Annals of R.S.C.B., ISSN:1583-6258, Vol. 25, Issue 1, 2021, Pages. 3815 - 3820

Received 15 December 2020; Accepted 05 January 2021.

Middle Ear Adenoma- A Rare Pathologic Encounter

Shweta Pandey[1], Arvind Bhake[2], Akriti Jain[3]

[1]Resident, Department of Pathology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Medical Sciences, Sawangi (M), Wardha;
[2]Director Professor, Department of Pathology, Jawaharlal Nehru Medical College, DattaMeghe Institute of Medical Sciences, Sawangi (M), Wardha;
[3]Resident, Department of Pathology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Medical Sciences, Sawangi (M), Wardha;

Email: [1]shweta.9152@gmail.com, [2]arvindbhake@gmail.com, [3]drakritijain@gmail.com

Address of corresponding author :-
Shweta Pandey
Department of Pathology, Datta Meghe Institute of Medical Sciences, Sawangi(M), Wardha
Email- Shweta.9152@gmail.com

Abstract:- Middle ear adenoma [MEA] is a rare pathologic condition and remains unsuspected clinically and radiologically. MEA has several clinical differential diagnoses that ranges from cholesteatoma to the one as rare as endolymphatic sac tumor.
A case has been reported of middle ear adenoma in a 62 years old male who was clinically unsuspected of it and had coexisting lesion of cholesteatoma and of Grade 3 facial nerve palsy which until has not been reported in literature to our knowledge.
Key words :- Middle ear adenoma, neuroendocrine differentiation, cholesteatoma, facial nerve palsy .

INTRODUCTION

Middle ear adenoma [MEA] is a rare pathologic entity and therefore reported infrequently in medical literature. It forms one of the differential diagnosis of middle ear masses even though its occurrence is unusual.

First known case series at reporting of middle ear adenoma was contributed by Hyams and Michaels(1).The entity of middle ear adenoma was previously described taxonomically as ceruminoma, ceruminous Adenoma, monomorphic adenoma and carcinoid tumor(2).
The Middle ear adenoma and carcinoid tumor of the middle ear possess less or more indistinguishable morphologies. But the studies published in the literature, on the parameter based on histopathology, Immunohistochemistry based on clinical behaviour and treatment outcome have revealed that middle ear adenoma is a distinct entity(3-5).

The histogenesis of this tumor was also debated in the literature for its origin from neuroendocrine cell and from middle ear mucosa. But by now there is a consensus amongst

Annals of R.S.C.B., ISSN:1583-6258, Vol. 25, Issue 1, 2021, Pages. 3815 - 3820

Received 15 December 2020; Accepted 05 January 2021.

the pathologists to unify adenomatous tumor of middle ear under the name of Neuroendocrine adenoma of the middle ear (NAME) [6-8].

Due to rarity of middle ear tumors its diagnostic encounters are infrequent to the pathologists[9-11]. So also the clinical and radiological features sufficiently varies to suspect middle ear adenoma[12-14].

This paper reports one such unusual case clinically and radiologically unsuspected of MEA in a 62 year male who concurrently suffered the lesion of cholesteatoma. The case is reviewed in the light of literature.

CASE REPORT

A 62 years man presented to Ear nose throat OPD of AVBRH with the symptoms of pain in left ear over a period of 7-8 months. He had discharge from the left ear for 5-6 months and difficulty in chewing food for past 3-4 months' time. The pain had started 8 months back in the left ear which was of gradual in onset. At his presentation the pain was throbbing type and was experienced intermittently. He took medicines for pain which did not subside on medication. The discharge from the ear was yellow, foul smelling and rarely stained of blood. It was of moderate quantity and had no relationship with chewing of the food. He also complained of progressive difficulty in chewing of food with a little drooping of saliva from the mouth.

The patient was on medication for diabetes mellitus and hypertension for two years. The examinations pertaining to ear revealed Normal preauricular, post auricular and auricular areas. The tympanic membrane of Right side was intact but left side tympanic membrane could not be visualized because of discharge. The abnormal Rinne's test for left ear could be elicited. The Air bone conduction was normal on right side and reduced on left side. The examination of the face revealed the signs of Left facial nerve palsy for closure of left angle of mouth, loss of nasolabial crease, reduced blowing of the cheek, watering from eyes and loss of frontal creases. The provisional clinical diagnosis was made as Left unsafe chronic serous otitis media with grade 3 facial nerve palsy. The patient underwent MRI and CT examination. The opinion offered was of left otomastoiditis with cholesteatoma with evolving small abscess with perilesional edema.

