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Case Report

Plasma cell granuloma middle ear: A rare lesion at a rare site

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ABSTRACT

Plasma cell granuloma (PCG) is a rare non-neoplastic lesion, characterised by proliferation of immunoreactive polyclonal plasma cells present in a fibrous background. We present a case of 30 year male who presented with left sided otalgia, tinnitus and otorrhea along with mild conductive hearing loss. On CT scan a diagnosis of CSOM with granulation tissue was made however on histological examination, on microscopy with immunohistochemistry (IHC), it was reported as plasma cell granuloma of the middle ear. Middle ear plasma cell granuloma is a rare entity only few cases are reported in literature so far.
Here, we will discuss clinical presentation, histological diagnosis with IHC and management of this rare entity at an unusual location along with review of literature.

1. Introduction

Plasma cell granuloma, is a rare benign lesion of unknown aetiology, it is also called as inflammatory psuedotumor. Most common site for plasma cell granuloma is lung and major airways with rare involvement of some other uncommon sites.[1] Head and neck lesions make up just 5% of all extrapulmonary lesions, with the orbit being the most frequent location, followed by the meninges, paranasal sinus, infratemporal fossa, and soft tissue.

Other rare sites which could be involvement by PCG are temporal bone, skull base and facial nerve. The PCG is rarely found in the middle ear, with only sporadic cases reported in the literature. As a result, the incidence of this rare occurrence at this specific site has not been determined.[1] These lesions can manifest as a tumor, consisting of sheets of polygonal plasma cells, which may imitate a malignant condition. The final diagnosis of these lesion could only be made on the basis of collective gross, radiographic, microscopic and IHC findings. Exact etiology of the lesion is unknown, however, it is also considered to develop as a response to post inflammatory process.[2] The World Health Organization states that PCG is mainly made up of polyclonal plasma cells with a small number of innate inflammatory cells found in a myofibroblastic background.[3]

2. Case Report

A 30-year male presented in otorhinolaryngeology department of our teriary care multispeciality hospital with left sided otalgia, and otorrohea along with heaviness in ear for two and half months. He also complained of mild decrease in hearing for last 15 days. There was no history of vomiting, fever, nausea or dizziness. Otoscopic examination of the left ear showed central perforation of the tympanic membrane along with posterosuperior granulations with active discharge and myringitis. Facial nerve function was intact. Pure tone audiometry showed mild conductive hearing loss on the left side.

High resolution computed tomography (HRCT) temporal bone showed left tympano-mastoiditis consist of left middle ear cavity filled with soft tissue/fluid density along with

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loss of pneumatisation. However ossicular chain was intact, ossicles appeared normal in size, shape, outline and density. There was no significant sclerosis or cholesteatoma formation. Left Eustachian tube was blocked. Patient was admitted for above complains and necessary investigations were carried out. Intraoperatively, after making post auricular Wilde's incision left cortical mastoidectomy with tympanoplasty and ossiculoplasty was performed under general anaesthesia. Post-operative period was uneventful.

The tissue was sent to the department of histopathology in the form of multiple irregular tissue pieces, collectively measuring 1x0.8x0.4 cm. All the tissue bits were processed and sections were prepared for evaluation. Histopathological examination of the tissue on microscopy revealed middle ear mucosa lined by stratified squamous epithelium. Subepithelial stroma showed diffuse sheets of bland appearing plasma cells with surrounding fibro-collagenous stroma along with few lymphocytes, polymorphs, histiocytes and small capillaries. No mitosis and significant nuclear atypia were noted. Occasional scattered plasma cells showed Russel bodies in their cytoplasm.(Figure 1) All these findings suggested a diagnosis of plasma cell granuloma. On Immunohistochemical examination of plasma cells showed diffuse positivity for CD138.(Figure 2) IHC for kappa and lambda light chain expressed positive cytoplasmic staining in equal amount on these plasma cells thus indicating a polyclonal population of plasma cells.(Figure 3) These findings differentiate plasma cell granuloma from neoplastic plasma cell lesion which express monoclonality.

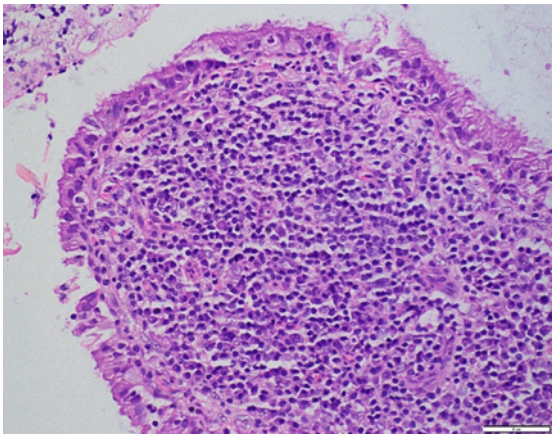

Figure 1: H&E (400x), showing sheets of plasma cells

3. Discussion

Plasma cell granuloma is a rare benign lesion, first described in 1973 by Bahadori and Liebow in 1973 to describe a psuedotumoral lesion of lung.[4] Plasma cell granuloma is also called as inflammatory psuedotumor and xanthomatous psuedotumor.[5,6] Although this lesion is infrequent, it can

