Tuberculous otitis media: Report of 2 cases on Long Island, N.Y., and a review of all cases reported in the United States from 1990 through 2003

Lisa M. Chirch, MD; Khalid Ahmad, MD; Warren Spinner, MD; Victor E. Jimenez, MD; Susan V. Donelan, MD; Eric Smouha, MD

Abstract

We report 2 cases of tuberculous otitis media that were diagnosed at Stony Brook University Hospital in New York since 1999. Both patients were women, aged 30 and 31 years. One patient had grown up in Russia, the other was a native-born American who had never left the East Coast region of the United States. Both patients had been symptomatic for many months; one complained of chronic otorrhea, and the other reported otorrhea, hearing loss, and discomfort. Neither patient responded to medical management, and both ultimately underwent surgery. One was diagnosed after surgical pathology revealed acid-fast bacilli on frozen-section analysis. In the other, pathology revealed chronic inflammation and granulomata, but stains were negative and her diagnosis was delayed for almost 2 years. We also review 9 other cases of tuberculous otitis media in the United States that have been reported in the literature since 1990. Our review suggests that the number of cases is rising in areas where tuberculosis is most common—that is, in major U.S. cities. Although 3 of these 9 cases occurred as reactivation disease in immigrants, most might have occurred as a result of local transmission. Clinicians should maintain a high degree of suspicion for tuberculosis in patients with chronic otitis symptoms, particularly those who are at higher risk of exposure to tuberculosis.

Introduction

Tuberculous otitis media (TOM) has become an uncommon diagnosis in the developed world since the introduction of effective antituberculous chemotherapy in the mid-20th century. However, new cases of tuberculosis are reported annually in the United States and worldwide, and at least 8 million people still develop tuberculosis each year; approximately 3 million of these patients die of the disease. In fact, tuberculosis remains the leading cause of death secondary to infectious diseases worldwide in persons older than 5 years.

Considering the increasing number of cases of tuberculosis, including extrapulmonary tuberculosis, that have been diagnosed over the past 2 decades in U.S. urban centers, one would expect a corresponding increase in the number of cases of TOM. However, such has not been the case. TOM is difficult to diagnose, in part because diagnosis requires special media and staining to identify the organism in ear secretions or biopsy specimens. Another reason TOM is difficult to diagnose is because it mimics other conditions, such as chronic bacterial otitis and cholesteatoma, and it is therefore frequently overlooked as a possibility. Untreated TOM can result in permanent, severe sequelae, such as facial paralysis, hearing impairment, and intracranial dissemination of infection. Therefore, early suspicion and a timely diagnosis are of paramount importance.

Since 1990, 9 cases of TOM diagnosed in the U.S. have been reported in the literature; all of these cases occurred in major urban centers. We report 2 new cases of TOM that were diagnosed at Stony Brook University Hospital, a large tertiary care center on Long Island, N.Y., located approximately 60 miles east of Manhattan. In this article, we describe these 2 new cases and we review the 9 previously reported cases.

Case reports

Patient 1. In June 2001, a 31-year-old white woman pre-
Presented to the Department of Otolaryngology with a history of chronic sinus problems since 1999. She reported chronic intermittent nasal drainage and sinus pain. Her symptoms had responded only minimally to various attempts at medical management. She did experience some relief following sinus surgery in 2000.

In the spring of 2001, she began to notice a discharge from her left ear. She denied pain or any notable change in hearing at the time. In addition to sinusitis, her medical history was significant for bronchitis, which had been treated with antibiotics and inhalers. She denied having traveled outside the continental United States or having lived in a shelter or prison, and she was not aware of ever having been exposed to tuberculosis. She did report a positive result on purified protein derivative (PPD) testing. She lived alone on Long Island in Suffolk County, N.Y., but she had previously had a roommate who was a native of Haiti. Since 1991, she had attended a church on Long Island whose members had emigrated from all over the world, particularly from Latin American countries.

The patient was referred to the ENT clinic at Stony Brook University Hospital in June 2001. Examination revealed that a dark-red soft-tissue mass had filled the middle ear. She underwent a mastoidectomy and tympanoplasty at that time for removal of a possible glomus tumor. A biopsy revealed chronic granulomatous inflammation (figure 1, A and B). Stains for acid-fast organisms and fungi were negative.

