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Clinical Record

Sinonasal inverted papilloma with malignant transformation in the middle ear: a multicentric origin?

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Abstract

Inverted papilloma is a rare, benign tumour representing only 0.5 to 4 per cent of all sinonasal neoplasms; its involvement of the middle ear is extremely rare. We present a case of multicentric inverted papilloma in the sinonasal region and middle ear in a 54-year-old man. The patient later developed neck metastasis secondary to malignant transformation of the inverted papilloma in the middle ear.

Key words: Papilloma, Inverted; Paranasal Sinuses; Ear, Middle; Epithelial Neoplasms, Malignant

Introduction

Inverted papilloma is a rare, locally aggressive, benign nasal neoplasm representing only 0.5 to 4 per cent of all sinonasal neoplasms;[1] it is remarkable for its high recurrence rate, multicentricity and association with carcinoma in 9 per cent of cases.[2] The lesion usually originates from the mucosa of the lateral nasal wall and can involve the paranasal sinuses, orbits, anterior skull base, pharynx and nasopharynx. Inverted papilloma involving the temporal bone or middle ear is an extremely rare occurrence; to our knowledge, there have been only 15 reported cases in the literature.[3,4]

We present the case of a middle-aged man diagnosed with sinonasal inverted papilloma who underwent tumour excision via lateral rhinotomy and medial maxillectomy, and who was found later to have malignant transformation of the inverted papilloma in the middle ear, with neck metastasis.

Case report

A 54-year-old Malay man experienced frequent left rhinorrhoea and persistent nasal blockage of one year's duration. These symptoms were associated with hyposmia, occasional epistaxis and dull headache. However, there were no ear symptoms.

On examination, the left nasal cavity was filled with a fleshy mass arising from the lateral wall and extending to the nasopharynx. The left tympanic membrane appeared retracted but not perforated. A 512 Hz tuning fork test revealed left conductive hearing loss.

Biopsy of the nasal mass was reported as inverted papilloma. A computed tomography (CT) scan showed a mass in the left nasal cavity extending to the maxillary and ethmoid sinus, with expansion and remodelling of the maxillary sinus.

The patient underwent a left partial maxillectomy via a lateral rhinotomy approach for tumour excision and histopathological confirmation of an inverted papilloma.

The patient did well post-operatively, until five months later, when he complained of epistaxis and left ear discharge, with reduced hearing.

Nasoendoscopy at this time showed a mass arising from the lateral wall of the maxillary sinus and extending to the nasopharynx. Otoscopic examination revealed a polypoidal lesion arising from the left middle ear. There was also a palpable lymph node at level II, measuring about 2×2 cm. Biopsies of the nasal lesions showed inverted papilloma with moderate dysplasia, whereas the ear polyps were shown to be composed of inverted papilloma with an area of malignant transformation in which the tumour had infiltrated into the underlying stroma (Figure 1). Fine needle aspiration of the lymph node showed poorly differentiated carcinoma. A CT scan of the paranasal sinus and the neck supported the above findings. However, there was no thickening or irregularity of the eustachian tube to suggest continuity of the primary tumour in the nose or nasopharynx with the middle ear. A high resolution CT scan of the temporal bone showed a soft tissue mass occupying the left external ear canal, mastoid air cells and middle ear (Figure 2). No erosion of the ossicles or inner-ear structures were seen.

The patient was advised to undergo endoscopic removal of the nasal tumour recurrence, a lateral temporal bone resection for the middle-ear cleft disease and a left radical neck dissection followed by radiotherapy. However, the patient refused surgery and opted for radiotherapy instead.

Discussion

Inverted papilloma is a rare, benign, epithelial neoplasm characterized by its tendency to recur following excision, its association with malignancy and its multicentricity. Sinonasal papilloma represents about 0.5 to 4 per cent of all nasal tumours,[1] but inverted papilloma involving the

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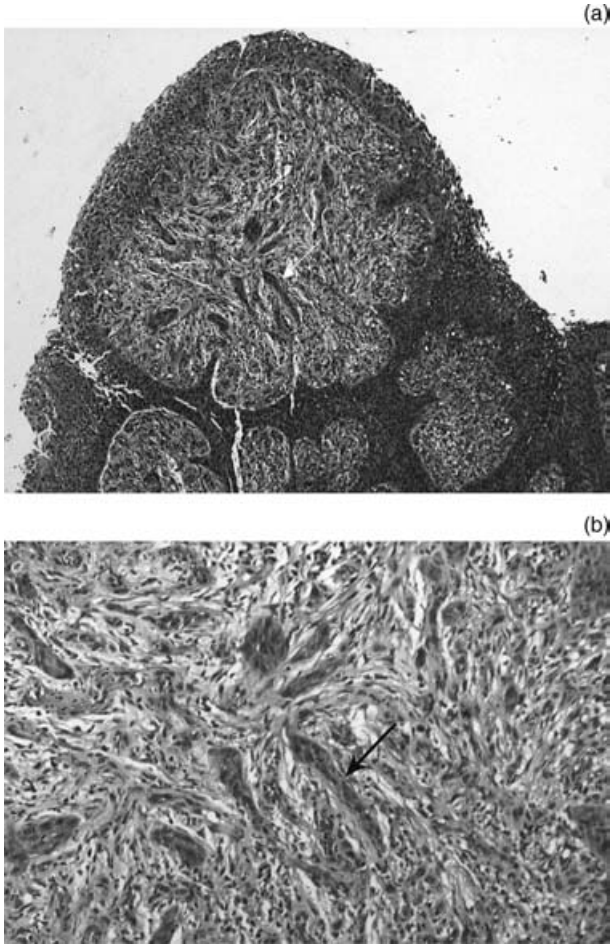

