouth is extremely rare. These cysts generally present slow and progressive growth, and even if they are congenital, they often are not diagnosed until the second or third decade of life.

Basal cell carcinoma and squamous cell carcinoma arising in the wall of an otherwise conventional epidermoid cyst has been seen.^{13, 14, 15} However, no carcinomatous transformation was noted in our case.

IV. Conclusion

Though tonsillitis is the commonest lesion diagnosed in all age groups, histopathology plays an important role in diagnosis of various other non-neoplastic and neoplastic lesions of tonsil. The histopathological examination of tonsils in our case report also documents Actinomyces colonies with infected epidermoid cyst. Dense acute inflammation caused by Actinomycosis might be a reason for development of epidermoid cysts in a rare site like tonsils. In conclusion, epidermoid cysts as well as actinomycosis, a rare occurrence in the head and neck area, can also be found inside the palatine tonsils and cause asymmetrical hypertrophy. The clinical importance of recognising this organism lies in the fact that definitive treatment requires a long course of penicillin group antibiotics even after surgical excision.⁷ There might be existence of two separate clinical entity in a single case, one being the causative factor of another. There has been evidence of carcinomatous transformations in epidermoid cysts hence the patient should be kept in regular follow up.

V. Clinical Significance

We herewith report a case of exceptional existence of multiple epidermoid cysts of the tonsil with actinomycosis. To the best of our knowledge, it is the first report in English-indexed literature. This study also emphasizes on the need for further research into the etiopathogenesis of the cysts and the sequels of actinomycosis infections.

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