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Case Report

Chronic Tuberculous Otomastoiditis: A Case Report

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Worldwide, tuberculosis is a widespread disease, with 8.7 million new cases occurring annually. Its etiologic agent, Mycobacterium tuberculosis, essentially causes pneumonia. However, this organism affects the middle ear in rare cases, accounting for 0.04–0.09% of all chronic middle ear otitis cases in Western countries. In this report, we describe the case of a young woman affected by tuberculosis of the middle ear. In our experience, empiric therapy was not beneficial. Adequate treatment was possible only after obtaining a specific diagnosis through a difficult process requiring surgical sampling for culture examination. We consider surgical sampling to be mandatory in all cases of chronic otitis media that do not respond to prolonged systemic and local therapies.

KEYWORDS: Mycobacterium tuberculosis, otomastoiditis, otitis media, biopsy

INTRODUCTION

Mycobacterium tuberculosis infection remains among the most insidious diseases worldwide. A report in 2012 by the World Health Organization estimated that almost one-third of the global population carries a latent form of tuberculosis [1]. In addition, significant data on mortality associated with M. tuberculosis infection revealed 1.4 million deaths among 8.7 million new cases detected annually.

Regarding extra-pulmonary M. tuberculosis infection, 95% of cases involve the head–neck region (larynx, lymph nodes, tonsils, pharynx, oral cavity, salivary glands, and external and middle ears) [2]. At the beginning of the last century, the incidence of tuberculous external and middle ear infection in industrialized countries ranged from 3% to 5%. This percentage massively decreased to 0.04–0.09% for all chronic otitis media cases with the introduction of tuberculosis-specific antibiotic therapies. Further, the incidence of pulmonary infection decreased to 10–20% [2, 3]. In Italy and other European countries, only a few tuberculous middle ear infection cases have been described.

Here we describe our experiences with a rare case of tuberculosis of the middle ear in an adult patient. Based on our experience, we suggest that the above-mentioned percentages are underestimations, given the difficulties associated with the diagnosis of tuberculous otitis.

CASE PRESENTATION

A 38-year-old woman presented with chronic right middle ear otitis. At the time of admission to the hospital, she signed an informed consent for medical and surgical treatment and for the use of her personal data. She was born and currently lived in Tuscany, Italy; worked as a freelance journalist; and had no recent history of travel abroad, contact with possible infection sources, or chronic and systemic pathologies. Her clinical history contained no suggestive elements, except for a pulmonary infection of unknown origin 5 years earlier that was successfully treated with ceftriaxone. Despite local and systemic antibiotic therapies for 2 months, she experienced tympanic membrane perforation and otorrhea. She additionally reported unilateral hearing loss, vertigo, and right-sided facial nerve paralysis (House–Brackmann grade V).

During our initial evaluation, clinical features included a yellowish, jelly-like auricular secretion; severe mixed hearing loss; peripheral right-sided facial paralysis with no fever; and a good general condition. Otoscopy revealed a posterior tympanic perforation and a jelly-like pultaceous secretion in the external auditory canal. Systemic oral and parenteral antibiotic therapies were prescribed as follows: amoxicillin with clavulanic acid (Augmentin; GlaxoSmithKline, Verona, Italy) at 2 g/day for 8 days, ceftriaxone (Rocefin; Roche, Milano, Italy) at 1 g/day for 7 days, and clarithromycin (Klacid; BGP products, Roma, Italy) at 1 g/day. The following corticosteroids were administered: betamethasone (Bentelan; Sigma Tau Industrie Farmaceutiche Riunite, Roma, Italy) at 1.5 mg/day intramuscularly for 7 days and prednisone (Deltacortene; Bruno Farmaceutici, Roma, Italy) at 25 mg/day orally for 5 days. Local therapy was with ceftazidime (Glazidim; Glaxo Smith Kline, Verona, Italy). A computed tomography (CT) scan of the middle ear revealed a wide area of mastoid cell opacification on the right side in a well-pneumatized mastoid bone, widespread opacity of the tympanic

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antrum, and a tympanic cavity in the ipotympanic region; notably, the ossicular chain was clear and morphologically undamaged. Imaging data indicated chronic right otomastoiditis. An ear secretion sample was negative for most common bacteria, fungi, and mycobacteria. Audiometry revealed mixed hearing loss (Figure 1).

