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A Rare Case in a Child: *Actinomycosis* of the Middle Ear

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ABSTRACT

Background: *Actinomyces* is an anaerobic, filamentous bacterium that causes granuloma formation and suppurative infection. Although it is part of the normal flora of the oropharynx, gastrointestinal tract, and genital tract, it is rarely encountered in the middle ear and mastoid cavity.

Case Report: We present the case of a 4-year-old child diagnosed with chronic otitis media who had experienced ear pain and discharge for more than 3 months and whose symptoms did not resolve despite antibiotic therapy. *Actinomyces* was identified histopathologically in a specimen obtained from the middle ear during endoscopic surgery.

Conclusion: This report aims to emphasize that actinomycosis should be considered in the differential diagnosis of patients with chronic suppurative otitis media who do not improve with medical treatment.

Keywords: *Actinomyces*, child, chronic otitis, middle ear, treatment.

INTRODUCTION

Actinomyces is a gram-positive, anaerobic, non-acid-fast, filamentous bacterium.[1] The most commonly affected sites include the head and neck, abdomen, thorax, and pelvis. Actinomycosis may also occur in the middle ear, with more than half of the reported cases affecting patients younger than 15 years.[2,3] Middle ear contamination may occur after mucosal damage caused by trauma, such as tooth extraction or dental caries; the infection can spread from the nasopharynx to the middle ear via the Eustachian tube. Furthermore, *Actinomyces* has been detected with increasing frequency in the nasopharynx of children with recurrent otitis media and can cause infection through mucosal damage. Another route is direct spread from the external auditory canal to the middle ear through damage to the tympanic membrane. In very rare cases, actinomycosis may also occur via hematogenous spread.[3,4]

Actinomyces forms a fibrous, pseudocapsulated inflammatory granuloma characterized by yellow sulfur granules. A careful medical history, a history of trauma, clinical and radiological findings,

and the identification of *Actinomyces* sulfur granules in patient specimens are crucial for diagnosis.[5]

In this report, we present the case of a 4-year-old child with a long history of ear pain and discharge, in whom histopathological examination of a specimen obtained from the middle ear during endoscopic surgery revealed *Actinomyces*.

CASE REPORT

A 4-year-old girl with no known underlying disease developed pain and discharge in the right ear 3.5 months before presentation. She was treated with antibiotics for a presumptive diagnosis of otitis; however, her symptoms did not completely resolve. Her ear pain and discharge improved during antibiotic treatment but recurred after treatment was discontinued. Temporal bone and inner ear computed tomography (CT) was reported as follows: "Loss of aeration in the right mastoid air cells; soft tissue densities compatible with otomastoiditis in the right mastoid air cells and middle ear. The right middle ear ossicles and inner ear structures appear normal. The left ear structures are normal." An endoscopic ear examination performed by an otorhinolaryngologist, with a preliminary diagnosis of cholesteatoma, revealed a lesion perforating the tympanic membrane and extending into the deeper middle ear spaces that was not externally visible. The patient underwent mastoidectomy with excision of the polypoid lesion, and the tympanic membrane was repaired during the procedure. Pathological examination reported "Inflammatory exudate in the middle ear, chronic inflammation, inflammatory granulation tissue, and gram-positive filamentous bacilli arranged in a fine, radial, and branching pattern with sulfur granules in the center, consistent with *Actinomyces* colonies" (Fig. 1, 2).

During our initial physical examination, the patient was in good general condition, and the systemic examination was unremarkable. No pathology was detected on oropharyngeal or ear examination. Laboratory evaluation revealed a white blood cell count of 7320/µL, an absolute neutrophil count of 2850/µL, an absolute lymphocyte count of 3620/µL, a hemoglobin level of 11.8 g/dL, and a platelet count of 274,000/µL. Liver and kidney function tests were normal. The erythrocyte sedimentation rate was 17 mm/hour (range, 0–20), and the C-reactive protein level was 1.61 mg/L (range, 0–8). Chest radiography was normal.

Middle ear actinomycosis was considered, and high-dose intravenous penicillin G therapy (300,000 U/kg/day in 4 doses) was initiated. Abdominal ultrasonography and brain magnetic resonance imaging were performed to assess organ involvement and revealed no abnormalities. Baseline immunological investigations were normal. The treatment regimen consisted of intravenous penicillin G for 2 weeks, followed by oral penicillin V (100,000 U/kg/day in 4 doses) for an additional 3 months. Post-treatment otoscopic examination was normal. The patient's symptoms resolved. The patient has been followed for 9 months after treatment and has had no complaints. Written informed consent was obtained from the patient's legal guardians for the publication of this case report and the accompanying images.

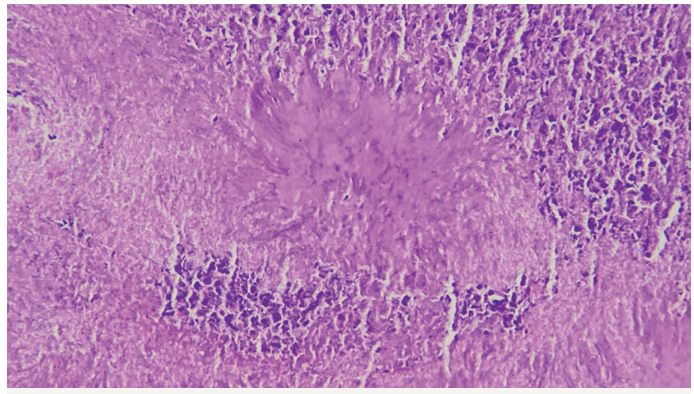

Figure 1. Central sulfur granules formed by a fine, radial, and branching arrangement of gram-positive filamentous bacilli, with *Actinomyces* colonies and inflammatory cells at the periphery (×40).