Several months after surgery, the patient again noted left ear drainage, which had stained her hair and pillowcase. She visited the ENT clinic several times, but again did not respond to medical management. In July 2003, the ear drainage was sampled in the clinic. Smears for acid-fast bacilli (AFB) were again negative, but 4 weeks later, *Mycobacterium tuberculosis* complex was isolated in culture and identified by nucleic acid hybridization (AccuProbe; Gen-Probe; San Diego).

![Figure 1. Patient 1. Immediately following surgery, granuloma and chronic inflammation are detected in the tympanic membrane specimen (A) and the middle ear contents (B). Two years later, CT scans (C and D) demonstrate erosion of the left mastoid process with opacification of mastoid air cells, a bony defect in the base of the left petrous bone, and a soft-tissue density that fills the middle ear cavity.](image-url)
The patient was admitted to Stony Brook University Hospital in September 2003 when culture results became available, and she was placed in isolation pending further evaluation and initiation of treatment with isoniazid, rifampin, pyrazinamide, ethambutol, and vitamin B$_6$. Computed tomography (CT) of the head and sinuses revealed erosion of the left mastoid process with opacification of mastoid air cells. A bony defect in the base of the left petrous bone was also identified, and a soft-tissue density filled the middle ear cavity. Erosion of the petrous bone and coalescence of mastoid air cells were present. The ossicles were intact, as was the VIIth cranial nerve (figure 1, C and D).

Chest films on admission were read as normal, but a chest CT revealed tiny nodular opacities in the posterior segment of the right upper lobe with bronchiectasis. Three sputum specimens for acid-fast staining were negative, but one of three sputum cultures later grew *M tuberculosis* that was sensitive to all of the aforementioned drugs. Audiometry revealed a mild-to-moderate conductive hearing loss on the left and normal function on the right. The patient continued antituberculous drug therapy for a total of 1 year.

**Patient 2.** A 30-year-old woman who had been born in Russia presented to the ENT clinic in March 1999 with a 4- to 5-month history of right ear drainage, fullness, and hearing impairment. She had earlier been treated with 10 days of amoxicillin/clavulanate and antibiotic eardrops, but her symptoms failed to resolve. She denied any constitutional symptoms or sick contacts, and her medical history was unremarkable. The patient had been vaccinated with bacille Calmette-Guérin (BCG) as a child. She had moved from Moscow to Huntington, N.Y., on Long Island in 1995. While in Russia, she had been trained as a microbiologist. During the preceding few years, she had traveled to the West Coast of the United States and to Europe. She denied tobacco, alcohol, and illicit drug use.

Findings on the physical examination were unremarkable with the exception of the right ear; a fleshy, bloody mass was present in the canal. Her complete blood count, chemistries, liver function, and urinalysis were within normal limits, as was a CT of the chest.

A mastoidectomy and tympanoplasty were performed to remove a possible cholesteatoma. Intraoperative exploration detected a 40% tympanic membrane perforation and a purulent discharge in the canal. Biopsy of the middle ear contents showed extensive necrotizing granulomatous inflammation, and stains on frozen section were positive for AFB (figure 2). Culture became positive for *M tuberculosis* complex 1 month later (AccuProbe), and sensitivities available soon thereafter revealed the presence of a pansensitive organism. The patient was treated with 4-drug therapy—isoniazid (plus vitamin B$_6$), rifampin, pyrazinamide, and ethambutol—for 1 year, and her symptoms resolved completely.

**Literature review**

We conducted a MEDLINE search to identify cases of TOM using the search terms *tuberculosis*, *otitis*, *middle ear*, and *tuberculous otitis media*. Our analysis included all cases diagnosed in the United States that had been reported in the literature during or after 1990. It is important to note that although all cases of suspected or confirmed tuberculosis should be reported to local departments of health, not all reported cases are submitted for publication, so the true incidence of TOM is unknown. The number of cases reported in this analysis likely underestimates the true number of cases that had occurred since 1990.

