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CASE REPORT

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Tuberculous otitis media case report: Clinical manifestation, evaluation and management

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ABSTRACT

Tuberculous otitis media, caused by *Mycobacterium tuberculosis*, is a chronic bacterial infection that affects the middle ear structures. Diseases with polymorphous and insidious clinical features can make diagnosis difficult and delay treatment. Herein we report a case of chronic tuberculous otitis media associated with cholesteatoma, highlighting the main factors that led to the diagnosis. Further, this report aimed to highlight the particularities in the clinical and surgical treatment of the case, given the association with chronic cholesteatomatous otitis media. This descriptive research, with qualitative analysis, was carried out at the Department of Otorhinolaryngology of the Hospital de Reabilitação de Anomalias Craniofaciais in São Paulo, Brazil. This study highlights the importance of investigating tuberculosis as a differential diagnosis of granulomatous diseases of the middle ear and temporal bone, given the risk of serious and irreversible complications as the disease progresses.

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KEYWORDS
Tuberculosis; cholesteatoma; chronic mastoiditis

Introduction

Tuberculous otitis media is a rare suppurative granulomatous infection of the middle ear and temporal bones. When comparing remote epidemiological data to the present day, a drastic reduction in the number of cases is observed due to antituberculous therapy. At the beginning of the twentieth century, 3–5% of cases of chronic suppurative otitis media had a tuberculous etiology; currently, such cases have reduced to 0.05–9% today. However, due to its morbidity and mortality, tuberculosis remains a public health problem, both nationally and worldwide [1–3].

Owing to its unfavorable socioeconomic conditions, Brazil shows the highest incidence of tuberculosis among American countries, with an average of 31.6 cases per 100,000 inhabitants in 2020. In 2019, 66,819 new cases and 4500 deaths were registered due to the disease. Another aggravating factor in this situation is the high rate of abandonment of treatment, approximately 65% of cases, which spreads the contamination of the bacillus. Thus, despite advances in treatment, tuberculosis remains the most common infection worldwide [4,5].

This pathology, in 90% of cases, tends to have pulmonary tuberculosis as its primary focus; the high incidence and rate of treatment abandonment directly reflects the incidence of the disease in the country. It is estimated that due to the lack of suspicion and difficulty in etiological confirmation, cases of tuberculous otitis media are often underdiagnosed, leading to underestimation of the disease in Brazil [1,3].

Patient information

A 26-year-old female patient, from the city of Fartura, in the state of São Paulo, visited the otorhinolaryngology outpatient clinic of the Hospital de Reabilitação de Anomalias Craniofaciais da Universidade de São Paulo, complaining of continuous and daily bilateral otorrhea for approximately one year. In addition, she complained of bilateral hearing loss associated with the condition, which was worse on the right side. She reported frequent

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episodes of otorrhea in childhood that resolved with advancing age. She had no other associated otologic and sinonasal complaints. Further, she denied any comorbidities or previous use of medications.

Clinical findings

On physical otoscopic examination, the tissue presented with a polypoid, hyperplastic appearance, preventing visualization of the tympanic membrane, and thick otorrhea, which was aspirated during the examination (Figure 1).

Consequently, oral treatment with ciprofloxacin and clindamycin was initiated for 14 days, along with the use of ear drops (ciprofloxacin and betamethasone) for 14 days, and an audiological evaluation was requested. The patient returned after 14 days with audiometry showing mild conductive hearing loss on the right (40 dB) and moderate mixed conductive loss on the left (60 dB), according to the classification of the World Health Organization. Moreover, despite clinical treatment, previous otoscopic alterations still persisted.

Diagnostic assessment

Therefore, the following tests were recommended to investigate granulomatous diseases: chest radiography, VDRL, HIV serology, and tuberculin test, in addition to magnetic resonance imaging (MRI) of the ear. The tuberculin skin test revealed a strongly reactive result of 15 mm.

An incisional biopsy was then performed in both ears, with a search for acid-resistant bacillus (BAAR). The BAAR test was positive on the left and negative on the right side, and showed vascular proliferation associated with a chronic granulomatous inflammatory process with intermingled neutrophils. Therefore, the diagnosis of chronic tuberculous otitis media was confirmed (Figure 2).

