A Case of Middle Ear Actinomycosis Treated by Non-Surgical Method

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－ ABSTRACT －

Actinomycosis is a chronic suppurative infection by Actinomyces species, which rarely involves the middle ear and seldom diagnosed prior to tympanomastoidectomy. Unlike actinomycosis of the other body part, surgical removal is generally conducted in middle ear actinomycosis. We present the case of a 72-year-old female with 1-month history of otalgia and otorrhea in the left ear. Otoscopic examination revealed voluminous granulation tissue obstructing the ear canal with serous discharge. Excisional biopsy of the granulation tissue revealed actinomycosis. Tympanomastoid surgery was advised to treat presumed residual mastoid disease, but it was refused by the patient. 2 weeks of intravenous antibiotics and 6 months of penicillin was administered, and with a follow-up of 2 years she is free of symptom. Early diagnosis of actinomycosis and proper medication with antibiotics could control the symptoms and disease without surgery in some selected cases. **(J Clinical Otolaryngol 2015;26:248-252)**

KEY WORDS：Actinomycosis · Mastoid · Temporal bone.

Introduction

Actinomycosis is a chronic suppurative infection by Actinomyces species, which are anaerobic, gram-positive filamentous bacteria.[1)] The presentations of most patients with middle ear actinomycosis are similar to those of patients with chronic suppurative otitis media, that is, these nonspecific clinical features result in long delays to diagnose.[2)] Furthermore, it is difficult to find Actinomyces through microbiologic culture because Actinomyces are anaerobic and frequently coexist with other bacteria.[2)]

Actinomyces rarely involves the middle ear and seldom diagnosed prior to tympanomastoidectomy. Unlike actinomycosis of the other body part, surgical removal is generally conducted in middle ear actinomycosis. We present the case of a 72-year-old female with middle ear actinomycosis who was treated non surgically.

논문접수일 : 2015년　8월 17일
논문수정일 : 2015년 10월　7일
심사완료일 : 2015년 11월　6일
교신저자 : 김성희, 41199 대구광역시 동구 아양로 99
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Case Report

A 72-year-old woman presented with an 1-month history of otorrhea and aggravating otalgia in her left ear. There was neither history of trauma to ear, dental manipulation nor otological surgery. The external auditory canal was filled with grayish polypoid soft tissue with serous otorrhea (Fig. 1). The pure tone audiogram revealed four frequency average (500, 1,000, 2,000 and 3,000 Hz) of 85 dB flat mixed hearing loss with poor speech discrimination (0%) in the left ear and 35dB in the right ear (Fig. 2). The hearing loss in the left ear was worsening gradually, but she hadn't had any work-up for her ear by then. On initial microbiologic study, Streptococcus viridans, multi-drug sensitive, was cultured. Bone window computed tomo-

graphic scan showed soft tissue density replacing the middle ear cavity and external auditory canal and total opacification of sclerotic mastoid air cells without evidence of bony destruction (Fig. 3).

The polypoid granulation tissue had a broad stalk on middle ear mucosa and was protruding through the perforated ear drum, thus occupying the ear canal. Excisional biopsy was done under the microscope in the outpatient clinic. Histopathologic examination showed middle ear mucosa infiltrated by lymphocytes, plasma cells, and also adjacent Actinomycoces colonies characterized by coccoid and filamentous forms surrounded by polymorphonuclear leukocytes (Fig. 4). Tympanomastoid surgery was advised to treat presumed residual mastoid disease, but it was refused by the patient due to old age and rapid improvement in symptom after polypectomy. Intravenous antibiotics (amoxicillin sodium 1 g, clavulanate potassium 0.2 gm three times a day) for 2 weeks and 6-month course of antibiotics with Amoxicillin/Clavulanate potassium (Amocla duo (7 : 1)® 875/125 mg, twice a day, Kuhnil

