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Middle ear and mastoid actinomycosis

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Abstract
Actinomycosis is a granulomatous infection which is most common in the cervicofacial region. It is very rare in middle ear and mastoid. Actinomycosis should be considered as one of the differential diagnosis of chronic otitis media, although it is rare. The diagnosis is often made at histopathological examination only. Surgical debridement and prolonged course of antibiotics (penicillin) is the treatment of choice in middle ear actinomycosis. Early diagnosis and prompt treatment make the prognosis good. Here we report a case of a 9 year old girl who presented with recurrent otorrhea, initially suspected as a case of chronic otitis media squamous disease, finally diagnosed to have actinomycosis.

Keywords: actinomycosis, middle ear, mastoid, otorrhea

1. Introduction
Actinomycetes are seen as commensals in oropharynx, gastrointestinal tract and female genital tract of healthy individuals [1, 2]. But they can cause chronic granulomatous disease in humans most commonly after minor trauma in cervicofacial region, abdomen and thorax [3, 4]. Actinomycosis of middle ear and mastoid is a rare entity [1] and thought to occur by direct spread from nasopharynx via eustachian tube. The clinical features of middle ear actinomycosis are nonspecific and very similar to those of chronic suppurative otitis media, hence there will be delay in diagnosis.The English literature has reported less than 50 cases of tympanomastoid actinomycosis. Here we are presenting a case of 9 year old girl affected by right middle ear and mastoid actinomycosis.

2. Case Report
A 9 year old girl came to our outpatient clinic with complaints of recurrent foul smelling, purulent and blood-stained discharge from right ear since early childhood. There was also decreased hearing on right side of 3 months duration. She had received many courses of topical and systemic antibiotics, but the symptoms persisted even though there was mild relief. Her past medical history was unremarkable.

On examination there was a congested polyp with granulation, in right external auditory canal. Polyp excision was done, but the canal was filled with polyp within 2 weeks. The histopathology report came as inflammatory polyp. Pure tone audiogram showed right sided mild conductive hearing loss. Then we proceeded with high resolution computed tomography of temporal bone which showed mildly enhancing soft tissue density lesion in right external auditory canal causing bony erosion of posterior canal wall & mastoid cortices (figure1 and 2).

From the clinical picture and imaging, we arrived at a diagnosis of chronic otitis media squamous disease. She underwent modified radical mastoidectomy with type III tympanoplasty. Incus and malleus were eroded and embedded in granulation tissue and polyp. The final histopathological report came as Actinomycosis (figure 3 and 4). Subsequently she was put on penicillin for 6 months. During third month of follow up there was granulation tissue in the mastoid cavity, the same was cauterised with trichloroacetic acid. She is asymptomatic for the last two years of follow up.

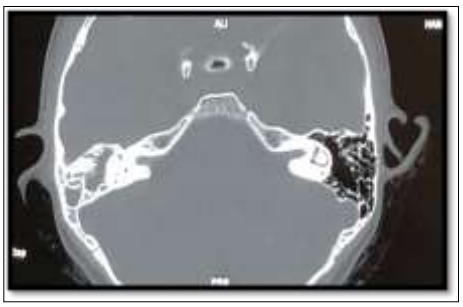

Fig 1: Preop CT- axial image

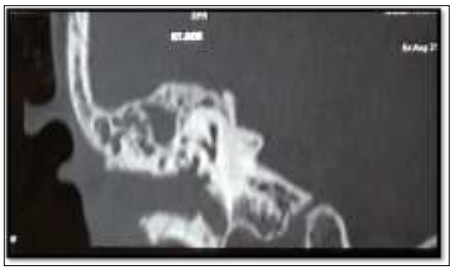

Fig 2: Preop CT- Coronal image

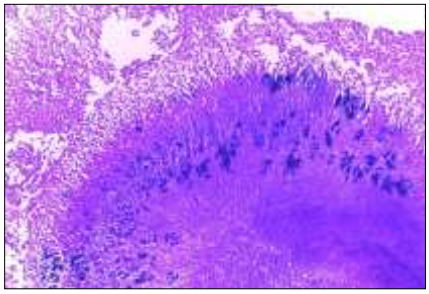

Fig 3: H&E stain-sulphur granules with basophilic radiating filaments in background of inflammation