We found that 9 cases of TOM in the U.S. had been reported in the literature since 1990 (table). Three of these cases had been diagnosed at New York Hospital in New York City and reported in 1995, 5 were seen at the University of Illinois Medical Center at Chicago (1995, 1996).
and 2003), and 1 at Memorial Sloan-Kettering Cancer Center in New York City (2000).\(^5\)

Combined analysis of our 2 cases of TOM and the 9 previously reported cases revealed that these patients (6 males and 3 females) ranged in age from 5 months to 69 years (mean: 32 yr); 7 patients were younger than 35 years. Four patients were immigrants. The time to diagnosis ranged from 1 month to more than 2 years (mean: ~8 mo; median: ~4 mo). Only 4 patients had positive smears or stains for AFB (another had an AFB-positive chest nodule), but 8 eventually had positive cultures. Four patients had documented pulmonary involvement. Nine patients had a positive PPD test, and 2 had previously been vaccinated with BCG, and they did not undergo PPD testing. Eight patients had temporal bone involvement demonstrated on CT, and 4 had central nervous system involvement. Seven patients experienced severe complications or permanent sequelae, particularly conductive hearing loss; 1 child experienced developmental delay.

**Discussion**

Mandatory reporting of tuberculosis to local health departments was instituted in 1953. In the literature, scattered descriptions and case reports have appeared since the 19th century. Clinical TOM was first documented in 1853, and the organism was first identified in otic discharge in 1883.\(^6\) From 1962 to 1984, TOM accounted for only 0.06% of all cases of chronic otitis media.\(^7\) Although more recent statistics on TOM are not available, our review suggests that the number of cases of TOM is rising in areas where tuberculosis is most often diagnosed—namely, in major U.S. cities. Although some cases of TOM have occurred as a reactivation disease among immigrants, most may have occurred as a result of the local transmission of tuberculosis, as was the case with our patient 1.

The pathogenesis of TOM is postulated to involve one of three mechanisms: (1) aspiration of mucus through the eustachian tube, (2) hematogenous dissemination from other tuberculous foci, or (3) direct implantation through the external auditory canal and a tympanic membrane perforation.\(^8\) The history of chronic sinusitis and bronchiectasis in our patient 1 suggests that inadequate clearance of tubercle bacilli in respiratory secretions may have played a role. Whether local factors present in the middle ear are required for establishment of the infection is unknown. Some 40 to 50% of patients with TOM have no evidence of tuberculosis elsewhere.\(^6\)

Abnormal chest films were seen in 58% of cases reviewed by Skolnik et al in 1986.\(^7\) Our review of cases reported since 1990 showed that only 36% of patients (4/11) had documented pulmonary involvement, but CT was not routinely performed. In our patient 1, the diagnosis of pulmonary tuberculosis was based on CT after the patient had already been diagnosed with TOM and after her initial chest film had been read as negative. This raises the issue that perhaps CT of the thorax, in addition to serial sputum examinations for AFB in smears and cultures, should be performed on patients who are diagnosed with TOM to rule out with certainty concomitant pulmonary tuberculosis.

As revealed by our review of cases, the clinical presentation of TOM can be variable and insidious; few patients present with the classic symptoms of painless draining ears, hearing loss, and facial paralysis.\(^4,6\) Intense suppuration and multiple tympanic membrane perforations have been described in some textbooks, but contemporary cases differ in that the presentation reflects a more chronic and indolent disease, and multiple perforations are rare. Our 2 patients presented in this way.