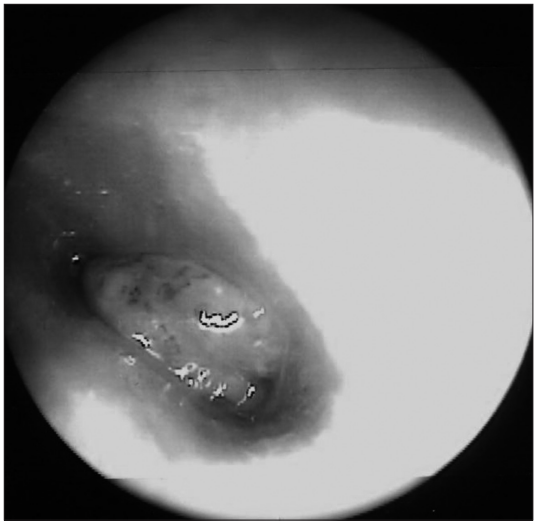

Fig. 1. Initial otoscopic finding of the left ear. Tympanic membrane is replaced by polypoid granulation tissue along with serous otorrhea. The polypoid granulation tissue had a broad stalk on middle ear mucosa and was protruding through the perforated ear drum, thus occupying the ear canal.

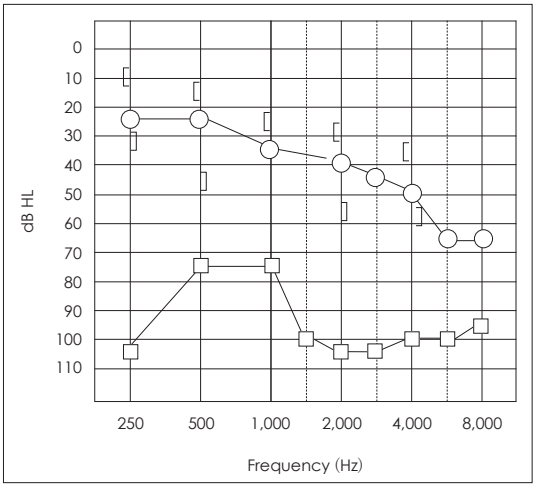

Fig. 2. Pure tone audiometry showed four frequency average (500, 1,000, 2,000 and 3,000 Hz) of 90 dB flat mixed hearing loss in the left ear and 35dB in the right ear.

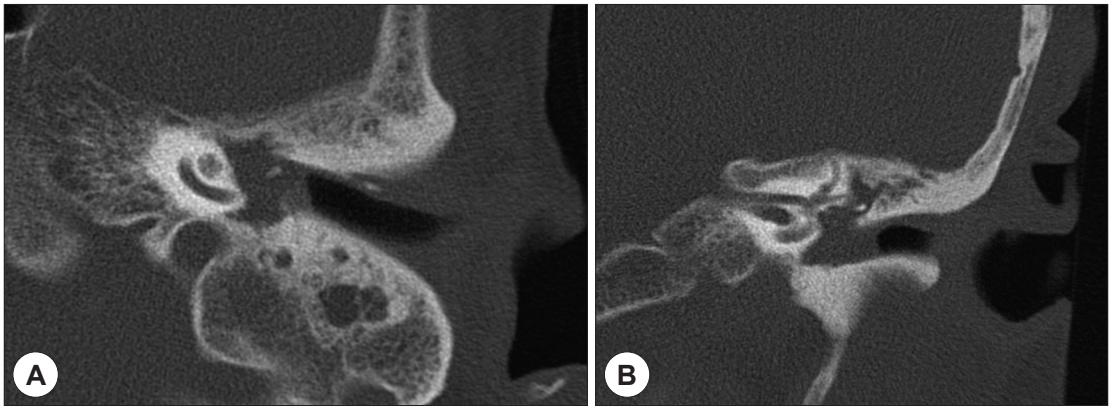

Fig. 3. Temporal bone CT showed soft tissue density occupying the whole middle ear and medial part of ear canal. There is a total opacification of sclerotic mastoid. Ossicle erosion is suspected (A : axial view, B : coronal view).