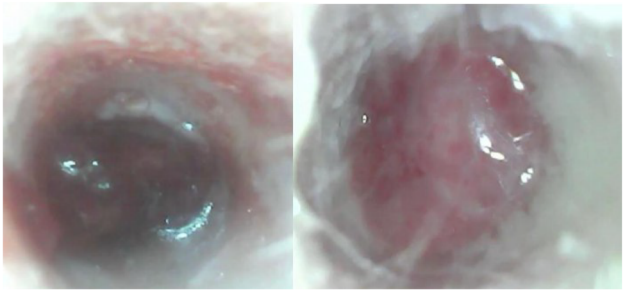

Figure 1. Polypoid tissue obliterating the external auditory canal of the right and left ear, respectively.

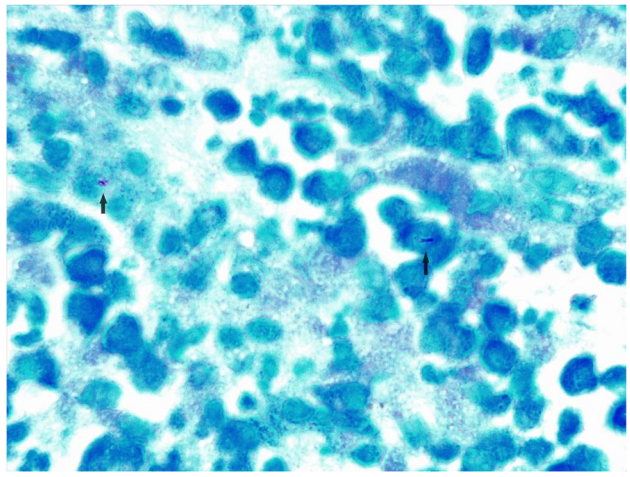

Figure 2. Histological image with hematoxylin-eosin coloration demonstrating the chronic inflammatory process, with bacillus (black arrows).

Therapeutic intervention

At the end of the recommended treatment (2 months of rifampicin, isoniazid, pyrazinamide, and ethambutol, followed by 4 months of rifampicin and isoniazid), bacillus research in residual polypoid tissue was repeated, which showed bilateral negative results. The patient reported improvement of otorrhea and hypoacusis of the left ear after treatment; however, she had intermittent episodes of otorrhea on the right side. Upon audiometric examination, she showed improvement in hypoacusis, with mild conductive hearing loss on the right (20 dB) and mild mixed conductive loss (35 dB) on the left side. On otoscopic examination, the right ear showed hyperplastic polypoid tissue obstructing the auditory canal, and the left ear showed thickening of the tympanic membrane with attic erosion and the presence of scaly lamellar tissue. Therefore, MRI of the mastoids was performed. MRI revealed the presence of material with T2 and FLAIR hypersignal, and an intermediate signal on T1, located inside the mastoid cells, middle ear, and external ear bilaterally, showing areas of diffusion restriction, which did not enhance after administration of contrast (Figures 3 and 4). Therefore, surgical treatment was indicated for the resolution and improvement of the condition. Subsequently, left tympanomastoidectomy was performed, opting for an open cavity.

During the intraoperative period, desquamative lamellar tissue was visualized in the middle ear and mastoid and was referred for anatomopathological analysis, which showed a nonspecific chronic inflammatory process with cholesterol crystals and areas compatible with an associated cholesteatoma.

Thirty days after the surgical procedure, the patient reported cessation of otorrhea and denied any worsening of auditory acuity.

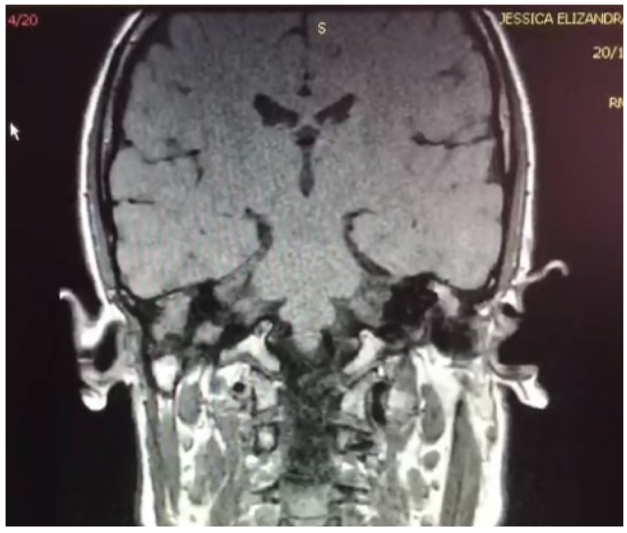

Figure 3. Temporal bone magnetic resonance imaging in coronal section, with areas of hypointensity in the topography of the mastoids, in T1 sequence.

