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CASE REPORT

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Tuberculous otitis media masquerading as malignancy: a diagnostic challenge

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Abstract

Background: Primary tuberculous otitis media is rare in the paediatric age group, and its neuro-otogenic complication of involvement of cerebellopontine angle in a child is very unusual. Tuberculosis should always be considered as a rare but possible aetiology for such neuro-otogenic lesions.

Case presentation: We report a case of a 13-year-old female patient who presented with left ear discharge and mass-like lesion on otoscopy. High-resolution computed tomography (HRCT) temporal bone showed erosion of petrous temporal bone, external auditory canal and ossicles. Contrast-enhanced MRI (CEMRI) revealed peripherally enhancing hetero-intense lesion epicentred in the petrous and mastoid part of left temporal bone extending into the left cerebellopontine angle and external auditory canal. Homogenously enhancing soft tissue was seen in the left occipital condyle with sigmoid sinus thrombosis and cervical lymphadenopathy. There was also a single enhancing left temporal lobe lesion. Radiological and clinical assessment was suggestive of malignant aetiology. However, biopsy revealed tuberculosis and anti-tubercular therapy (ATT) was initiated. Interval imaging showed an adequate response to treatment.

Conclusions: Tuberculous otitis media often masquerades as malignancy on clinical and imaging assessment.

Keywords: Tuberculous otitis media, Temporal bone tuberculosis, Petrous temporal bone, Contrast-enhanced magnetic resonance imaging (CEMRI), Case report

Background

Extra pulmonary tuberculosis refers to tuberculosis affecting regions other than the lungs. Tuberculosis affects head and neck regions in 10% of cases, out of which cervical lymphadenopathy is the most common presentation [1]. Tuberculous otitis media (TOM) is rare, comprising 0.04% of chronic otitis media (COM) cases [2]. Primary TOM is even more uncommon, and only a few paediatric cases have been reported in the English literature [3].

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Plain radiography is rarely used nowadays for the evaluation of COM due to the availability of advanced imaging modalities. HRCT due to its superior spatial resolution is used for evaluating ossicles and bony confines of the middle ear. But since it has low contrast resolution, HRCT cannot be used for differentiating between various aetiologies of middle ear opacification such as long-standing infection/inflammation, neoplasms, haemotympanum or vascular anomalies. MRI is useful in indeterminate and complicated COM cases for evaluation of the inner ear, integrity of facial canal and intracranial complications since it has excellent contrast resolution [4].

Tuberculosis (TB) of the middle ear commonly presents with features of COM and is usually suspected late when there is no improvement with conventional treatment [5]. Here, we present a 13-year-old female patient with chronic ear discharge for 6 months which was suspected

as malignancy and subsequently diagnosed with temporal bone tuberculosis. There are no case reports of TOM in the paediatric population with intracranial extension into the cerebellopontine angle. Here, TOM presented as a mass in the external auditory canal mimicking malignancy of the temporal bone.

Case presentation

A 13-year-old female patient presented to the otorhinolaryngology department with complaints of left ear discharge for 6 months. She did not have any constitutional symptoms of loss of weight or appetite, evening rise of temperature or chronic cough. There was no family history of tuberculosis. On examination of the left ear, a red granular mass was seen completely occupying the external auditory canal obscuring the tympanic membrane. The post-aural region was unremarkable. Enlarged, mobile, firm, non-tender left level V and level II lymph nodes were palpable. These symptoms were not relieved with topical and systemic antibiotics given from another hospital. Hence, in view of clinical suspicion of malignancy, contrast-enhanced MRI of the temporal bone with corroborative HRCT temporal bone was performed.

HRCT temporal bone showed erosion of sigmoid sinus plate, destruction of petrous temporal bone and posterior wall of external auditory canal with associated extra-axial collection along the petrous temporal bone. There was an erosion of handle of malleus, body and long process of incus as well as thinning of left facial canal. Imaging diagnosis of osteomyelitis left petrous temporal bone with otitis externa, otitis media and mastoiditis was considered (Fig. 1).

