Case Report

Insights Journal Of Surgery And Clinical Case Reports

Bone Osteomyelitis Secondary To Tuberculous Otitis Media. A Pediatric Case Report

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Author Contributions:
This work has been conceived by Venegas, M. P. and López de Heredia, M. The experiments were designed by Venegas, M. P.; López de Heredia, M.; León, X.; De Juan, J.; Boronat, S.; Diez, S. and Orús, C. All authors read the manuscript and have been involved in the critical discussion of this work. Finally, Venegas, M. P. and López de Heredia, M. wrote the paper.

Received Date: 09 Aug 2023
Accepted Date: 25 Aug 2023
Published Date: 31 Aug 2023

Citation:
María del Prado Venegas Pizarro. Bone Osteomyelitis Secondary To Tuberculous Otitis Media. A Pediatric Case Report. Insights Journal Of Surgery And Clinical Case Reports 2023.

1. Abstract

1.1. Background: Tuberculous otitis media (TOM) is a rare cause of chronic suppurative infection of the middle ear and mastoid in developed countries. Diagnosing tubercular otitis media requires a high index of suspicion even in the absence of pulmonary tuberculosis. Demonstration of acid-fast bacilli (AFB) in the ear discharge is difficult and requires complementary tests for diagnosis.

1.2. Case Presentation: We describe the case of a 3-year-old girl with left-ear otorrhea refractory to usual treatment. Computed tomography revealed occupation of the tympanic and mastoid cavities, bone erosion, insufflation, and periosteal reaction in the left temporal bone. Magnetic resonance revealed contrast enhancement in the tympanic cavity and a soft-tissue mass in the mastoid-petrous-squamous temporal bone area. T2 cochlear hyperintensity was partially absent. The differential diagnosis included chronic mastoiditis, osteomyelitis, Langerhans cell histiocytosis and tumours. A biopsy yielded a diagnosis of a rare and potentially life-threatening temporal bone osteomyelitis caused by Mycobacterium tuberculosis complex.

1.3. Conclusions: In the presence of recurrent otitis that is resistant to the usual treatments in the pediatric age group, a differential diagnosis with tuberculous otitis is necessary to avoid diagnosis delays that can lead to further hearing damages. Treatment resolved the lesions after 6 months except for fibrosis at the level of the cochlea. Therapeutic options to restore hearing are presented.

2. Keywords:
Osteomyelitis, Otitis Media, Tuberculous, Otorrhea, Hearing

3. Background

Tuberculous otitis media (TOM) is a rare cause of chronic suppurative infection of the middle ear and mastoid. It was first described by Jean Louis Petit in the 18th century [1]. Its incidence is very low, accounting for only 0.04% of all cases of chronic suppurative otitis media in developed countries. Diagnosis is often delayed because TOM is easily confused with other acute or chronic middle ear conditions [2]. Rare and potentially life-threatening complications of TOM are osteomyelitis of the temporal bone and permanent hearing loss or cophosis. Rapid and accurate diagnosis is required to urgently start treatment and reduce morbidity and mortality [3]. We present the case of a 3-year-old female pediatric patient with TOM, cophosis, and temporal bone osteomyelitis.

4. Case Presentation

We describe the case of a 3-year-old girl who consulted for bilateral chronic otorrhea without otalgia that resembled a bilateral benign chronic otitis. However, the left side otorrhea was refractory to usual medical treatment with beta-lactams and derivatives along with oral and topical corticosteroids. In the otoscopic examination of the left ear, oedema with granulation tissue was observed in the external auditory canal, and central eardrum perforation was observed in the tympanic membrane, ruling out signs of chronic cholesteatomatose otitis media. Blood parameters and

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chest X-ray results were both normal, ruling out systemic pathology and pulmonary infection. A computed tomography (CT) scan of the petrous bone revealed occupation of the tympanic and mastoid cavities, bone erosion, insufflation, and periosteal reaction in the left temporal bone (Figure 1a-1b). In addition, a magnetic resonance imaging (MRI) exam with intravenous contrast was carried out, which revealed contrast enhancement in the tympanic cavity and a soft-tissue mass in the mastoid-petrous-squamous area of the temporal bone. Normal T2 hyperintensity of the cochlea was partially absent (Figure 2a-2b), which indicated localised infection in the temporal bone.

