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Letter to the editor

Recurrent neuroendocrine adenoma of the middle ear: A case report

Un cas d'adénome amphicrine récidivant de l'oreille moyenne

The authors report the case of a 27-year-old man with recurrent neuroendocrine adenoma of the left middle ear. The tympanum was explored via a posterior approach due to suspicion of cholesteatoma of the middle ear, allowing resection of tumour tissue during this surgical procedure. Histological examination was in favour of neuroendocrine adenoma.

Six years later, the patient reported onset of left-sided hearing loss. Audiogram demonstrated unilateral left-sided conductive hearing loss estimated at 40 db HL. Otoscopic examination revealed a whitish retrotympanic mass. Computed tomography of the petrous temporal bone demonstrated a round tumour occupying the epitympanic recess and mesotympanum with signs suggestive of ossicular lysis (Fig. 1).

Exploration of the tympanum via a posterior approach allowed resection of two recurrent tumours in the additus-ad-antrum and mesotympanum. Partial lysis of the incudostapedial joint was observed, but the ossicular chain nevertheless remained mobile and continuous. The bony implantation base of the lesion was drilled and the ossicular chain was preserved.

Histological examination demonstrated 2 sites of recurrence of neuroendocrine adenoma measuring 7 mm and 2 mm, with no signs of cytonuclear atypia. An immunohistochemical study demonstrated labelling of the cells with synaptophysin antibody and chromogranin (Fig. 2).

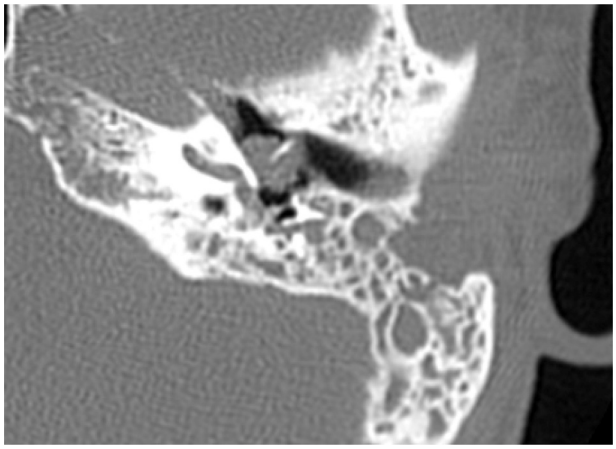

Fig. 1. Computed tomography visualizing a tumour around the long process of the incus with opacified mastoid air cells.

An audiometric gain of 10 db HL was observed one month after the surgical procedure. At 3-year follow-up, the patient did not present any recurrence and the audiometric gain was maintained.

Neuroendocrine adenoma of the middle ear (NAME) is a very rare benign tumour, first described in 1976 [1]. About fifty cases have subsequently been reported in the literature [2]. The mean age at diagnosis is 45 years [2]. The most frequent complaint is onset of unilateral conductive hearing loss [2–4]. Other possible symptoms are a feeling of ear fullness, earache, vertigo, tinnitus, superinfection, otorrhagia and facial paralysis [2–4]. Computed tomography

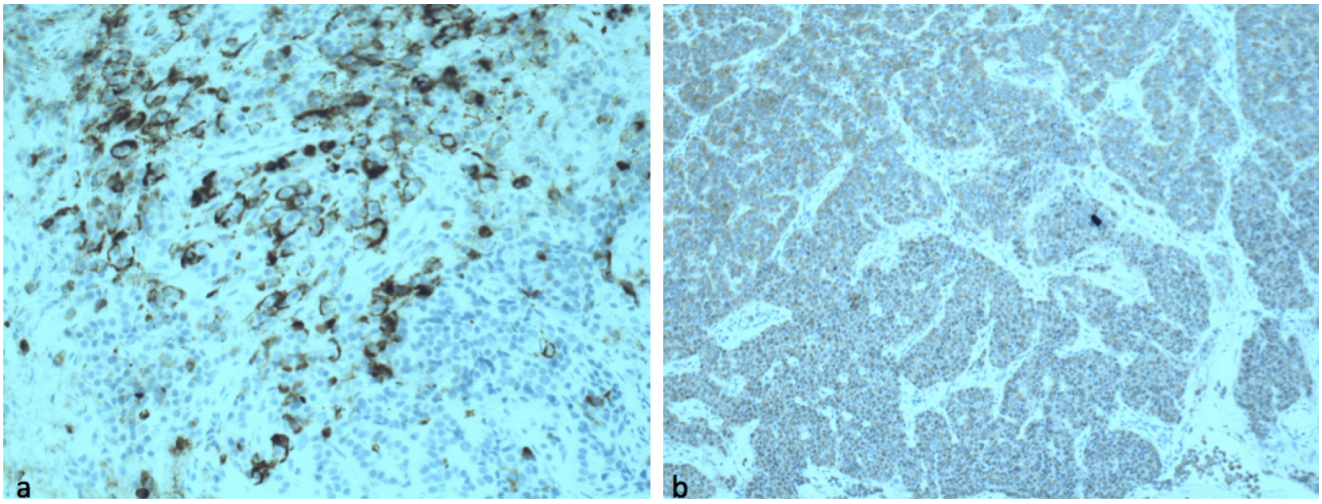

Fig. 2. Demonstration of chromogranin (a) and synaptophysin (b) immunohistochemical markers in favour of neuroendocrine differentiation.

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reveals a non-specific tumour without osteolysis, localized in the middle ear, intimately related to the ossicular chain and associated with opacified mastoid air cells. The lesion can spread to the mastoid, external auditory canal and Eustachian tube [2]. Treatment consists of surgical resection [2,3,5]. The recurrence is 18% to 22% [2,3]. Preservation of the ossicle chain, when involved, increases the risk of recurrence [2].

NAME is a benign tumour presenting double adenomatous and neuroendocrine on differentiation [4,5]. Neuroendocrine differentiation is demonstrated on immunohistochemistry by labelling of tumour cells by chromogranin A and synaptophysin [2,4]. Benign neuroendocrine tumours have been described by various terms: middle ear adenoma, amphicrine, carcinoid or adenocarcinoid tumour. These various terms actually correspond to a single histological entity, but presenting varying degrees of glandular and neuroendocrine differentiation. The term carcinoid tumour of the middle ear was defined by analogy with intestinal or bronchopulmonary carcinoid tumours. Carcinoid tumour is the most aggressive form of middle ear adenoma [5].

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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