CASE STUDY

Inverted Papilloma of Middle Ear and Temporal Bone

Papiloma invertido de oído medio y hueso temporal

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A fifty-year-old male presenting with right painless non-fetid otorrhea of several months onset and severe ipsilateral mixed hearing loss, with no other symptoms. His history included an operation for a right inverted nasosinusal papilloma 2 years earlier.

Otoscropy showed a granuloma occluding the external auditory canal. A CT scan was requested (Fig. 1A and B), that revealed occupation of the mastoid, with destruction of the cell walls, the tegmen tympani and the ossicular chain. MRI was recommended (Fig. 1C, T2 weighted axial image and D, coronal FLAIR image), and reported a mass with poorly defined margins with areas of necrosis on the posterior superior slope with intracranial expansion to the temporal fossa and integrity of the dura mater.

A subtotal petrosectomy was performed, obtaining various samples that were cystic, granulous and brown in appearance. The histological study (Fig. 2) showed a proliferation of epithelial squamous cells with no atypias, monomorphic, arranged in papillary formation with expansive growth towards the stroma, with a diagnosis of inverted papilloma. After 5 years of follow-up there are currently no signs of recurrence.

Discussion

Inverted papilloma is a benign tumour with locally invasive characteristics that most frequently present in the nasal cavity, and are very rarely located in the temporal bone. 1

Thirty-three cases of temporal bone inverted papilloma (TBIP) have been described between the years 1987, when it was first described, 2 and June 2016. Nineteen of the cases were male and 14 female, with a mean age of presentation of 53 years (19–81 years).

There are theories regarding the pathogenesis of TBIP ranging from direct extension from the sinonasal territory through the Eustachian tube to the middle ear, persisting ectopic embryological remnants of Schneiderian membrane to iatrogenic aetiology. An example of probable extension through the Eustachian tube is that of the studies as a whole, 20 cases (60.6%), associate a history of either synchronous or metachronous SNIP; the remaining 13 (39.4%) were primary tumours in the middle ear and/or temporal bone, with no history of sinonasal disease of any type.

If we divide TBIP into 2 groups, those that are primary and those that are secondary to a sinonasal disorder, we observe that the primary group tend to start at earlier ages and are more prevalent in females. In addition, their behaviour...
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Figure 1  CT and MR images in axial (A and C) and coronal slices (B and D).

Figure 2  (A) small magnification image showing a polypoid lesion with an endophytic growth pattern towards the stroma (H&E). (B) Epithelial proliferation comprising monomorphic squamous cells without atypias and without keratinisation (H&E, ×200).

very much resembles the sinonasal primary, with a low rate of intracranial extension, neuropathy and carcinogenesis. By contrast, those that are secondary to a sinonasal disorder seem to be more aggressive, and more frequently turn malignant and invade intracranially.

The distribution by sex of SNIP ranges from 2:6 and 6:1 in favour of the male gender. In TBIP, the majority of the first cases published were female and attempts were made to relate it with finding hormonal progesterone receptors, positive in 2 primary and 4 secondary TBIP patients. With the publication of new cases, this tendency for female predominance has adjusted until, with the present case, the ratio is currently a 1.4:1 male predominance.

The principal prognostic factor is the risk of malignant transformation to a squamous cell carcinoma, which the different reviews of SNIP set as varying between 1% and 13%. With TBIP, this percentage increases to 42.4% (14/33). Of the 14 cases, 8 were squamous cell carcinomas and 6 were carcinomas in situ. This might be explained by the late diagnosis of this type of disorder since the clinical signs and symptoms generally appear when after wide extension.

There are other factors and theories that might predispose to malignant transformation such as altered expression
of p53 and human papilloma virus (HPV) infection in the mucosa of the inverted papilloma. These factors have been listed and cited in different studies referring to the sinonasal location, without any evidence or demonstrated influence.

Another characteristic of inverted papillomas is recurrence; in the case of SNIP, recurrence rates of around 10% to 15% have been observed. The persistence and recurrence of TBIP is greater. Of the 14 cases of recurrence, 85% were treated by more limited mastoidectomy-type surgery. Only 2 cases (9.5%) recurred when the surgery was more extensive, with subtotal petrosectomy, infratemporal fossa approaches or retrosigmoid approaches.

Finally, given that TBIP has a higher recurrence rate and risk of carcinogenesis than SNIP, we should adapt the type of surgery to achieve wide and complete exeresis.

Conflict of Interests

The authors have no conflict of interests to declare.

References