



A RARE CASE OF PAEDIATRIC CERVICOFACIAL ACTINOMYCOSIS WITH OBSTRUCTIVE ADENOTONSILLAR HYPERTROPHY

Dr. Shilpa H

Senior Resident, Department of Otorhinolaryngology, MVJ Medical College & Research Hospital, Hoskote, Bangalore-562114, Karnataka.

Dr. Amrita Suzanne Mathew *

Assistant Professor, Department of Otorhinolaryngology, MVJ Medical College & Research Hospital, Hoskote, Bangalore-562114, Karnataka. *Corresponding Author

ABSTRACT

Actinomycetes are commensal bacteria within the oral cavity. Actinomycosis is seen more commonly in adults and is rarely encountered in children. Actinomycosis of the tonsillar tissue is a rare pathological condition whose clinical significance has been disputed by authors worldwide. When present in tonsillar tissue, they may present with either features of recurrent tonsillitis, or obstructive sleep apnoea secondary to tonsillar hypertrophy. Craniofacial actinomycosis can present with a swelling/abscess at any head and neck site. Its importance lies in its diagnosis on histopathological examination, and management additionally with long term penicillin. We therefore report a paediatric case of cervicofacial actinomycosis with obstructive adenotonsillar hypertrophy.

KEYWORDS : Actinomycosis/diagnosis; Tonsillitis/microbiology; Tonsillitis/complications

INTRODUCTION

Actinomycetes are Gram-positive, spore-free, facultative anaerobic bacteria showing branching filaments with varying cellular morphology. They do not exist in nature. Humans are the only natural reservoir.¹ Actinomycetes are common bacteria found in the oral cavity and palatine tonsils and is prevalent in 2-3% of subjects.² Actinomyces israelii and Actinomyces naeslundii are the most common species isolated in humans.^{3,4}

Actinomycosis is a rare chronic bacterial infection that affects humans, with an incidence rate as low as 1 in 300,000 per year.⁵ It commonly occurs in the 4th to 6th decades and commonly involves cervicofacial, thoracic, abdominopelvic regions and the central nervous system.² Actinomycosis is not frequently encountered these days because of widespread use of antibiotics.⁶ Cervicofacial actinomycosis is the most common manifestation of actinomycosis, accounting for 50% of all cases. It is a chronic disease characterised by abscess formation, draining sinus tracts, fistulae and tissue fibrosis and should be included in the differential diagnosis of any soft tissue swelling in the head and neck.⁷

Actinomycetes are found in the gingival clefts and tonsillar crypts in normal structure of oral flora, especially in periodontal pockets, dental plaques, rotten teeth and upper respiratory tract.^{1,8} Its presence in tonsillar tissue was first detected in 1896. The role of Actinomycetes in the tonsillar tissue pathology is not fully understood. Actinomycotic infection has been reported in a small percentage of people presenting with chronic tonsillitis.¹ Infection occurs if the mucosal barrier is impaired and spread occurs to the surrounding tissues due to the proteolytic enzymes they produce.^{9,10}

Actinomycosis infection of the head and neck, although fairly uncommon, represents an important entity because of its varied presentation that may mimic other common diseases, the difficulties involved in its diagnosis, and the long course of treatment mandatory to eradicate the disease.¹¹ We therefore report a paediatric case of peritonsillar abscess with acute on chronic adenotonsillitis with cervical lymphadenopathy and obstructive sleep apnoea, diagnosed with actinomycosis, treated surgically and medically.

CASE REPORT

A 4 year old male patient presented to the Department of Otorhinolaryngology and Head and Neck Surgery with the complaints of throat pain, difficulty swallowing and fever in the preceding week. History of similar recurrent episodes of throat pain with fever, bilateral neck swelling, nasal obstruction, mouth breathing, snoring, excessive daytime sleepiness and fatigue for the

past 1 year for which he was prescribed antibiotic and analgesic by local practitioners for each episode. A swelling was noticed below the left half of the jaw for the last 8 months.

Oral and oropharyngeal examination revealed dental caries, bilateral enlarged tonsils (Grade III) with congested overlying mucosa and left peritonsillar abscess. Anterior rhinoscopy was normal. On diagnostic nasal endoscopy, adenoid enlargement was evident. Neck examination revealed tender left submandibular and bilateral jugulodigastric nodes. The patient was diagnosed as a case of left peritonsillar abscess with acute on chronic adenotonsillitis with cervical lymphadenopathy and obstructive sleep apnoea.