His base level hematological and biochemical investigations were Normal. He was advised surgery. Intraoperative findings were of presence of cholesteatoma in mastoid antrum, intact facial canal and ossicle along with floating bone present in external auditory canal and a mass deeply situated. Intraoperatively a diagnosis of suspicion of glomus tumor or else other neoplastic etiology associated of facial nerve palsy was made. The facial nerve palsy (Grade 3) was attributed to mastoid bone involvement of mastoiditis and Chronic serous otitis media. The tissue collected at surgery from a suspected area of growth on gross was multiple tiny tissue bits of approximately 1 x 1 cm together which were grey white in colour and little soft in consistency. The whole tissue was submitted for paraffin blocks. The histological section stained by hematoxylin and eosin showed the following morphological features of fibrocollagenous tissue lined at a single place by skin along with subsequent soft tissue with hypercollagenosis. A small area showed discrete lobulated tissue separated by thick

Annals of R.S.C.B., ISSN:1583-6258, Vol. 25, Issue 1, 2021, Pages. 3815 - 3820

Received 15 December 2020; Accepted 05 January 2021.

fibrocollagenous bands. Lobules were made of cells with eosinophilic cytoplasm and basally oriented nuclei in pseudoglandular pattern. Nuclei appeared benign and no evidence of malignancy was seen[Figure 1 & 2] The diagnosis of Middle ear adenoma was offered.

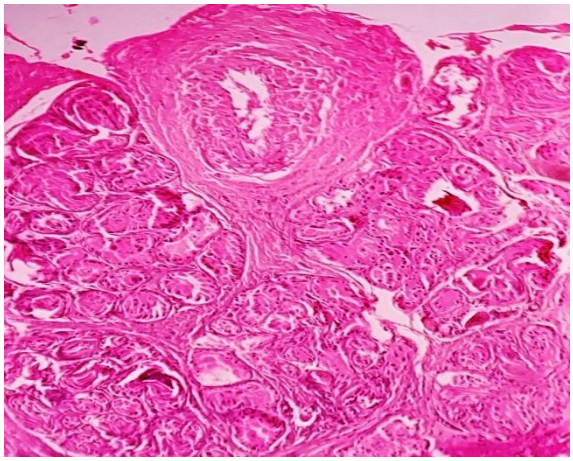

Figure 1:- Middle Ear Adenoma :Lobules and pseudoglandular structures of cells with eosinophilic cytoplasm and basal hyperchromatic nuclei [H & E, 10X]

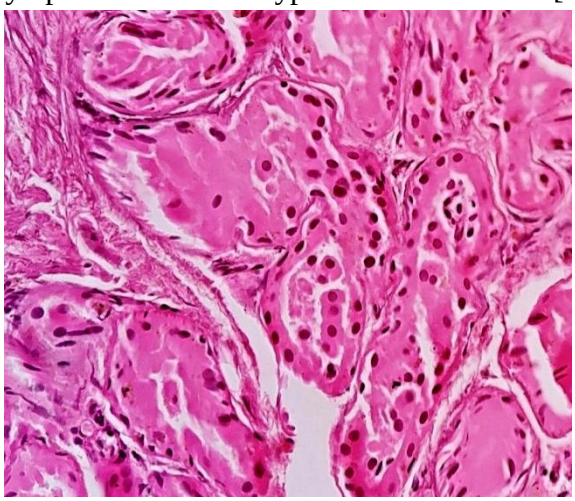

Figure2:- Middle Ear Adenoma :The cells with eosinophilic granular cytoplasm with a little of neuroendocrine differentiation [H & E, 40X]

The another separately sent container labelled as perilesional soft tissue revealed chronic inflammatory granulation tissue and cholesteatoma with spicules of calcification.