Contrast-enhanced MRI revealed a T1 hypo-intense, T2 hetero-intense, peripherally enhancing lesion epicentred in the petrous and mastoid part of the left temporal bone which had extension towards the left cerebellopontine angle, indenting the lateral aspect of left cerebellar hemisphere. Anteroinferiorly, it was extending into the external auditory canal causing its obstruction and expansion. Anteromedially, extension along the left eustachian tube was also seen (Fig. 2 A). The soft tissue in the external auditory canal, left petromastoid air cells and along left cerebellopontine angle showed restricted diffusion, predominantly along the periphery with the corresponding drop on ADC images (Fig. 3 A and Fig. 3 B). There was an erosion of the left sigmoid sinus plate with the associated attenuated calibre and thrombosis of the left distal sigmoid sinus (Fig. 3 C and Fig. 3 D). A homogenously enhancing lesion was also seen in the left occipital condyle with communication with the prior mentioned lesion (Fig. 2 B & 4 A). Enlarged homogenously enhancing left level V cervical lymph nodes and left intraparotid lymph nodes were noted (Fig. 4 B & 2 B). An ill-defined T1 iso-intense, T2 hypo-intense lesion with peripheral hyper-intense rim, having no evidence of restricted diffusion on diffusion-weighted images and showing ring enhancement was noted in the left temporal parenchyma which was abutting the tentorium cerebelli with surrounding perilesional oedema (Fig. 4 B).

Imaging differentials of rhabdomyosarcoma of the middle ear with brain metastasis and Langerhans cell

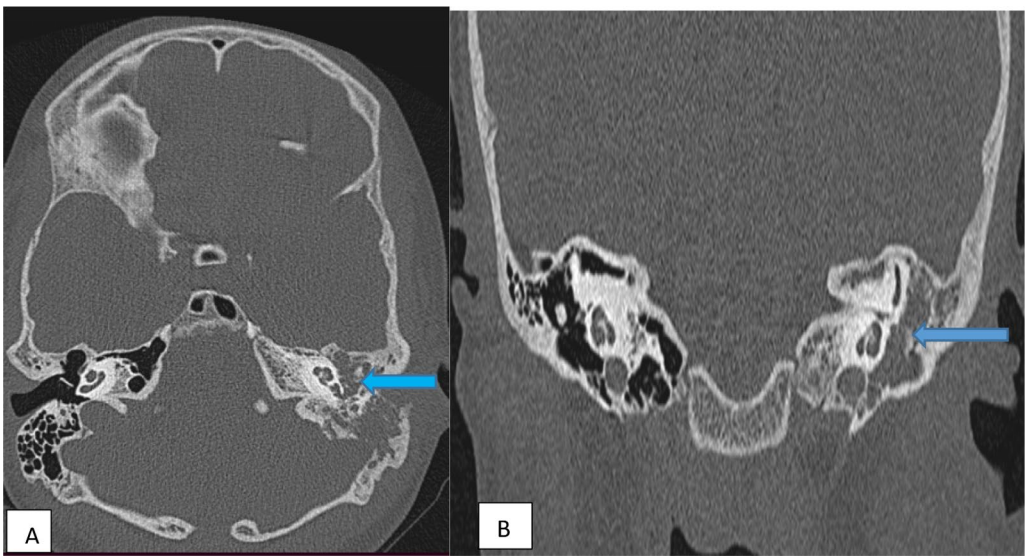

Fig. 1 HRCT temporal bone axial section (**A**) and coronal section (**B**) showing destruction of left petrous temporal bone with erosion of ossicles (blue arrows)

