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Inflammatory Myofibroblastic Tumor of the Temporal Bone: A Rare Pathology at an Unusual Anatomical Site

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Abstract

Background: Inflammatory myofibroblastic tumor (IMT) is a rare spindle-cell neoplasm that predominantly affects the lungs. Temporal bone involvement is exceptionally uncommon and poses significant diagnostic and therapeutic challenges. Case Report: A 37-year-old female presented with progressive right-sided otalgia, hearing loss, tinnitus and aural fullness for 10 months. Imaging revealed a lesion involving the external auditory canal, middle ear, and mastoid cavity. Histopathology and immunohistochemistry confirmed ALK-positive IMT. The patient underwent sub-total petrosectomy with complete tumor excision. Histopathological examination of the surgical specimen confirmed the diagnosis. Recovery was uneventful, and a follow-up MRI at 3 months showed no residual disease. Conclusion: Temporal bone IMT is a rare entity with non-specific clinical and radiological features. Histopathology with immunohistochemistry is essential for diagnosis, while complete surgical excision remains the treatment of choice. Reporting such rare cases may help improve understanding and guide future management.

Keywords

Inflammatory Myofibroblastic Tumor, Temporal Bone, Surgical Resection

1. Introduction

Inflammatory Myofibroblastic tumor was first observed in the lung and was first described by Brunn in 1939 [1]. According to the World Health Organization 2002, IMT is defined as "a distinctive lesion composed of myofibroblastic spindle cells accompanied by an inflammatory infiltrate of plasma cells, lymphocytes, and

eosinophils" [2]. It has several different names, most common being inflammatory pseudotumor as it mimics malignant neoplasms clinically, radiologically and microscopically. This makes it difficult to accurately assess the incidence rate.

Usually younger age group is affected, but other age group is not immune to it. Patient presentation along with its management is variable depending on the primary origin of tumor. It is mostly seen in the lungs, whereas IMT of head and neck account for fewer than 5% of all extra-pulmonary cases. The most common in this region is the orbit, followed by meninges, paranasal sinuses, and infratemporal fossa. Temporal Bone, skull base and facial nerve are very unlikely involved [3].

IMT of the temporal bone appears to have a more aggressive and unpredictable course. Involvement of critical structures in the temporal bone makes the diagnosis and treatment more challenging than IMT of the lung. For the diagnosis, pathological assessment of tissue is a must.

This limited understanding of the disease in this location makes it crucial to document and analyze such cases to improve our knowledge in diagnosing and treating it. We present a case of an extensive inflammatory Myofibroblastic tumor of temporal bone in an adult female.

2. Case Report

A 37-year-old-female presented with complain of otalgia and decreased hearing in right ear for ten months. Pain was dull aching in nature, radiating to the right side of head with a feeling of aural fullness. She also complained of tinnitus on and off. The patient gave negative history of ear discharge, vertigo, facial weakness and involvement of opposite ear. Her complain of otalgia was gradually progressive in nature for which she took analgesics from multiple doctors but there was no relief. The otorhinolaryngologist did otoscopic examination revealing presence of mass in external auditory canal (EAC). She was then advised to get pure tone audiometry and magnetic resonance imaging (MRI) of temporal bone done. Audiometry was suggestive of severe mixed hearing loss in right ear and normal in other. The T2-weighted MRI revealed a hyper-intense lesion in right mastoid cavity, middle ear, and EAC (**Figure 1**). High-resolution computed tomography (HRCT) of the temporal bone was also performed, demonstrating a soft-tissue density mass causing extensive erosion of the posterior canal wall, middle ear ossicles, tegmen tympani, and extension toward the petrous apex. In accordance with the MRI reports, she was planned for the biopsy from the EAC mass for histopathological examination.

Microscopic section of biopsy sample revealed proliferation of spindle shaped cells having elongated wavy nuclei and ill-defined cytoplasm, arranged in vague storiform pattern with inflammatory cell infiltrate of lymphocytes and plasma cells. Separate evaluation of cellular features noted no significant nuclear atypia, very low mitotic activity (fewer than 2 mitoses per 10 high-power fields), and an absence of tumor necrosis, yielding an initial impression of a low-grade

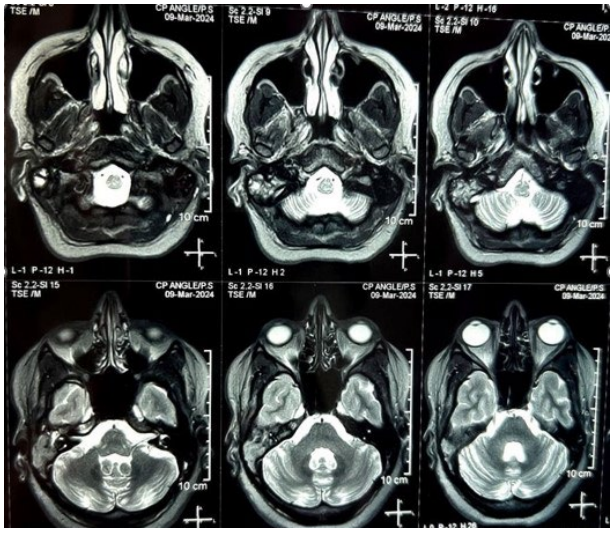

Figure 1. Axial section of T2 weighted MRI of temporal bone shows hyper intense lesion in the right temporal bone.

spindle-cell proliferation. To differentiate this from other temporal-bone spindle-cell lesions—such as schwannoma, leiomyosarcoma, nodular fasciitis, and fibromatosis—immunohistochemical (IHC) staining was performed. The lesional cells tested positive for smooth muscle actin (SMA), caldesmon, and anaplastic lymphoma kinase (ALK), while testing negative for desmin, muscle-specific actin (MSA), CD34, and S100. Diffuse ALK positivity combined with SMA and caldesmon expression conclusively distinguished IMT from smooth muscle sarcomas (which are typically desmin-positive), schwannomas (S100-positive), and fibrous proliferations (ALK-negative), confirming the diagnosis of ALK-positive IMT.