The paraffin sections on which the diagnosis of middle ear adenoma was offered underwent further investigation for CK7 immunostaining. The Immunohistochemistry for CK7 was Positive. The other immunohistochemical markers on the tissue of chromogranin and synaptophysin were weakly positive in rare focal cells.

DISCUSSION

The middle ear adenoma is a rare condition. Cardoso et al[(6)] published a systematic review on the adenomatous tumors of middle ear and has reported it to occur in 5th decade of life with common hearing symptom of conductive hearing loss.

Annals of R.S.C.B., ISSN:1583-6258, Vol. 25, Issue 1, 2021, Pages. 3815 - 3820

Received 15 December 2020; Accepted 05 January 2021.

Torske et al[5] reported adenoma versus carcinoid tumor of middle ear with a study of 48 cases. This study has observed that the mean age of patient with middle ear adenoma was 45 years with common symptom of hearing loss and pain with mean duration of 1.7 years. The presented reported case was a 62 year old male with multiple complaints of pain and discharge in left ear accompanied by hearing loss and facial nerve palsy.

The middle ear adenoma accompanied by facial nerve palsy has not been reported yet in the published literature. The present case intraoperatively did not show capsule around the tumor mass but showed a little bit of bony erosion. The similar findings of absence of capsule of middle ear adenoma has been reported in the reviews of Cardoso et al[6], Torske et al[5] and Bittencourt et al[8].

The middle ear adenoma clinically and radiologically simulating as cholesteatoma has been reported [5-6] as well as paraganglioma, schwannoma, retrotympanic vascular masses, ceruminal gland adenomas, endolymphatic sac tumors and others have been reported in the literature[3-6,9]. The present case was peculiar that middle ear adenoma pathology was present with a distinct another lesion of cholesteatoma. Such a coexistence of two different pathology has not been confronted in the reviewed reports for the present case.
The histomorphology of middle ear adenoma has been described by a few authors[3-14]. The similar histomorphology observed in the present case was similar to the one described by the aforesaid authors. The middle ear adenoma, with neuroendocrine differentiation has been reported by Kim et al[4], Torske et al[5], Cardoso et al[6], Tsiouvaka et al[7], Bittencourt et al[8], Saliba et al[12], Isenring et al[13] and Lella et al[14] alternatively been referred as Neuroendocrine adenoma of the middle ear (NAME)[4].

Torske et al[5] bought out a review that differentiated adenoma from carcinoid tumor of the middle ear. The study comprises of 48 cases. The comprehensive reporting for the features was on the points of clinical demographics, presentation, radiological findings, microscopic findings and immunohistochemical findings. The immunohistochemistry of middle ear adenoma revealed the high intensity staining for CK7 and absent to low staining for Chromogranin, Synaptophysin, Serotonin, Neuron-specific enolase and Human pancreatic polypeptide. The other immunohistochemical markers that enabled the differentiation were cytokeratin cocktails, CK20, S100 and vimentin. The study of Torske et al[5] concluded that MEA was most correctly described by their morphological features and clinical behaviour. It may instead be called as Neuroendocrine adenoma of middle ear by virtue of the immunoreactivity. The study further concluded that there is sharing of immunohistochemical markers between the lesion of middle ear adenoma and that of carcinoid.

The present case however was weakly positive in focal cells of chromogranin and synaptophysin but was positive for CK7, retaining its designation as middle ear adenoma without neuroendocrine differentiation.

Cholesteatoma upon the histochemical staining being redesignated as Neuroendocrine adenoma of middle ear has been reported by Kim et al[4]. However the presently reported case

Received 15 December 2020; Accepted 05 January 2021.

was distinct where the tumor mass histologically revealed morphologic evidence of middle ear adenoma and the tissue sent separately was showing a distinct cholesteatoma. The present case of middle ear adenoma holds clinicopathologic peculiarities that it was associated with facial nerve palsy. The tumor was appearing like a change of mastoiditis on CT and MRI, immunohistochemically lack of neuroendocrine differentiation and coexistence of cholesteatoma.

Some of rare cases and studies of adenoma have been reported[15-17]. Middle ear adenoma is uncommon, itshistomorphological mimics are many. It is known to have neuroendocrine differentiation and confuse with that of carcinoid and paraganglioma. The learning from the present case report was that middle ear adenoma immunohistochemically shows CK7 positivity. It may be associated with facial nerve palsy and it may be accompanied by a distinct lesion of cholesteatoma which is not yet been reported to the best of our knowledge.