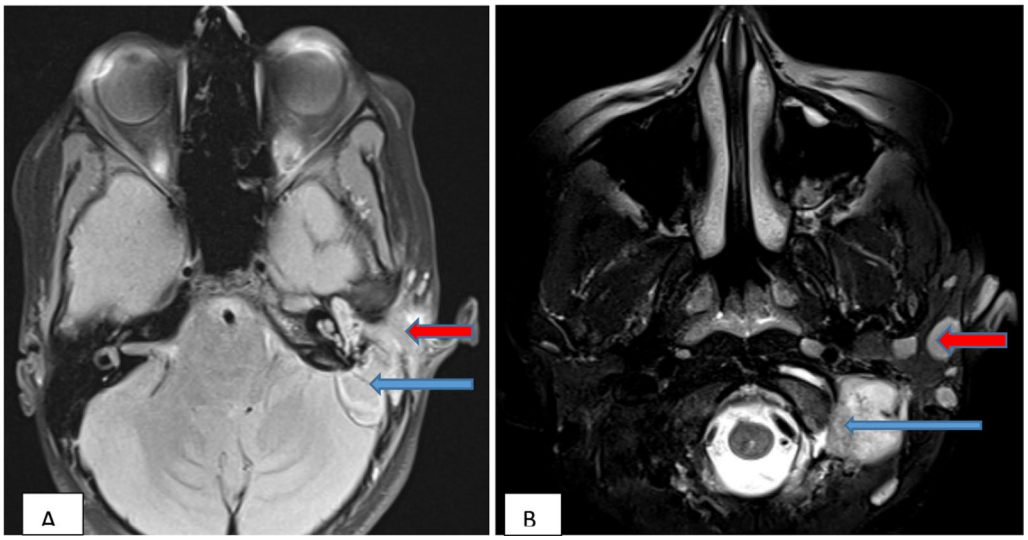

Fig. 2 Axial sections of MR image at base of skull shows: **A**) T1 fat-suppressed image: T1 hypo-intense soft tissue lesion in left external auditory canal (red arrow); extension of soft tissue along left cerebellopontine angle and indenting left cerebellar hemisphere (blue arrow). **B**) T2 fat-suppressed image: Prominent left intraparotid lymph nodes (red arrow) with T2 hyper-intense soft tissue lesion in the left occipital condyle (blue arrow)

histiocytosis (LCH) were considered with less likely differential of benign diseases like cholesteatoma and tubercular infection.

A biopsy was taken from the lesion in the external auditory canal and the specimen was sent for histopathological examination. Histopathological examination (HPE) showed granulomatous inflammation consistent with tuberculosis with inflamed granulation tissue and marked pseudoepitheliomatous hyperplasia (Fig. 5). Acid-fast bacilli were seen on Ziehl–Neelsen stain. Sputum and gastric aspirate were negative for acid-fast bacilli in this patient. A chest radiograph was also done to rule out pulmonary TB, which was normal.

The patient was started on antitubercular drugs. In this case, the intensive phase of ATT was for 2 months with isoniazid, rifampicin, pyrazinamide and ethambutol after which the patient had significant clinical improvement with complete resolution of the mass in the external auditory canal and a dry ear, although there was a moderate degree of residual conductive hearing loss. The maintenance phase was started with isoniazid, rifampicin and ethambutol for 10 months instead of 4 months because of intracranial involvement. Interval contrast-enhanced MRI following 6 months of initiation of drug therapy showed interval resolution of brain parenchymal lesion with significant interval reduction in soft tissue in left petrous and mastoid and left cerebellopontine angle. There was near total resolution of soft tissue infiltration in the left external auditory canal, eustachian tube and left occipital condyle with a reduction in enlarged cervical lymph nodes. But there was persistent thrombosis of left sigmoid sinus with its attenuated calibre (Fig. 6). There were no adverse and unanticipated events during the course of treatment to date.