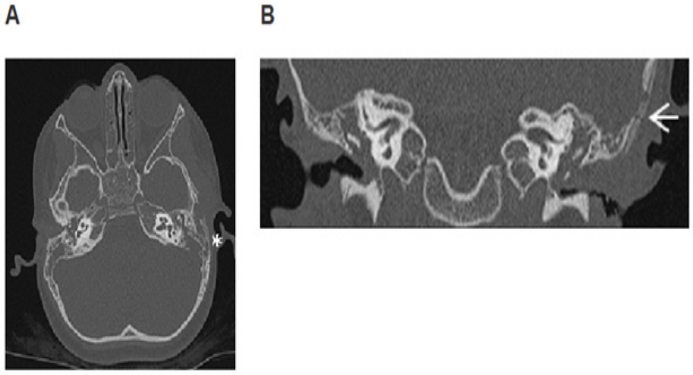

Figure 1: Computed tomography (CT) scans of the petrous bone. **A**. Coronal computer tomography. Occupation of the tympanic and mastoid cavities, bone erosion, insufflation, and periosteal reaction in the left temporal bone is indicated by a white arrow. **B**. Transverse computer tomography image showing a left side occupation of the tympanic and mastoid cavities (white asterisk). Bone extensive cortical bony destruction involving left petrous, squamous, and mastoid bone.

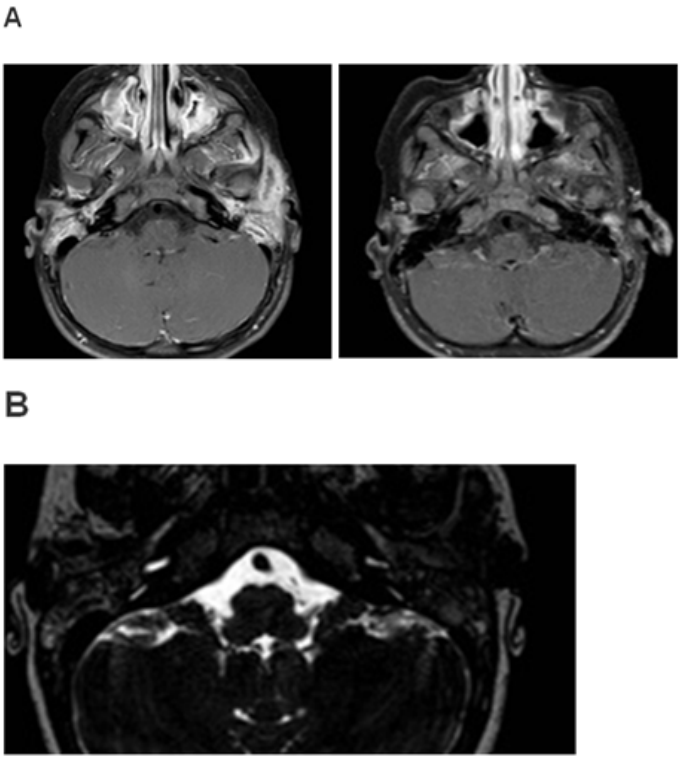

Figure 2: Magnetic resonance imaging (MRI). **A**. T1 sequence. Contrast enhancement in the tympanic cavity and a soft-tissue mass in the mastoid-petrous-squamous area of the temporal bone. **B**. T2 sequence. Hyperintensity of the cochlea is partially absent.

In our setting, the differential diagnosis included chronic mastoiditis, osteomyelitis, Langerhans cell histiocytosis and tumors. A craniotomy was performed in collaboration with a neurosurgeon with the patient under general anesthesia, and a large and deep biopsy was performed of bone and soft tissues. The diagnosis was confirmed as temporal bone osteomyelitis secondary to TOM. The microbiological study of biopsy showed growth of a strain of Mycobacterium tuberculosis complex sensitive to the usual antituberculostatic treatment. Treatment was started with pyrazinamide, isoniazid, and rifampin at pediatric weight-based doses for 2 months, followed by isoniazid and rifampin for 13 months. Tuberculin tests were performed on family members. Only the father tested positive but was clinically asymptomatic – in his chest X-ray, only some nonspecific adenopathies were visualised. Despite being asymptomatic, his positive test result was classified as latent tuberculosis, and he started treatment with rifampicin and isoniazid 300 mg/15 mg, 2 tablets every 12 hours for 3 months. The father tested negative after accomplishment of treatment.