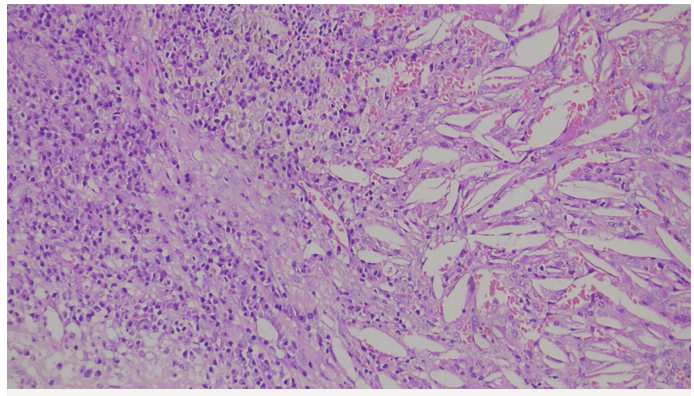

Figure 2. Lymphoplasmacytic inflammation on the left and a foreign body reaction to cholesterol clefts on the right (×40).

DISCUSSION

Middle ear actinomycosis typically presents with recurrent ear discharge resembling chronic suppurative otitis media. A history of acute otitis media may also be present. Mild fever has been reported in approximately 50% of cases, whereas pain is rare. In our case, recurrent ear discharge and ear pain were present, along with a history of acute otitis media. Although hearing loss has been reported in some cases, our patient had no hearing complaints. Because of limited resources, audiometry could not be performed. This is a limitation of our study.

Because otoscopy often fails to provide definitive findings, clinicians may prefer CT imaging. A definitive diagnosis requires isolation and identification of the pathogen by culture or histopathological examination.[5] In our case, the definitive diagnosis was established through histopathological examination. Middle ear swab cultures are generally not helpful for identifying *Actinomyces*, and cultures yield false-negative results in more than 70% of cases.[1] Surgically excised biopsy specimens or purulent material drained from collections are the most appropriate samples for diagnosis.[2] When a surgeon encounters "yellow granules" or unusually inflamed tissue during surgery, an anaerobic culture with prolonged incubation should be requested from the microbiology laboratory.[1]

In a case report by Modi et al.,[4] a 35-year-old patient with a 9-month history of yellow otorrhea was diagnosed with actinomycosis based on biopsy findings. Similarly, Sheikh et al.[6] reported middle ear actinomycosis in a 24-year-old patient with an 8-year history of ear pain and hearing loss. In a study conducted at a tertiary care hospital in Singapore between January 2004 and December 2020, 3 of 10 pediatric patients diagnosed with actinomycosis had ear pain; all but 1 underwent surgical excision, and treatment included penicillin, amoxicillin, or amoxicillin-clavulanate.[7]

Cholesterol granuloma is a foreign body reaction to cholesterol crystals that develops during an inflammatory process, and, as in our case, cholesterol granuloma and *Actinomyces* may sometimes be found together.[8] A study by Erdem showed that *Actinomyces* accompanies cholesterol granuloma in the mandible.[9] Similarly, a study by Maradeix et al.[10] revealed that *Actinomyces* and cholesterol granuloma are found together in the hip. This coexistence may suggest a synergistic relationship between the two conditions. In our case, excision of the polypoid lesion and mastoidectomy promoted aeration and healing by physically removing not only the infected tissue but also the cholesterol granuloma.

Long-term antibiotic treatment should be administered after surgery to achieve disease remission. Depending on disease severity, intravenous penicillin G may be administered for 2 to 6 weeks, followed by oral penicillin V or amoxicillin for 3 to 6 months.[2] Current literature recommends 3 months of treatment for surgically treated patients; however, if the patient has not undergone surgery or has not shown improvement, treatment may be continued for up to 12 months.[1,5] Penicillin V is superior to amoxicillin because of its narrower spectrum of activity, high sensitivity against *Actinomyces*, and the virtually complete absence of resistance. Therefore, penicillin V may be the preferred choice. In patients with penicillin allergy, clindamycin, doxycycline, or fluoroquinolones may be used.[1]

CONCLUSION

Although middle ear actinomycosis is rare, it is a serious infection that is often misdiagnosed or overlooked, primarily because of low culture sensitivity. In our case, a patient with a history of acute otitis media and long-standing ear discharge and pain underwent debridement with a preliminary diagnosis of cholesteatoma and was subsequently diagnosed with actinomycosis on the basis of pathological findings. Middle ear actinomycosis should be considered in patients with chronic otitis that does not respond to treatment.

Ethics Committee Approval: This is a single case report, and therefore ethics committee approval was not required in accordance with institutional policies.

Informed Consent: Written informed consent was obtained from the patient's legal guardians for the publication of this case report and the accompanying images.

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