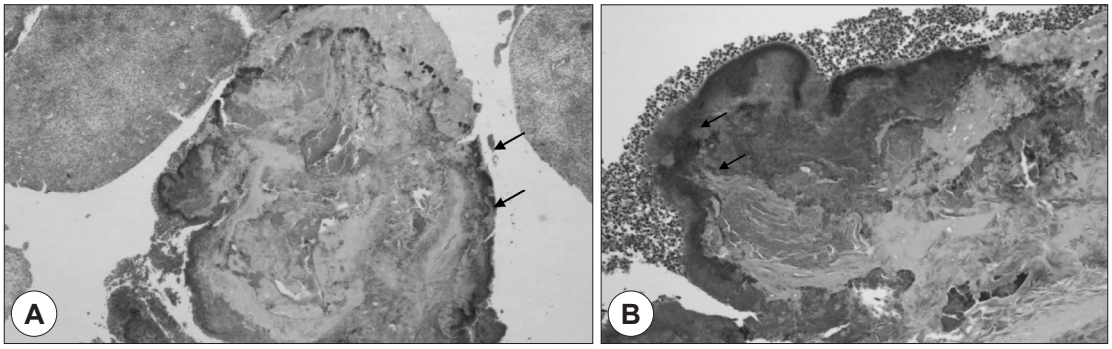

Fig. 4. Histopathologic finding. Histopathologic examination shows middle ear mucosa infiltrated by lymphocytes, plasma cells, and also adjacent Actinomycoces colonies characterized by coccoid and filamentous forms surrounded by polymorphonuclear leukocytes (sulfur granule : arrow) (A : H&E stain ×40, B : H&E stain ×100).

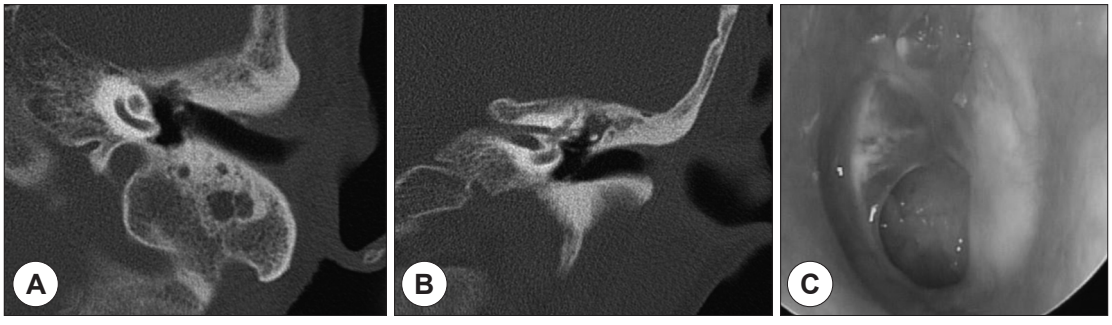

Fig. 5. Follow-up temporal bone CT scan (A and B) and ear drum finding (C) at 3 months post-presentation. Temporal bone CT scan showed well aerated middle ear space, but soft tissue density in sclerotic mastoid cavity (A : axial view, B : coronal view). Otoscopic exam shows dry medium sized perforation of the tympanic membrane.

pharm, Seoul, Korea) were implemented. Follow-up computed tomographic scan after 3 months showed an improvement in middle ear and mastoid air cell (Fig. 5A, B). There was no recurrence of otorrhea and otalgia with a follow-up of 2 years. Tympanic membrane perforation (Fig. 5C) and hearing loss remained.

Discussion

Actinomycosis is a chronic suppurative infection of the cervicofacial region caused by Actinomyces species, which are anaerobic, gram-positive filamentous bacteria.[1)] It is an uncommon infection of the middle ear and is seldom diagnosed prior to tympanomastoidectomy. It was once a fatal disease, however in the antibiotic era, it responds to the antibiotic treatment well.

The presentations of most patients with middle ear actinomycosis are similar to those of patients with chronic suppurative otitis media, that is, these infections are characterized by a prolonged, indolent course.[2,21)] Even though middle ear actinomycosis tends to be refractory to conventional treatment, these nonspecific clinical features result in long delays to diagnose.