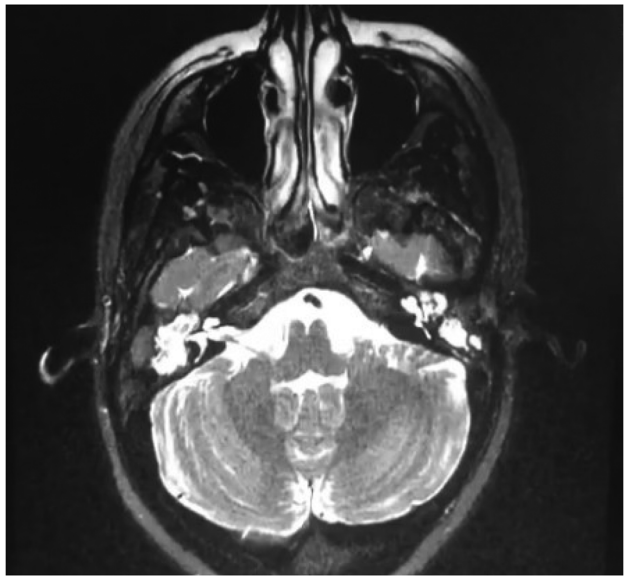

Figure 4. Magnetic resonance imaging in axial view, with areas of hyperintensity in the mastoids, in T2 sequence, axial view.

Despite resolution of the suppurative condition, the patient presented with a tympanic membrane with retraction and attic erosion on the left, suggesting the presence of cholesteatoma in the left ear. Therefore, after six months, the patient underwent radical left mastoidectomy (Figure 5). During the intraoperative procedure, desquamative lamellar tissue was visualized in the topography of the middle ear, with abundant inflammatory tissue in the attic and ossicular chains, which was referred for histopathological analysis. Tissue analysis was compatible with cholesteatoma.

Three months after the operation, the patient still had otorrhea on the left side, with hyperplastic and

Figure 5. Removal of scaly lamellar tissue during intraoperative period.

exudative tissue in the cavity on otoscopic examination, despite local cleaning and use of topical anti-inflammatory and antimicrobial medication. Therefore, serial cauterization of the hyperplastic tissue was performed with 70% trichloroacetic acid, with insidious improvement of the suppurative condition. Three cauterizations were necessary, and a hypothesis of tuberculosis recurrence was suggested. Therefore, a new tuberculin skin test was requested, with a PPD value of 15 mm. The results obtained were referred to the department of infectology for further evaluation. However, owing to the serological scar, the value was considered normal. On follow-up, the patient remains stable and has not reported any new suppurative episode with the left ear. After cauterization and use of ear drops with an acidifying solution, the patient underwent otoscopy which revealed no new polyps in the middle ear.

Follow-up and outcomes

The patient remains under follow-up at this department, with resolution of the suppurative condition, and otoscopy showing healing and epithelialization of the mastoid cavities (Figure 6). Because of hearing loss, she was referred to use a bilateral individual sound amplification device.

Given the case, we discussed with the patient the possibility of publishing the clinical case as a case report for scientific purposes. The patient agreed, and the work was submitted to the hospital's Department of Education and Research's Ethics and Research

Figure 6. Right ear otoscopy six months after surgery. The contralateral ear had the same appearance.

Committee, where it was approved (publication number: 5.682.457). The informed consent form was completed and signed by the patient for publication. Additionally, the patient signed the Publication Agreement, granting permission for the publication. It's important to note that no conflicts of interest have been reported by the authors.

Discussion

Mycobacterium tuberculosis bacillus can affect the middle ear and temporal bone in a primary or secondary manner, the latter being considered the most common. The primary form initially affects the middle ear and may spread to other neighboring structures by contiguity [3]. The secondary form, comprising 90% of cases, usually presents initially as pulmonary tuberculosis, extending to the middle ear and temporal bone by hematogenous dissemination [1].