Discussion

Temporal bone tuberculosis is rare and is usually secondary to pulmonary, nose and laryngeal infection [6]. In a study by Akkara S.K. et al. [7], tuberculous otitis media accounted for only 6 out of 211 cases of tuberculosis occurring in the otorhinolaryngeal region. Painless otorrhoea, pale granulation tissue, multiple tympanic membrane perforations, facial nerve palsy and severe hearing loss are the most common symptoms and signs of middle ear tuberculosis [2].The rarity, lack of disease specific symptoms or signs along with high false negative cultures make it difficult to diagnose this condition early [8].

Complete opacification of the tympanic cavity and mastoid air cells with soft tissue, no scutum erosion, thickening of the external auditory canal and ossicular destruction in the absence of cholesteatoma are the various features which help in differentiating tubercular otitis media from chronic otomastoiditis. These findings are not pathognomonic of TB [8, 9]. MRI was done primarily because we suspected malignancy with differentials such as rhabdomyosarcoma and Langerhans cell histiocytosis. In a case report by Niemczyk K et al. [10], tuberculoma mimicked cerebellopontine angle tumour, post-BCG vaccination in a case of tuberculous otitis media.

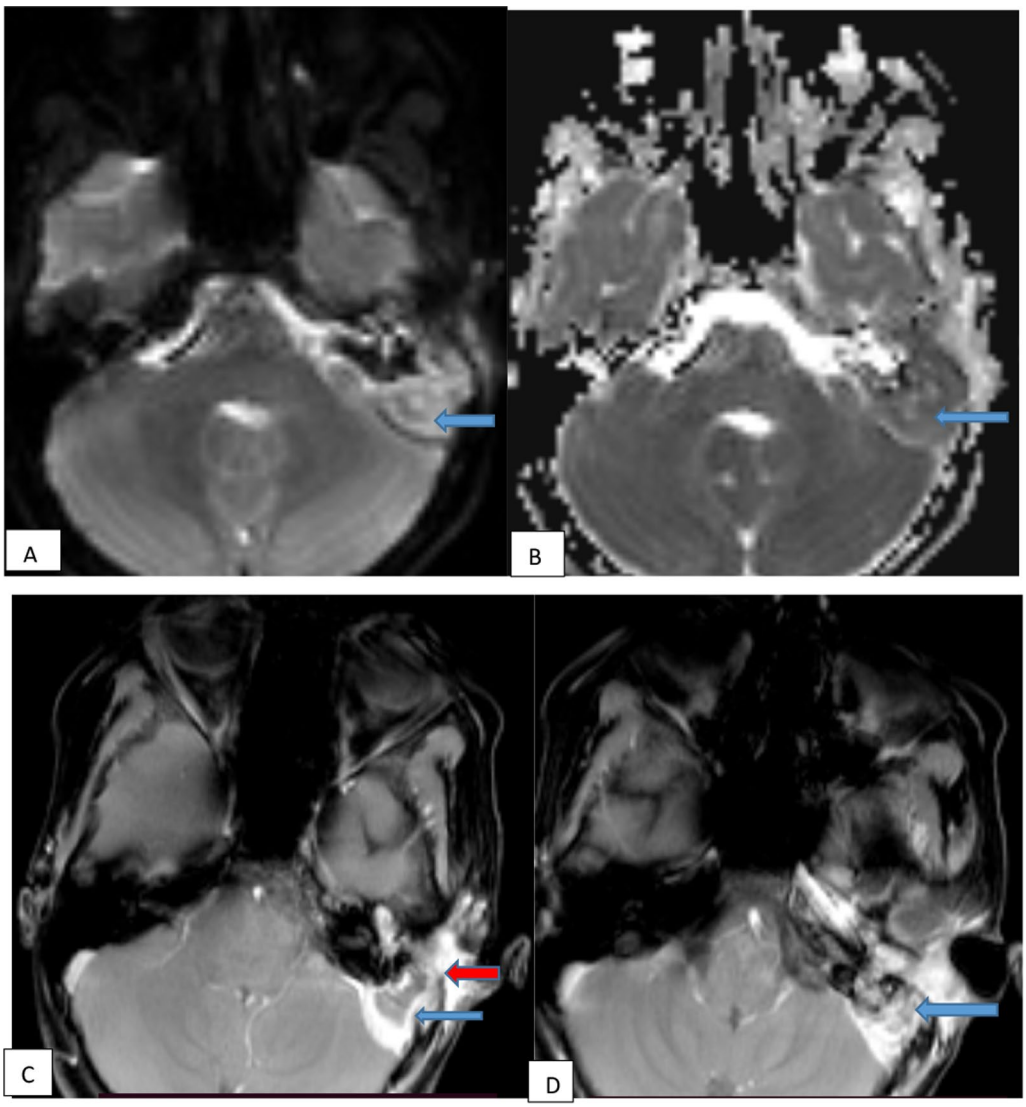

Fig. 3 Axial sections of MR image at base of skull shows: **A**) DWI image: restricted diffusion, predominantly in periphery, in the soft tissue along left cerebellopontine angle (blue arrow). **B**) ADC image: corresponding signal drop (blue