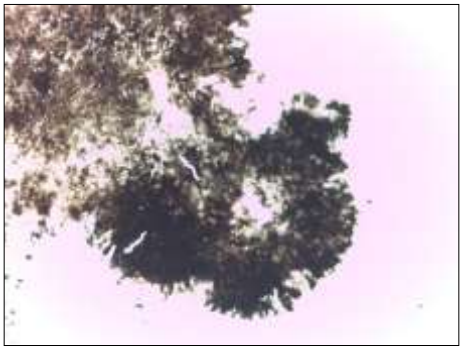

Fig 4: GMS stain showing actinomycotic colonies

3. Discussion

Actinomycosis is a rare, chronic and slowly progressive granulomatous disease caused by, Gram-positive, facultative anaerobic, non-spore forming, non-acid-fast filamentous bacteria of the 'Actinomycetaceae' family which comes under genus 'Actinomyces' [5]. In 1878 Israeli first described human actinomycosis. Actinomyces israeli is the most common pathogen involved. There is no human to human transmission known [6].

Actinomycosis, caused by Actinomyces israelii, occurs mainly in the cervicofacial area (50%) and affects the neck, face, mandible and tongue. The next common sites are abdomen (25%) and the thorax (15%) [3]. The pelvic organs may be infected from an intra-uterine device [6]. Middle ear and mastoid are rare sites for actinomycosis.

The disease is characterized by abscess formation, tissue fibrosis and draining sinuses. The disease can reach the middle ear through any of the following three routes-

1. From nasopharynx through the eustachian tube into the middle ear (most common).
2. Directly through external auditory canal as a result of external trauma and perforation.
3. Via blood stream (only in major infections) [3].

It can be seen that middle ear actinomycosis occurs more commonly at younger age in contrast to the ones in other parts of the body [7]. The middle ear actinomycosis usually presents like chronic otitis media with an indolent course and intermittent otorrhea, which is resistant to several antibiotics as in our case. Otoscopic examination may reveal a congested tympanic membrane, sometimes bulging out or a perforation with presence of inflammatory polyp or granuloma, but certain diagnosis from otoscopy alone is difficult. In our patient, there was an inflammatory polyp.

CT scan does not help in diagnosis and is useful only in defining the extent of the disease [8]. The isolation and culture of the causal microorganism is the gold standard method, but cultures are negative in more than 70% cases [6], because actinomyces are anaerobic and coexist with other bacteria [9]. So, to date, almost all reported cases of middle ear and mastoid actinomycosis have been diagnosed by histopathological examination. Pus drained from actinomycotic collection characteristically contains the organism in distinct granules (sulphur granules). These granules comprise a mass of actinomyces included in a complex of polysaccharides, calcium, proteins. The granules are surrounded by granulomatous inflammatory tissue [3]. They are stained by Grocott-Gomori methanamine silver nitrate stain (GMS), Maccallen-good posture stain and PAS stain [2, 10]. Even in our case, the diagnosis was made by histopathological examination of the tissue removed during surgery. Newer techniques like 16S rRNA sequencing polymerase chain reaction are good diagnostic tools, but in clinical practice they are not routinely used [1, 9, 11].

The differential diagnoses of chronic otorrhea with negative routine bacteriological cultures include;

- Tuberculosis: Chronic otorrhea and hearing loss are the main symptoms and is associated with facial paralysis in 40% of cases [12].
- Nocardiosis: It is a granulomatous and suppurative infection like actinomycosis. It also can produce sulphur granules, but not in visceral tissue. They are acid fast bacillus.
- Cholesteatoma: most common diagnosis in case of otorrhea [3].

Middle ear and mastoid actinomycosis can lead to complications like facial nerve palsy, labyrinthine fistula, sigmoid sinus thrombosis [2, 13]. In pre-antibiotic era there was more chance for intracranial complications.

The recommended treatment for middle ear and mastoid actinomycosis is surgery and prolonged antibiotic course with penicillin for 6-12 months [14]. In case of allergy to penicillin other antibiotics like tetracycline, erythromycin, clindamycin or chloramphenicol can be used [15]. Surgical debridement helps in removal of necrotic debris and granulation which is the cause for anaerobic environment for the growth of organism. However, in selected cases if there is good ventilation and drainage, surgery would not be mandatory. So, if early diagnosis is possible, the patient can be managed with nonsurgical treatment [8].

4. Conclusions

Actinomycosis of middle ear and mastoid is a rare clinical entity and only less than 50 cases were reported in the world literature so far. Due to the nonspecific presentation and culture negativity, there is often a delay in diagnosis and treatment. Early treatment is necessary to prevent complications. Even though it is a rare entity; it should always be included in the list of differential diagnosis of chronic otitis media resistant to standard therapy.

5. Acknowledgements

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6. Declarations

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Conflict of interest: None declared

Ethical approval: NA

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