The patient was admitted and given parenteral antibiotics. Purulent discharge from incision and drainage of the peritonsillar abscess showed no growth. Ultrasonography of the neck confirmed cervical lymphadenopathy. Fine needle aspiration cytology was suggestive of reactive lymphadenitis. 4 weeks following the resolution of the peritonsillar abscess (Fig.1), the patient underwent endoscopic microdebrider assisted adenoidectomy with dissection tonsillectomy. Intra and postoperative period was uneventful. Resected tissue was routinely submitted for histopathological examination which revealed hypertrophic tonsil with dense neutrophilic infiltration and bacterial colonies with sulphur granules (Fig.2). These granules stained positive with Gram stain and periodic acid Schiff (PAS) stain and negative with acid-fast stain, confirming the presence of actinomycosis. Patient was then put on a course of intravenous penicillin G for 1 month followed by a course of oral penicillin for 3 months. Postoperative recovery was satisfactory. At the end of 1 month, there was complete resolution of lymphadenopathy. There was considerable symptomatic relief from obstructive symptoms at the end of 3 months.

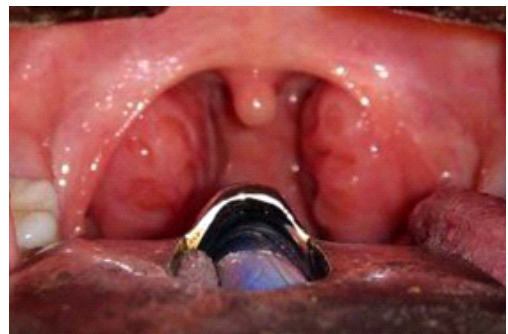


Fig 1-Preoperative picture following resolution of left peritonsillar abscess

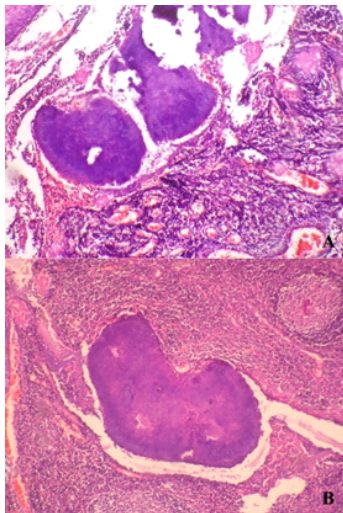


Fig 2 A-Section of tonsillar tissue showing Actinomycotic colonies within the crypts; B-Magnified view of an Actinomycotic colony within a tonsillar crypt

DISCUSSION

Actinomycosis is a rare soft tissue infection caused by a gram-positive, anaerobic bacteria. The principal cause of human Actinomycosis is *A. israelii*.¹² It is seen only approximately once a year in major medical centers, and is rarer still in the pediatric population, which includes our case.¹³ Actinomycotic infection has been reported in a small percentage of people presenting with chronic tonsillitis.²

A significant male predominance was found in both the pediatric and adult groups.¹⁴ Tonsils in children younger than 3 years showed no Actinomycosis.¹⁵ Malgarejo et al. reported that actinomycosis was more prevalent in patients aged 5-16 years.¹⁶ Lierop et al. demonstrated a statistically significant correlation between Actinomycosis colonisation and age, Actinomycosis being more common in older children, especially those over 5 years of age.⁹ In a study by Kansu, the incidence of Actinomycosis was significantly higher in the adult group than the pediatric group.¹⁴ Our patient is a male aged 4 years, being younger than the average age in the literature reviewed.

Commonest form of actinomycosis is cervicofacial and results from direct invasion of commensal oral actinomycetes into oral tissue by oromaxillofacial trauma, dental implantation and dental caries.¹⁷ Some systemic diseases increase the incidence of Actinomycosis. Bhargava et al. found that Actinomycetes in the tonsillar tissue of people with sickle cell anemia, thalassemia and bronchial asthma was at a higher rate and the probability of disease formation was also higher. It was thought that poor dental hygiene caused this situation.¹⁴ Literature has also revealed that infection due to Actinomycetes sp. was related with long term immunosuppressed conditions.⁴ However, none of these factors other than dental caries was present in our case.