arrow). **C**) Post-contrast T1 fat-suppressed MR image: Enhancing soft tissue in left external auditory canal, petromastoid air cells (red arrow) with attenuated calibre of left sigmoid sinus (blue arrow). **D**) Post-contrast T1 fat-suppressed MR image: Markedly attenuated calibre with filling defect within distal sigmoid sinus (blue arrow)

The various complications that can occur in tubercular infection are labyrinthine and post-aural fistula, subperiosteal abscess, meningitis, petrous pyramid osteomyelitis, cerebral and cerebellar abscess and facial nerve palsy. Early institution of antitubercular drugs which are the first line of treatment can prevent the development of these complications [2].

Middle ear rhabdomyosarcomas constitute about 7% of head and neck rhabdomyosarcomas and have symptoms similar to chronic otomastoiditis [11]. Imaging findings include enhancing soft tissue lesion with adjacent osseous erosion. There would be cranial nerve involvement and perineural spread of disease in cases of rhabdomyosarcoma of head and neck. Brain parenchymal and dural metastasis are seen with other distant sites of metastasis such as lymph nodes, lungs, liver, bones and extremities [12].

Langerhans cell histiocytosis is a reactive disease caused by abnormal immune regulation. Temporal bone involvement is seen in 15–61% of cases of LCH. Radiological imaging findings mimic otomastoiditis but with significant bone erosion and marked oedema which are disproportionate with respect to the clinical symptoms. MRI shows extensive bony destruction