Actinomyces are known to be a commensal flora of the oral cavity; hence, it is postulated that entry to the middle ear is most likely the result of seeding from the nasopharynx via the Eustachian tube.[1,2)] Many cases of middle ear actinomycosis report intact bulging ear drum with middle ear granulation tissue, and these are supporting the idea of pathogens are coming upward through Eustachian tube.[3-6)] Direct infections of the external auditory canal or haematogenous spread are possible but less likely alternatives.[1)] In this patient, the patient didn't have any history of ear infection previously, but according to poor pneumatization of mastoid and gradual aggravation of hearing loss in the left

ear, it is presumed that she had a chronic history of ear infection in the left ear. Also considering the old age unlike most reported cases, it can be plausible that Actinomyces could have been introduced in the middle ear through the external auditory canal and preexisted ear drum perforation rather than infection through the Eustachian tube.

Since 1980s, 19 cases in available English literature and 2 cases in Korean literature about middle ear and temporal bone actinomycosis were reported.[1-18] Among them, 13 cases are children below 15 years old[1-3,5,9-16,18] and 9 cases under 10 years old.[1,2,11-16,18] There is a tendency of middle ear actinomycosis being related to somewhat younger age than actinomycosis in the other part of body. It is presumed that the children in those ages are prone to infection through the Eustachian tube. Intact bulging ear drum is frequently reported in middle ear actinomycosis, so if middle ear actinomycosis occurs frequently in young children, it would be difficult to differentiate from regular bacterial acute suppurative otitis media.[3,5]

Furthermore, it is difficult to find Actinomyces through microbiologic culture because Actinomyces are anaerobic and frequently coexist with other bacteria.[2,7] Therefore, all the case reports except 2 cases are confirmed by pathologic result during operation.[7,10] In pathologic exam, Actinomyces form basophilic and eosinophilic colonies with club-shaped filaments radiating into rosette patterns, so called 'Sulfur granules'.[1] So, the identification of Gram-positive filamentous organisms with Sulphur granules is strongly suggestive of actinomycosis.[1] Newer diagnostic techniques such as polymerase chain reaction, 16S rRNA sequencing and mass spectrometry hold promise, but are not routinely used in clinical practice.[1,8]

In our case, luckily the patient had protruding polypoid granulation occupying middle ear space with stalk, so it could be removed quite easily, and actinomycosis could be confirmed by pathologic reports. However, in several reports, excisional biopsy through polypectomy showed just inflammatory polyp not actinomycosis, so the patients needed an operation to get the diagnosis.[1,8,9] Even though excision of granulation is not enough for mastoid ventilation, persistent tympanic perforation could be a mean for producing aerobic conditions in the middle ear. However, at 3 months post- presentation, the temporal bone CT scan shows mastoid haziness, thus long term follow up will be needed because there is a tendency to recur after insufficient treatment.

Among 21 cases reported so far, 2 cases were diagnosed as actinomycosis by biopsy examination, which preceded surgical extraction, and they all had non-surgical treatment with antibiotics and reported good result.[7,10] So far, in middle ear actinomycosis, surgical debridement and proper antibiotics treatment were recommended.[1-4] Especially the need for surgical debridement has been brought up because thick granulation tissue can provide anaerobic environment for the Actinomyces. However in non-severe case with confirmation of actinomycosis through culture or tissue biopsy, medical treatment can be optional. In actinomycosis of other body part, for example in lung, medical treatment alone gives good result.[19] And in cervicofacial actinomycosis, full recovery occurs in most of patients managed by antimicrobial treatment alone.[20] Surgical debridement is often necessary for complete resolution, as this anaerobic species can survive for long periods within poorly vascularized tissues, where antibiotics may not reach therapeutic concentrations.[12]

However, in selected cases with already confirmed actinomycosis and expected good aeration and drainage of inflammation, surgery would not be mandatory. It is possible that early diagnosis may facilitate successful non-surgical treatment of actinomycosis. It is hoped that alert awareness and clinical suspicion in patients with culture-negative infections or intractable inflammation might lead to earlier diagnosis and more success with non-surgical treatment regimens in selected patients with middle ear actinomycosis.

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