REFERENCES

[1]. Hyams VJ, Michaels L. Benign adenomatous neoplasm (adenoma) of the middle ear. Clinical Otolaryngology & Allied Sciences. 1976 Jan;1(1):17-26.

[2]. Mills SE, Fechner RE. Middle ear adenoma. A cytologically uniform neoplasm displaying a variety of architectural patterns. The American journal of surgical pathology. 1984 Sep 1;8(9):677-85.

[3]. Berns S, Pearl G. Middle ear adenoma. Archives of pathology & laboratory medicine. 2006 Jul;130(7):1067-9.

[4]. Kim YH, Lee SH, Jung DJ. Neuroendocrine adenoma of the middle ear confused with congenital cholesteatoma. Brazilian journal of otorhinolaryngology. 2014 Apr;80(2):180-1.

[5]. Torske KR, Thompson LD. Adenoma versus carcinoid tumor of the middle ear: a study of 48 cases and review of the literature. Modern Pathology. 2002 May;15(5):543-55.

[6]. Cardoso FA, Monteiro EM, Lopes LB, Avila MN, Scarioli BO. Adenomatous tumors of the middle ear: a literature review. International Archives of Otorhinolaryngology. 2017 Sep;21(3):308-12.

[7]. Tsiouvaka S, Papadopoulou I, Lachanas V, Koltsidopoulos P, Skoulakis C. Neuroendocrine adenoma of the middle ear presented as paragaglioma: a case report. Hippokratia. 2017 Oct;21(4):201.

[8]. Bittencourt AG, Tsuji RK, Cabral Junior F, Pereira LV, Fonseca AC, Alves V, Bento RF. Middle ear adenoma with neuroendocrine differentiation: relate of two cases and literature review. International archives of otorhinolaryngology. 2013 Sep;17(3):340-3.

[9]. Zan E, Limb CJ, Koehler JF, Yousem DM. Middle ear adenoma: a challenging diagnosis. American journal of neuroradiology. 2009 Sep 1;30(8):1602-3.

[10]. Shim MJ, Song CI, Yoon TH. A case of middle ear adenoma. Korean journal of audiology. 2012 Apr;16(1):27.

[11]. Shim MJ, Song CI, Yoon TH. A case of middle ear adenoma. Korean journal of audiology. 2012 Apr;16(1):27.

[12]. Saliba I, Evrard AS. Middle ear glandular neoplasms: adenoma, carcinoma or adenoma with neuroendocrine differentiation: a case series. Cases Journal. 2009 Dec 1;2(1):6508.

[13]. Isenring D, Pezier TF, Vrugt B, Huber AM. Middle ear adenoma: case report and discussion. Case reports in otolaryngology. 2014 Jan 1;2014.

[14]. FA D. Middle Ear Adenoma: A Rare Location for a Benign Tumor. Journal of Otolaryngology-ENT Research. 2017;7(1).

[15]. Fidvi, M., A. Jadhav, A. Kasatwar, and R. Borle. "Pleomorphic Adenoma of the Cheek: Report of a Rarity." Journal of Datta Meghe Institute of Medical Sciences University 13, no. 1 (2018): 63–65. https://doi.org/10.4103/jdmimsu.jdmimsu_108_17.

Received 15 December 2020; Accepted 05 January 2021.

[16]. Gulve, S.S., and S.V. Phatak. "Parathyroid Adenoma: Ultrasonography, Doppler, and Elastography Imaging." Journal of Datta Meghe Institute of Medical Sciences University 14, no. 1 (2019): 47–49. https://doi.org/10.4103/jdmimsu.jdmimsu_91_18.

[17]. Lamture, Y., and A. Ghunage. "Ectopic Pleomorphic Adenoma in Intramuscular Plane of Sternocleidomastoid Muscle: A Rare Case Report." Journal of Datta Meghe Institute of Medical Sciences University 14, no. 4 (2019): 417–19. https://doi.org/10.4103/jdmimsu.jdmimsu_89_19.