The patient was hospitalized and underwent magnetic resonance imaging, which revealed dehiscence of the second facial bony canal area continuous with endotympanic phlogistic material that entirely filled the tympanic cavity and mastoid cells omolaterally (Figure 2). A direct vestibular examination and stabilometry revealed right-sided ipo-reflexivity. Laboratory examinations and specific antibody analyses were performed to screen for antibodies against herpes simplex virus, varicella zoster virus, cytomegalovirus, Epstein–Barr virus, human immunodeficiency virus, and Borrelia burgdorferi as well as autoantibodies, but they yielded negative results. Leukocytosis and count alterations were not observed; however, increases in fibrinogen levels and erythrocyte sedimentation rates were detected.

During recovery, antibiotic therapy was modified as follows (all intravenously administered): piperacillin/tazobactam (Tazocin; Pfizer Limited, Sandwich, UK) 4.5 g×3/day and vancomycin (Vancomicina Hikma; Hikma Italia Spa, Pavia, Italy) 500 mg×4/day for 3 days, replaced with linezolid (Zyvoxid; Pfizer Italia, Latina, Italy) 600 mg×2/day for 5 days, cefepime (Maxipime; Bristol-Myers Squibb S.r.l., Roma, Italy) 2 g×2/day for 5 days, and ciprofloxacin (Ciproxin; Bayer Spa, Milano, Italy) 500 mg×2/day for 5 days. No change was observed after 5 days of therapy. Infection-negative chest radiography results and a Quantiferon test positive for tuberculous disease were received 2 weeks later. While waiting, canal wall-up tympanoplasty was performed to drain the infection and obtain biopsy samples. During surgery, we observed granulation tissue and widespread purulent material throughout the mastoid cavity and aditus, with a small dehiscence of the mastoid cortical bone emitting purulent secretion. Facial nerve neurostimulation evoked no response. Finally, PCR (Polymerase Chain Reaction)-based mycobacterial screening of the collected sample was positive for M. tuberculosis complex DNA, allowing us to make the correct diagnosis. The patient began specific systemic therapy comprising ethambutol, rifampicin, and isoniazid. After a 6-month follow-up, conductive hearing loss and peripheral facial palsy (House–Brackmann grade V) were unchanged, but otorrhea was no longer present.

DISCUSSION

We reported the rare case of a patient with chronic tuberculous otomastoiditis. This infection is more frequent in women (1.36-fold) than in men, in medical doctors and paramedics (4.3-fold) than in the general population, and in people near affected patients or carriers [4, 5]. Infection frequently reaches the tympanic cavity via the external auditory canal, or through rhinopharyngeal inspiration via the Eustachian tube, or hematogenous spread (79%) [6].

Wallmer [7] first described clinical features of tuberculous otitis in 1953, including painless otorrhea, multiple tympanic membrane perforations, granulation tissue in the tympanic cavity and mastoid, and bone necrosis with progressive conductive hearing loss and facial palsy. Symptoms have become more variable and polymorphic over time. Bacterial superinfection may cause purulent serous otorrhea.

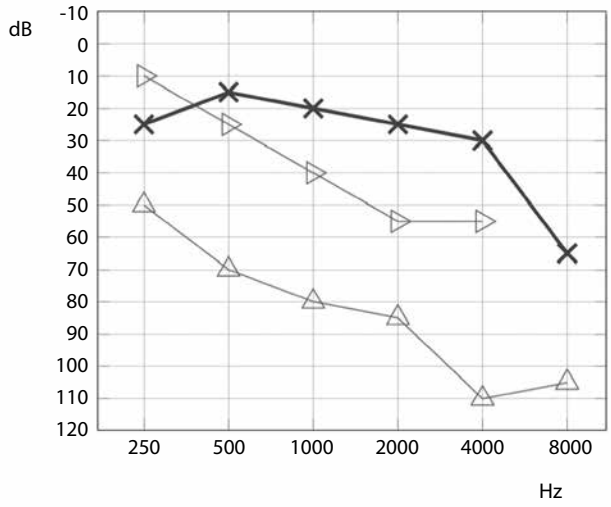

Figure 1. Audiogram data collected on admission to hospital. Red triangles indicate the air threshold of the right ear, red arrows indicate the bone threshold of the right ear, and blue crosses indicate the air threshold of the left ear