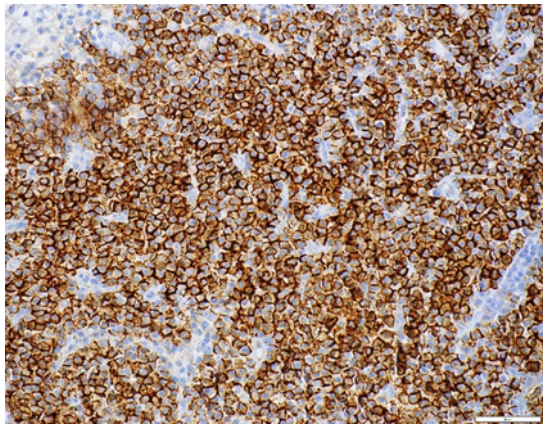

Figure 2: CD138 (400x), showing strong membranous positivity in plasma cells

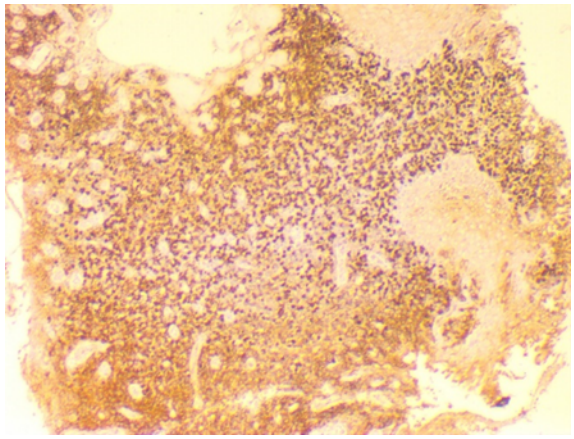

Figure 3: IHC for kappa light chain (200x), showing diffuse positive staining in plasma cells

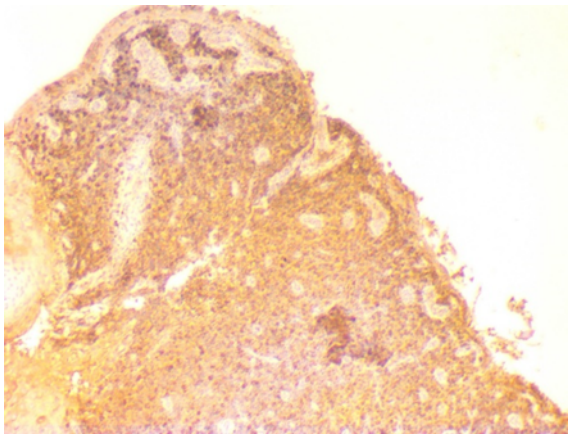

Figure 4: IHC for lambda light chain (200x), showing diffuse positive staining in plasma cells

manifest in any part of the body and possesses the capability to invade nearby tissue or erode bone. Therefore, it should be taken into account when considering the differential diagnosis of plasma cell neoplasm. While the cause of these lesions remains unknown, they are believed to develop as an atypical response to injury.[7] They can also develop in response to infection.[8] Recent literature indicates that PCG falls within the spectrum of IgG4-related diseases.[9] In the head and neck region, PCG typically presents as a locally destructive lesion, distinguishing it from other parts of the body where IgG4-related diseases are commonly identified incidentally during radiological assessments.[10]

Plasma cell lesions found in this area could potentially be plasma cell granuloma, inflammatory myofibroblastic tumor, IgG4 related disease, or Plasmacytoma. Nevertheless, distinct histological, clinical, and serological characteristics play a crucial role in differentiating between these diagnoses (Table 1).[11]

Table 1: Comparative features of the differentials of plasma cell granuloma

Inflammatory myofibroblastic tumor	**Plasma Cell Granuloma**	**Plasmacytoma**	**IgG4 related disease**
Myxoid (fibrous histiocytoma like)	heets of plasma cells	Sheets of plasma cells	Storiform fibrosis
Dense sclerosing (desmoid like), Spindle cell	Mature phenotype	May show nuclear atypia	Obliterative phlebitis
Variable number of plasma cells	Polyclonal	Monoclonal	Plasma cell infiltrate
ALK By IHC or FISH			IgG4:IgG ratio>40%

Our case was identified as Plasma cell granuloma, as there were no other features suggestive of alternative differentials. The polyclonality of plasma cells, confirmed through IHC, helped in excluding Plasmacytoma. Absence of sclerosis, spindle cell or fibroblastic proliferation, or myxoid background ruled out inflammatory myofibroblastic tumor. IgG4 disease was also ruled out due to the absence of obliterative thrombophlebitis, concentric fibrosis, and other systemic symptoms.

There is no definite age or sex predilection for this lesion. Due to few reported cases in the literature, there is no definite data regarding incidence or prognosis of this lesion is available, similarly there is very less information regarding pathogenesis, clinical behaviour as well as regarding treatment of the lesion. The preferred treatment to prevent the recurrence is surgical excision with a clear margin. In cases where the lesion is locally aggressive and cannot be surgically removed, or in patients with significant comorbidity but a resectable lesion, steroid or radiation therapy may be necessary.[12–14]

A recurrence rate of 22% has been reported during the 12-month follow-up after the surgery.[15] At the most recent follow-up, patient was disease-free and doing well. Otoscopic examination revealed intact tympanic membrane.

4. Conclusion

Plasma cell granuloma is a non-neoplastic proliferation of polygonal plasma cells, mainly present in lung, with rare occurrence at other sites. Middle ear is a rare site for plasma cell granuloma. Its clinical presentation may mimic many benign and malignant lesions, due to its ability to erode bone and local infiltration. Thus, appropriate diagnosis and management are critical. Surgical excision with clear margin is usual treatment for this lesion along with use of steroid and chemotherapy is few cases.

5. Sources of Funding

None.

6. Conflict of Interest

None.

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