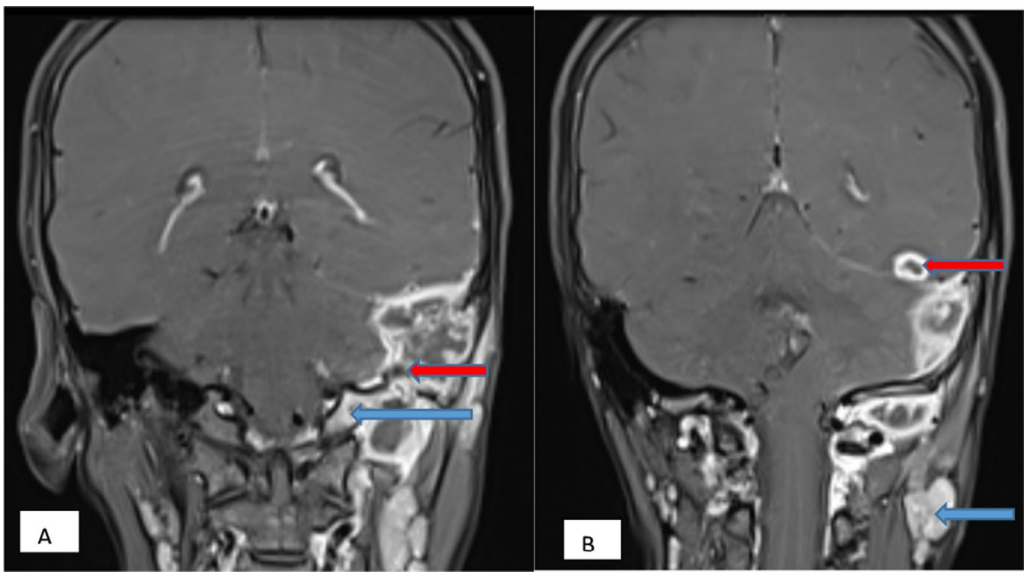

Fig. 4 Coronal post-contrast T1 fat-suppressed MR image shows: **A**) peripherally enhancing soft tissue intensity lesion in left petromastoid temporal bone (red arrow) with extension in left occipital condyle (blue arrow). **B**) Peripherally enhancing lesion in left temporal parenchyma abutting the tentorium cerebelli (red arrow) with homogenously enhancing enlarged cervical lymph nodes (blue arrow)

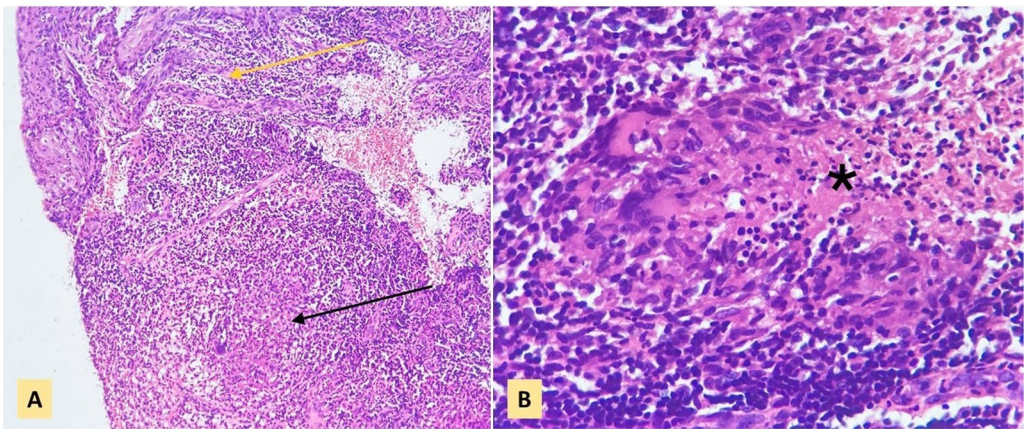

Fig. 5 Biopsy from external auditory canal shows **A**) ulceration of the covering stratified squamous epithelium showing marked pseudoepitheliomatous hyperplasia (yellow arrow). The ulcerated areas show several granulomas (black arrows) surrounded by dense acute on chronic inflammation and granulation tissue, 100 × **B**) The granulomas show central foci of necrosis and microabscess formation (star), 400x. Haematoxylin and eosin stain

with T1 hypo-intense, T2 hyper-intense soft tissue lesion with marked contrast enhancement and marked surrounding oedema [13]. The bony labyrinth and the cranial nerves are generally spared in LCH. CT scan demonstrates well-demarcated radiolucent areas with bony destruction of petrous apex. Involvement of various reticuloendothelial organs is commonly seen with bilateral temporal bone involvement in a few cases [14]. The role of MRI is mainly to assess the soft tissue and bone marrow [15].