Hearing loss is usually acute in onset and disproportionate to the extent of disease. Conductive hearing loss is the most common type, appearing in approximately 90% of cases.\(^6\) Examination of the infected ear may reveal pale-yellow granulation tissue on a thickened, hyperemic tympanic membrane. A tumorlike mass was present in both of our patients. Perforations usually occur in areas of granulation tissue.\(^6\) Discharge may be serous or, if bacterial superinfection has occurred (79% of cases), thick and mucoid or purulent.\(^6\) Destruction of the ossicles may be visible through perforations.\(^6\)

TOM may be clinically indistinguishable from the usual forms of chronic otitis media, although it is exceedingly less common. As such, biopsy with staining and culture should be considered for patients who have not responded to usual therapeutic measures. The differential diagnosis of TOM is wide-ranging and includes both infectious and noninfectious etiologies. Among the latter are Wegener’s granulomatosis, cholesteatoma, lymphoma, and histiocytosis X.\(^6\) Bone erosion on CT is quite common in both TOM and cholesteatoma. Temporal bone involvement in histiocytosis X may masquerade as chronic otitis media. Otic involvement in Wegener’s granulomatosis may present as otitis media with effusion secondary to eustachian tube obstruction by luminal granuloma. Infectious etiologies that mimic TOM include fungal infections (e.g., histoplasmosis and blastomycosis), congenital or acquired syphilis, nocardiosis, chronic bacterial otitis, and necrotizing external otitis.\(^6\)

The diagnosis of TOM relies heavily on clinical suspicion, as microbiologic testing is often delayed and unreliable. Patients with known or suspected tuberculosis and chronic otitis should be evaluated for TOM by an ear examination, PPD test, and chest films. Based on the small series of cases reported herein, we propose that all patients with chronic otitis media undergo PPD testing, especially those in urban centers or other areas with significant numbers of tuberculosis cases. External canal smears are positive in as many as 20% of cases, and cultures are positive in 5 to 35% of cases.\(^4\) One would expect to see higher figures with intraoperative specimens; our patient 2 had positive stains on intraoperative frozen-section analysis, and 8 of the 11
patients (73%) in our series eventually had positive cultures obtained from ear drainage or intraoperative specimens, despite the fact that only 4 (36%) had a positive smear or stain for AFB. Inoue et al suggested that polymerase chain reaction (PCR) testing provides a more rapid and reliable diagnosis of TOM. They reported the case of a 26-year old woman with TOM whose otorrhea smear was negative but whose PCR assay was positive. The use of PCR to diagnose TOM has not been reported elsewhere, but the experience of Inoue et al supports the idea that a diagnosis cannot rest on staining of secretions alone; it more than likely requires a biopsy with staining and culture.

<table>
<thead>
<tr>
<th>Pt.</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>31 yr</td>
<td>30 yr</td>
<td>3 yr</td>
<td>19 mo</td>
<td>44 yr</td>
<td>61 yr</td>
<td>69 yr</td>
</tr>
<tr>
<td>Time to Dx</td>
<td>&gt;2 yr</td>
<td>4 to 5 mo</td>
<td>2 to 3 mo</td>
<td>1 mo</td>
<td>15 mo</td>
<td>Several months</td>
<td>1 yr</td>
</tr>
<tr>
<td>Smear/stain for AFB</td>
<td>–</td>
<td>+</td>
<td>+</td>
<td>Caseating granuloma</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Culture (from otorrhea or surgery)</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Pulmonary involvement</td>
<td>CT+, sputum culture+</td>
<td>–</td>
<td>CXR+</td>
<td>CXR+</td>
<td>–</td>
<td>Unknown</td>
<td>CXR+</td>
</tr>
<tr>
<td>PPD</td>
<td>+</td>
<td>N/A†</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Bone Involvement‡</td>
<td>+</td>
<td>–</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>–</td>
</tr>
<tr>
<td>CNS Involvement§</td>
<td>–</td>
<td>–</td>
<td>+</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>+</td>
</tr>
<tr>
<td>Surgical intervention**</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Location</td>
<td>Stony Brook</td>
<td>Stony Brook</td>
<td>Chicago</td>
<td>Chicago</td>
<td>Chicago</td>
<td>Chicago</td>
<td>Chicago</td>
</tr>
<tr>
<td>Sequela</td>
<td>Hearing loss</td>
<td>–</td>
<td>Hearing loss</td>
<td>–</td>
<td>–</td>
<td>Hearing loss</td>
<td>–</td>
</tr>
</tbody>
</table>

* Patients 4, 5, and 7 were diagnosed in the U.S., but their country of origin was not specified.  
† Patients 2 and 8 had been vaccinated with bacille Calmette-Guérin; no PPD (purified protein derivative) testing was performed.  
‡ Bone involvement was detected by CT.  
§ CNS involvement included facial paralysis, ataxia, tuberculomas, shunt placement, vertigo, and tinnitus.  
** Surgical intervention included mastoidectomy alone or tympanoplasty plus mastoidectomy.  