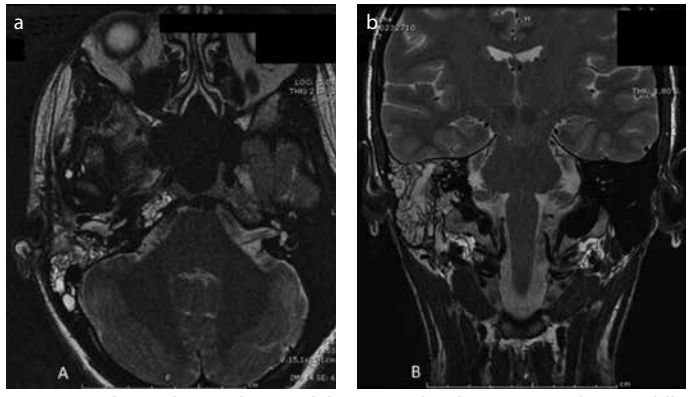

Figure 2. a, b. Axial (a) and coronal (b) T2-weighted MR images show middle ear and mastoid inflammation

Cervical and pre- and retro-auricular lymph nodes may be involved in the spread of infection [2, 8].

Complications mainly occur with delayed diagnosis and include retro-auricular fistulae, meningitis, petrous pyramid osteomyelitis, cerebral and cerebellar abscesses, and peripheral facial palsy (occurring in 15–40% of cases). Functional recovery is strictly related to the time lapse between palsy and specific therapy initiation. Complete recovery may occur if therapy begins within 5 days; the likelihood of recovery decreases over time, with permanent palsy expected after 2 months.

Useful diagnostic tools for M. tuberculosis of the ear include the bacteriological examination of ear secretions, although this is positive in only 5–35% of cases. The tuberculin (PPD or Mantoux) and Quantiferon tests have very low specificity due to their potential for positive results in cases of previous infection or vaccination [9]. CT imaging studies could detect inflammatory tissue within the tympanic cavity and mastoid cells with or without bone erosion, and in chronic infection, they may identify ossicular resorption and mastoid sclerosis. Similar findings, however, may be detected with chronic otitis and cholesteatoma [6]. As all these noninvasive diagnostic tools are nonspecific,

the surgical exploration of the tympanic cavity and mastoid cells is mandatory [6, 7]. Canal wall up tympanoplasty is useful for removing the necrotic bone and granulation tissue and collecting analytical samples. Finally, histopathological examination is the most effective diagnostic tool for chronic tuberculous otomastoiditis [9].

We note that the role of surgery with therapeutic intent remains controversial and that the effectiveness of surgery has not been definitively proven. Some authors oppose this practice unless complications are present and there is no indication of ossicular chain reconstruction, at least prior to symptom resolution [9, 10]. On the other hand, Kwon et al. [11] recently demonstrated that higher rates of dry ear are achieved when surgery preceded chemotherapy than with chemotherapy alone. Varty et al. [12] further asserted that the combination of surgery and medical therapy improves prognosis. In our opinion, surgery plays a minor role in the therapeutic approach to tuberculous otitis media. We conclude that without very specific and prolonged antibiotic therapy, infection cannot be eradicated and any type of surgical effort would be useless.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

Peer-review: Externally peer-reviewed.

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Erratum

In the article by Salam et al., entitled "Comparison of Scalar Location and Insertion Depth of Cochlear Implant Electrode Implanted Through the Round Window Versus Cochleostomy Approach" (Int. Adv. Otol. 2013; 9:(1) 30-37) that was published in the January 2013 issue of the Journal of International Advanced Otology, submission date of the manuscript was erroneously published as "16 November 2013" instead of "16 November 2012". The submission date of the manuscript has been corrected as "16 November 2012".