The progression of the disease can start as a single or multiple foci, depending on the dissemination route (hematogenous, *via* the Eustachian tube, *via* the external auditory canal, or *via* the intracranial route, close to the temporal bone) [2]. The mucous tubercle promoted by the bacillus can develop, producing necrosis and ulceration, and culminating in the formation of caseous exudate that invades air spaces. Thus, granulation tissue can spread at the tympanic membrane, ossicular chain, and mastoid cells, promoting osteitis and tympanic perforations [5].

The clinical manifestation is usually varied; however, unilateral mucopurulent otorrhea is found in most patients. It may initially present with tympanic membrane thickening, which generally evolves into single or multiple tympanic perforations. Polyps are common in the middle ear and external auditory canal, making it difficult to visualize the tympanic membrane. Otalgia is less frequent. With disease progression, patients can present with peripheral facial paralysis, retroauricular abscess, and temporal bone osteitis [3,6].

Hearing impairment usually presents a conductive pattern, initially due to the accumulation of exudate in the middle ear and the formation of granulation tissue. However, extension of the inflammatory process can lead to necrosis and erosion of the ossicular chain, otic capsule, and mastoid cells, resulting in moderate to severe sensorineural hearing loss [2,6].

Due to the clinical presentation being similar than chronic suppurative otitis media, failure in the standard treatment of this condition, associated with a more aggressive evolution of the condition, should suggest the differential diagnosis of tuberculous otitis. Thus, in patients undergoing surgical treatment for chronic otitis media, the material found should be subjected to histopathological analysis, with the aim of ruling out granulomatosis such as tuberculous otitis media [7].

The diagnosis begins with clinical suspicion; however, there is a need for additional tests to elucidate the condition. Some radiological findings may be suggestive of tuberculous etiology, such as mastoid filling well pneumatized by soft tissue content, mastoid cortical sclerosis, ossicular chain destruction, and middle ear opacification [2,3].

A tuberculin test can also be performed, indicating previous contact with the bacillus. However, it does not distinguish a patient from an individual infected by the agent. Thus, sputum bacilloscopy for acid-fast bacillus (BAAR) and aural secretion culture can also help in diagnosis, despite their low sensitivity [1].

Histopathological study, although variable, is considered the most reliable method for diagnostic confirmation. The finding of Langerhans cells, caseous necrosis, and specific granulation, associated with the clinical manifestation, can elucidate the diagnosis of the disease [3]. However, in most cases, therapeutic failure and disease aggressiveness lead to suspicion of tuberculous etiology [2].

The treatment is performed clinically with antituberculosis chemotherapy, whose basic regimen lasts six months. The initial use of rifampicin, isoniazid, pyrazinamide, and ethambutol for two months, followed by rifampicin and isoniazid for four months, is considered a national protocol for controlling extrapulmonary tuberculosis in the country. In cases of treatment failure, such therapy may be extended

for 18–24 months [8]. In the case presented in this report, such a clinical regimen was duly followed, with six months of clinical treatment. However, the condition was refractory, suggesting the possibility of extending clinical treatment or surgical intervention [9].

Surgical treatment is indicated in the presence of clinical treatment failure or complications, such as subperiosteal abscess, facial paralysis, labyrinthitis, or central nervous system involvement. In addition, mastoidectomy is performed when there is an association with cholesteatoma [1]. Thus, the surgical indication in this report is supported by the literature, given the associated cholesteatomatous condition.

Conclusion

In conclusion, the present case report highlights the importance of the differential diagnosis of granulomatous diseases of the middle ear when there is refractoriness to the treatment of chronic otitis media. Therefore, appropriate treatment including proper clinical and surgical management protocols are required, which can reduce the risk of sequelae and improve the quality of life of affected patients.

Patient perspective

In relation to the treatments performed, the patient in question had full confidence in the team, being well-informed about the seriousness of the disease and potential sequelae, despite the clinical and surgical treatment. After undergoing the treatments, the patient was very satisfied with the outcome, experiencing an improvement in the quality of life following the cessation of otorrhea episodes in both ears.

Disclosure statement

No potential conflict of interest was reported by the authors.

Informed consent

Written informed consent for publication was obtained.

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