Girl's otorrhea resolved after 9 months of treatment, which was confirmed by magnetic resonance imaging. By then, her ear lesions had disappeared at the level of the middle ear, mastoid and temporal bone, except for the absence of intracochlear permeability, possibly due to the fibrosis caused by the infection (Figure 3). As a sequela, the patient developed permanent hearing loss or cophosis in the affected ear. It was not possible to place a cochlear implant in the affected ear to restore binaurality due to the fibrosis in the cochlea caused by the infection. To avoid the head shadow effect of the ear affected by cophosis and to help localise the sound, it was decided to place an integrated osteosystem with an attached external processor.

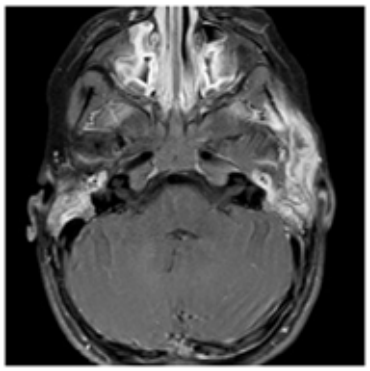

Figure 3: Magnetic resonance imaging (MRI) after 9 months of treatment. T1 sequence. No evidence of ear lesions can be detected.

5. Discussion

Tuberculosis is a major infectious disease that involves mainly the lungs

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and lymph nodes. Tuberculosis of the middle ear is an uncommon and frequently misdiagnosed form of tuberculosis (accounting for only 0.04% of all cases of chronic suppurative otitis media in the UK) [4,5]. In most cases, tuberculosis is pulmonary, and primary manifestations as otitis are rare in developed countries 6].

Tuberculosis of the middle ear can occur by hematogenous or lymphatic spread or by extension into the middle ear cleft through the Eustachian tube [1,5,6] due to vascularization of the mastoid bone in young children. One of the most frequent complications is osteomyelitis of the temporal bone, which is a rare but very aggressive disease. It should be recognised early and treated urgently to reduce morbidity and mortality risks. The pediatric patient had no pulmonary symptoms, and her chest X-ray was normal. Her only symptom was left ear otorrhea refractory to medical treatment. Diagnosing tubercular otitis media requires a high index of suspicion even in the absence of pulmonary tuberculosis. Demonstration of acid-fast bacilli (AFB) in the ear discharge is difficult.

Diagnostic suspicion was based on CT and MRI studies, followed by a large and deep biopsy of the lesioned temporal bone by the neurosurgeon. Microbiological analysis of the biopsied material yielded the definitive diagnosis of temporal bone osteomyelitis. Treatment should start immediately to minimise sequelae and comorbidities and avoid hearing impairment. In children, it is important to maintain the plasticity of the auditory pathway and binaurality if possible, so that they can interact with their environment and learn without hindrance. Binaurality is the ability to analyse and process auditory signals through the two ears simultaneously, so that such signals can later be integrated at the level of the central nervous system, allowing information to be extracted from the environment where the patient is. This is essential to be able to obtain stereophony and correctly perceive sounds in noisy environments.

In the case of the 3-year-old female patient, MRI performed after 6 months of treatment showed absence of bone lesions except for fibrosis in the cochlea. As a sequela, there was cophosis of the affected ear, but due to cochlear fibrosis it was impossible to place a cochlear implant. To aid in sound localisation and avoid the head shadow effect, a soft band osseointegrated implant was placed. This system does not provide binaurality. The sound perceived by the processor is transmitted in the form of vibrations through the skull bone to the contralateral cochlea, that of the normal ear. This system complements the auditory process although it does not restore it, avoids the head shadow effect, and improves the perception of the world in 360º and noisy environments.

6. Conclusions

Chronic pediatric otorrhea refractory to medical treatment is a very infrequent cause of temporal bone osteomyelitis. This serious infection requires a high index of suspicion, rapid and accurate diagnosis, and urgent treatment. In developed countries, tuberculous otitis should be considered in the differential diagnosis of recurrent otitis in children, regardless of whether there is no tuberculous history of the family. The diagnosis is difficult to make in the case of otorrhoea, and it is necessary to resort to a biopsy of the lesions caused by the infection. Injuries to the temporal bone and mastoid can cause irreparable hearing loss. In the case of temporal bone involvement with associated total hearing loss, the placement of an osseointegrated implant may be a good solution to improve the patient's hearing.

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