The primary treatment of temporal bone tuberculosis is with antitubercular drugs for at least 6 months [6]. Here in this case, the intensive phase of ATT was given for 2 months followed by the maintenance phase which was initiated for another 10 months. The patient was evaluated with a CEMRI at 6 months of treatment which showed a significant interval reduction in the size of the lesion and the same treatment was advised for the next 6 months as per National Tuberculosis Elimination Programme [16]. The patient is now due for her follow-up

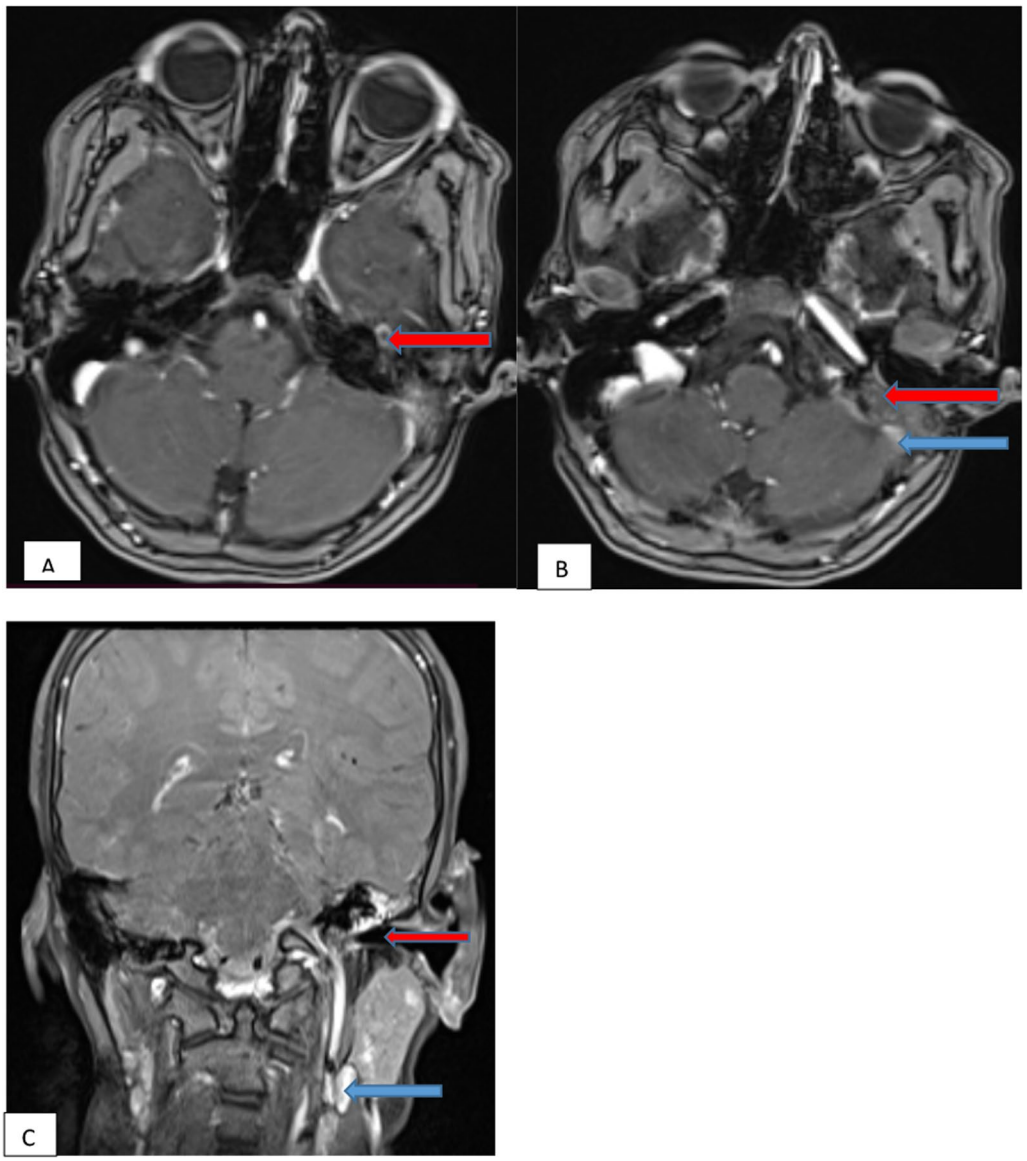

Fig. 6 Post-contrast T1 fat-suppressed MR image at the base of skull after 6 months of treatment shows: **A**) Axial section: minimal residual enhancing collection in the petromastoid temporal bone with a significant interval reduction in its extension (red arrow). **B**) Axial section: Reduction in extension into the left cerebellopontine angle (red arrow) with narrow calibre and thrombosis of adjacent sigmoid sinus (blue arrow). **C**) Coronal section: resolution of soft tissue infiltration in left external auditory canal (red arrow) and reduction in enlarged left cervical lymph nodes (blue arrow)

after 3 months. Surgery is reserved only for the purpose of obtaining tissue for HPE, draining a subperiosteal abscess, facial palsy or removing sequestered bone [17].

The greatest strength of this case report is the availability of histopathology which is the direct evidence of the exact aetiology of clinical symptoms and imaging features. Also, very good treatment response both clinically and radiologically after 6 months of ATT is another positive point in favour of this case report. One limitation in this case was that the sample was not sent for CBNAAT and Gene-Xpert for assessing drug resistance. The workup of cases of TB is incomplete without sending the sample for drug resistance testing in endemic countries like India where the incidence of multidrug-resistant tuberculosis (MDR-TB) is high.

Patient's perspective on treatment

The patient was very satisfied with the treatment because of the resolution of her symptoms and the complete resolution of the mass in the external ear within 2 months of initiation of treatment. The interval contrast-enhanced MRI findings were also direct evidence of a very good response to ATT. The patient is ready to continue treatment for the remaining 6 months. Under the Revised National Tuberculosis Control Programme (RNTCP) of the Government, direct monitoring of the treatment (DOTS—directly observed treatment, short course) is made and compliance of the patient with treatment is documented.

Conclusions