Clinically, patients present with varied symptoms ranging from sore throat, fever, discharging sinuses and hypertrophied inflamed tonsils, etc. Such patients generally give history of recurrent tonsillitis as seen in our case.¹¹ Actinomycosis is believed to have a pathological influence on tonsil size. Kutluhan et al. showed that there was a significant relation between Actinomycosis and enlargement of tonsillar tissue.³ Studies showed that patients with obstructive sleep apnoea had a higher incidence of Actinomycosis in the tonsillectomy specimens, than in those with recurrent attacks of tonsillitis.^{2,18,19} There was a statistically significant higher rate of Actinomycetes colonization in children undergoing tonsillectomies for sleep disordered breathing compared to recurrent tonsillitis.¹⁹ As a result, it is thought that Actinomycosis settled in the tonsillar tissue

can play a role in tonsillar hypertrophy.¹⁴ Our patient had features of obstructive sleep apnoea secondary to adenoiditis and grade 3 tonsillar hypertrophy.

Tonsils with Actinomycosis in both the pediatric and the adult groups were found to be significantly larger than those without Actinomycosis in a study by Kansu.¹⁴ In the paediatric group, while the mean volume of tonsils with Actinomycosis in their tonsillar tissue was 9.6 ± 3.9 cc, the mean volume of tonsils without Actinomycosis was 7.2 ± 3.0 cc.^{4,14} In our patient, the volume of tonsils was 11.8 cc, correlating with the average volume of tonsils with Actinomycosis.

Özgürsoy et al. in a study involving histological examination of tonsillar specimens, found that there was a significant increase in lymphoepithelial squamous cell metaplasia and secondary follicles in the Actinomycosis-positive tonsillar tissue.^{4,14} Several other authors have also studied histopathological sections from tonsillectomy specimens and have arrived at similar conclusions.^{12,18} It is thought that the microorganisms settled in the tonsil produce some toxins, resulting in tonsil hypertrophy.^{9,19}

The common indications for tonsillectomy are episodes of recurrent tonsillitis or tonsillar hypertrophy which can cause obstructive symptoms such as snoring and sleep apnoea, both of which were present in our case preoperatively.¹⁷ One of the most important complications of Actinomycosis settled in the tonsillar tissue, other than tonsil hypertrophy is bleeding in the late postoperative period, but the postoperative period was uneventful in this case.¹⁹

For the diagnosis of actinomycosis to be established, two of the following conditions must be present: positive cultures, sulfur granules or biopsy specimens showing the organism. However, cultures are difficult to obtain and histopathological identification is the gold standard.¹¹ As identified in our case, the typical findings include the presence of sulfur granules seen as basophilic masses with a granular center and a radiating fringe of club-shaped protrusions as well as the distinctive filamentous and beaded actinomycetes, the so-called "ray-fungus" appearance of an Actinomycosis colony.^{6,11,2,18} Hematoxylin-eosin staining has been shown to be highly effective in detecting Actinomycosis colonies.¹⁸

Classical surgical excision is thought to be a definitive treatment for head and neck actinomycosis, and tonsillectomy is usually curative for tonsillar actinomycosis.² However, the current recommended therapy which was followed in our case includes 4 weeks of high dose IV Penicillin followed by a 3 to 6 month course of oral Penicillin, continuing treatment even after total resolution of symptoms to prevent recurrence. Other antimicrobials that have been found to be effective include tetracycline, erythromycin and clindamycin. These can be used in Penicillin-allergic patients with good success rates. Ciprofloxacin has been used for treatment of recalcitrant cases.¹⁷

CONCLUSION

Actinomycosis are microorganisms that can be found in the tonsillar tissues opportunistically. There is a higher incidence in those with features of obstructive tonsillar hypertrophy than in recurrent tonsillitis. Actinomycosis can present with a swelling or abscess at any head and neck site and should therefore always be considered in the differential diagnosis. Definitive diagnosis is by histopathological examination. Appropriate treatment is necessary for eradication. Our case highlights the importance of microscopic examination of routine tonsillectomy specimens in all age groups for the identification of the possible etiological agent for tonsillar hypertrophy.