Key: P.R. = Puerto Rico; AFB = acid-fast bacilli; CXR = chest x-ray.
**Table. Summary of the 11 cases of tuberculous otitis media reported in the U.S. from 1990 through 2003**

<table>
<thead>
<tr>
<th>Pt.</th>
<th>Country</th>
<th>Age</th>
<th>Time to Dx</th>
<th>Location</th>
<th>Involvement</th>
<th>Surgical intervention</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>U.S.</td>
<td>31 yr</td>
<td>&gt;2 yr</td>
<td>Stony Brook</td>
<td>+ + + + + + + + + +</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Russia</td>
<td>30 yr</td>
<td>4 to 5 mo</td>
<td>Brook</td>
<td>– + – + + + + + +</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>U.S.</td>
<td>3 yr</td>
<td>2 to 3 mo</td>
<td>Chicago</td>
<td>– – – + – – – +</td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>?</td>
<td>19 mo</td>
<td>1 mo</td>
<td>Chicago</td>
<td>+ + + + + + + + +</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>U.S.*</td>
<td>44 yr</td>
<td>Several</td>
<td>Chicago</td>
<td>+ – – – – – – –</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>U.S.*</td>
<td>61 yr</td>
<td>1 yr</td>
<td>N.Y.C.</td>
<td>+ + + + + + + + +</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>India</td>
<td>69 yr</td>
<td>3 mo</td>
<td>N.Y.C.</td>
<td>+ + + + + + + + +</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>Pakistan</td>
<td>21 yr</td>
<td>4 mo</td>
<td>N.Y.C.</td>
<td>+ + + + + + + + +</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>Philippines</td>
<td>33 yr</td>
<td>1.5 yr</td>
<td>N.Y.C.</td>
<td>+ + + + + + + + +</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>? U.S.*</td>
<td>33 yr</td>
<td>1.5 yr</td>
<td>N.Y.C.</td>
<td>+ + + + + + + + +</td>
<td></td>
</tr>
</tbody>
</table>

**Key:**

- **§** CNS involvement included facial paralysis, ataxia, tuberculomas, shunt placement, vertigo, and tinnitus.
- **‡** Bone involvement was detected by CT.
- **†** Patients 2 and 8 had been vaccinated with bacille Calmette-Guérin; no PPD (purified protein derivative) testing was performed.
- ***** Patients 4, 5, and 7 were diagnosed in the U.S., but their country of origin was not specified.

CT is the best imaging modality available for diagnosing TOM and identifying mastoid involvement. A soft-tissue density may be visible in the middle ear cavity with opacification of mastoid air cells. There may be erosion of the mastoid or petrous bone, destruction of ossicles, or VIIth cranial nerve involvement. Widespread bony destruction on CT, as was seen in 8 of the cases we reviewed, in the absence of any clinical signs of aggressive infection should suggest a mycobacterial process. However, it is important to note that some of these changes seen on CT may also be attributable to postoperative changes in patients who have undergone mastoidectomy.

Treatment of TOM is the same as that currently recommended for extrapulmonary tuberculosis. The recommended duration of therapy is at least 6 months; in most of the cases reviewed herein, patients were treated for at least 1 year. Surgical intervention is warranted if complications arise, but surgery in the absence of antituberculous therapy can result in the development of fistulae and a failure of suture lines to heal.

In conclusion, the progression of TOM can be variable and insidious. In addition to its rarity, factors that contribute to the difficulty in diagnosing TOM include the unreliability of smears, stains, and cultures in identifying AFB and/or *M tuberculosis* in middle ear specimens. Delayed diagnosis and treatment can result in severe complications and permanent sequelae. Our findings underscore the importance of maintaining a high degree of clinical suspicion of tuberculosis in patients with chronic otitis media, particularly those whose PPD test is positive.

### References


### Further reading