This case was reported because of the rarity of primary tuberculous otitis media in the paediatric population and more so the uncommon neuro-otogenic presentation. The importance of diagnosing and starting treatment early so as to prevent potential temporal bone and intracranial complications has been emphasized. Tuberculous otitis media can often masquerade as or mimic malignancy on clinical and imaging assessment. The need for close clinical and radiological follow-up in cases of tuberculous otitis media has also been highlighted in this case report. TB should always be considered a rare but possible cause for such neuro-otogenic lesions.

Abbreviations

HRCT: High-resolution computed tomography; CEMRI: Contrast-enhanced magnetic resonance imaging; COM: Chronic otitis media; TOM: Tuberculous otitis media; TB: Tuberculosis; ATT: Anti-tubercular therapy; LCH: Langerhans cell histiocytosis; HPE: Histopathological examination.

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Author contributions

The first author has contributed to the radiological findings and imaging assessment of the patient as well as writing the original draft. The second author has contributed to the description of clinical presentation and assessment of treatment response of the patient as well as writing the original draft. The third (corresponding author), fourth and fifth authors have contributed to review and editing of the original manuscript and supervision of the entire process till article submission. The sixth author has contributed to analysing and providing histopathological images as well as supervision of entire submission process. All authors read and approved the final manuscript.

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Availability of data and materials

All the data used in this report in the form of images were collected after obtaining proper permission from the authorized person. The entire images of the MRI and HRCT done are available with the first author, and treatment records are available with the second author and can be made available on reasonable request.

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from the next kin of patient. Approval from ethics committee is not required for publishing case report in our institution, Christian Medical College and Hospital, Ludhiana, because it is a case report and identity of patient is not revealed in any stage and kept anonymous. However, the head of institution (Principal) was intimated through proper channel prior to submitting the article for submission.

Consent for publication

Written informed consent was obtained from the next kin of patient for the information regarding clinical data of this case report and case images.

Competing interests

The authors have no conflicting interests to declare.

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