REFERENCES

1. Can S, Bayındır T, Kuzucu C et al. Cervicofacial Actinomycosis: A Case Report Of Acute Suppurative Clinical Progress. *Bozok Med J*. 2014;1(1):12-16.
2. Bhargava D, Bhushnurmath B, Sundaram KR et al. Tonsillar Actinomycosis: A clinicopathological study. *Acta Trop*. 2001;80:163-168.
3. Kutluhan A, Şalvız M, Yalçın G et al. The role of the actinomycetes in obstructive

- tonsillar hypertrophy and recurrent tonsillitis in pediatric population. *Int J Pediatr Otorhinolaryngol*.2011;75:391-394.
4. Ozgursoy OB, Kemal O, Saatci MR et.al. Actinomycosis in the etiology of recurrent tonsillitis and obstructive tonsillar hypertrophy: Answer from a histopathologic point of view. *J Otolaryngol Head Neck Surg*.2008;37:865-869.
 5. Aini RI, Ratunanda SS, Wiyana W et.al. Aktinomikosis di tonsil lingualis dan supraglotis sebagai manifestasi klinis pertama pada pasien imunokompromais. *Oto Rhino Laryngologica Indonesiana*.2017;47(1):81-88.
 6. Kenji T, Kyoko K, Satoru K et.al. A case of actinomycosis causing unilateral tonsillar hypertrophy. *Acta oto-laryngologica*.2006;126(9):1001-1004.
 7. Kononen E, Wade WG. Actinomycosis and related organisms in human infections. *Clin Microbiol Rev*.2015;28:419.
 8. Yasan H, Çiriş M, Özel BF et.al. The significance of histopathologic tonsillar actinomycosis in pediatric patients with recurrent acute tonsillitis. *KBB-Forum*.2006;5:1-4.
 9. Lierop AC, Prescott CA, Smith CC. An investigation of the significance of actinomycosis in tonsil disease. *Int J Pediatr Otorhinolaryngol*.2007;71:1883-1888.
 10. Ashraf MJ, Azarpira N, Khademi B et.al. Relation between actinomycosis and histopathological and clinical features of the palatine tonsils: An Iranian experience. *Iran Red Crescent Med J*.2011;13:499-502.
 11. Mohanty S. Actinomycosis of faucial tonsil masquerading as oropharyngeal malignancy. *Indian J Otolaryngol Head Neck Surg*.2006;58:82-84.
 12. Sujatha N, Manimaran M, Rao R et.al. Histopathological Features of Tonsils & Significance of Actinomycosis in Chronic Tonsillitis. *IOSR Journal of Dental and Medical Sciences* 2015 Apr;14(4):105-109.
 13. Stewart MG, Sulek M. Pediatric actinomycosis of the head and neck. *Ear Nose Throat J*.1993;72(9):614-616, 618-619.
 14. Kansu L. Relation of Actinomycosis with Tonsillar Hypertrophy and Antibiotic Use. *Turk Arch Otorhinolaryngol*.2017;55:17-21.
 15. San T, Gürkan E, Erdoğan B et.al. Does actinomycosis have any role in tonsillar diseases in children? *Istanbul Med J*.2014;15:209-212.
 16. Melgarejo MP, MeSeguer MD, Garrido MA et.al. A correlation between age and Actinomycosis in the adenotonsillar tissue in children. *B-ENT*.2006;2:95-97.
 17. Hasan M, Kumar A. Actinomycosis and tonsillar disease. *BMJ Case Reports*.2011;doi:10.1136/bcr.01.2011.3750.
 18. Pransky SM, Feldman JI, Kearns DB et.al. Actinomycosis in obstructive tonsillar hypertrophy and recurrent tonsillitis. *Arch Otolaryngol Head Neck Surg*.1991;117:883-885.
 19. Riffat F, Walker P. Prevalence of tonsillar actinomycosis in children undergoing tonsillectomy for sleep disordered breathing compared with recurrent tonsillitis. *Int J Pediatr Otorhinolaryngol*.2009;73:1111-1113.
 20. Schrock A, Send T, Heukamp L et.al. The role of histology and other risk factors for post-tonsillectomy haemorrhage. *Eur Arch Otorhinolaryngol*.2009;266:1983-1987..