The patient underwent a mastoid exploration surgery under general anesthesia through a post auricular approach. The tumor was in the mastoid cavity eroding the tegmen tympani and was extending up to the petrous apex. A subtotal petrosectomy was executed, completely exenterating all mastoid air cells to achieve macroscopically complete gross tumor excision (in toto), Because complex skull-base structures (such as the carotid canal and dura) preclude wide three-dimensional clear soft-tissue margins, formal microscopic margin assessment of the deep skull base was not anatomically possible, though all gross disease was removed (**Figure 2**). It was then sent for histopathological examination. Following this the eustachian tube was obliterated with pieces of muscle and the mucosal lining of the middle ear was removed and a cul-de-sac closure was done. Histopathological study of mass and immunohistochemistry was consistent with finding of IMT. The post-operative course was uneventful. The patient was placed on a structured surveillance protocol consisting of clinical evaluations and serial contrast-enhanced MRIs every 3 months for the first year. A 3 and 6 month postoperative MRI confirmed the absence of residual disease, establishing disease control.

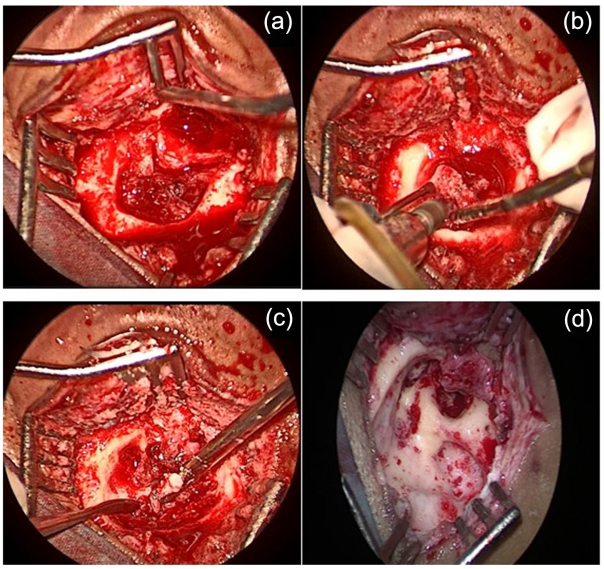

Figure 2. (a) Microscopic image showing tumour in mastoid antrum, (b) Drilling in mastoid cavity, (c) Removing tumour from mastoid cavity, (d) after subtotal petrosectomy.

3. Discussion

Inflammatory myofibroblastic tumors are rare and enigmatic group of lesions tend to be locally aggressive, but the malignant version (presence of distant metastatic disease) is very rare, making up less than 5%. There are no well-defined risk factors, and symptoms are often non-specific relating to the organ that the tumor is originating from [4].

The radiological examination shows soft tissue mass with no defined border. There is no specific radiological sign for diagnosing IMT. The histopathological and immunochemical reports serve as the primary sources in accordance with making an accurate diagnosis. Pertaining to the rarity of this entity, treatment modality of temporal bone IMT is still controversial due to the lack of available literature. However, according to H. S. Ong *et al.*, surgical resection with negative surgical margins is the gold standard treatment for head and neck IMT cases because of its better clinical outcome. The chances of recurrence are nearly 50% in cases with incompletely removed tumor. The other treatment modalities suggested are systemic cortisone or non-steroidal anti-inflammatory drugs as a medical treatment for the incompletely resected tumor. In aggressive cases, radiotherapy and chemotherapy have been tried [5]. In aggressive, recurrent, or metastatic settings, targeted biologic therapies—specifically small-molecule ALK inhibitors (e.g., crizotinib)—have emerged as highly effective therapeutic options for ALK-positive tumors, alongside conventional radiotherapy and chemotherapy.

4. Conclusion

Temporal bone is an extremely rare location of IMT. The non-specific symptoms and rarity of this disease make the pre-operative diagnosis difficult. The complete surgical resection offers good clinical outcomes. The increase in reporting of these

rare malignancies is the need of an hour to develop prompt treatment guidelines for the better long-term outcomes.

Ethical Clearance

It was taken from the institutional ethics committee.

Author Contributions

DT: Investigation, data curation, literature review, and writing—original draft. **PS:** Supervision, conceptualization, critical revision, and final approval. **AB:** Clinical evaluation, investigation, data curation, and manuscript preparation. **EB:** Clinical evaluation, literature review, interpretation, and writing—original draft. **AA:** Investigation, literature review, and manuscript revision. **AD:** Investigation, clinical documentation, and figure preparation. **BS:** Data collection, investigation and editing. **AK:** Literature review and critical review. **PG:** Clinical assistance, data verification, and manuscript revision. **YK:** Clinical assistance, data verification, and proofreading. **All authors:** Reviewed and approved the final manuscript and agreed to be accountable for the work.

Conflicts of Interest

There is no conflict of interest.

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