FIG. 1

(a) Polypoidal tissue predominantly showing inverted papilloma with an area of malignant infiltration into the stroma (arrow) (H&E; ×40), (b) High power view showing strands of malignant squamous clumps within the stroma (arrow) (H&E; ×200).

temporal bone or middle ear is an extremely rare occurence. To our knowledge, only 15 cases have been reported.[3–5] Sinonasal inverted papilloma occurs predominantly in men (4:1 male-to-female ratio)[6] but involvement of the middle ear shows a 2:1 female-to-male ratio.[5] The hypothesis that the influence of sex steroid hormone receptors differs in sinonasal and temporal bone or middle-ear inverted papilloma should be considered.[5]

The cause of this middle-ear or temporal bone lesion is poorly understood. Direct extension from the sinonasal cavity via the eustachian tube, multicentric primary origin and ectopic migration of the ectodermally derived Schneiderian membrane that lines the sinonasal cavities have been hypothesized.[3,7] In our patient, we assume this tumour was of multicentric primary origin as the CT scan was unable to detect any continuity of lesions or any irregularities of the eustachian tube

Over the years, several aetiologies have been proposed for inverted papilloma, including allergy, chronic inflammation, and carcinogen and occupational exposure. There has been growing evidence to support the association of human papilloma virus types 6B, 11, 16 and 18 in the pathogenesis of inverted papilloma. Human papilloma virus has been isolated from the epithelium in as many as 76 per cent of patients with sinonasal inverted papilloma[8] and in 12 per cent of patients with inverted papilloma of the middle ear.[9]

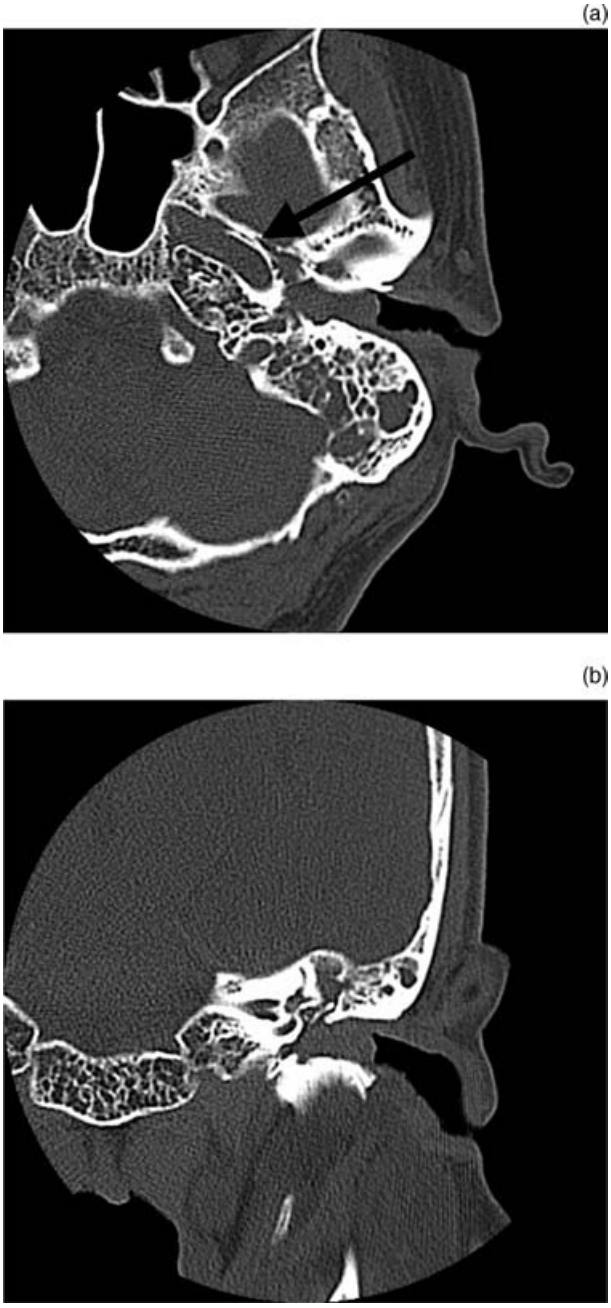

FIG. 2

High resolution computed tomography scans in the (a) axial and (b) coronal planes showing the presence of soft tissue mass in the external ear canal, mastoid air cells and middle ear. Note that the eustachian tube (arrow) is not enlarged.

A higher recurrence rate has been observed in inverted papilloma of the middle ear (82 per cent; six patients underwent a mastoidectomy or tympanomastoidectomy and three patients underwent a simple excision after myringotomy)[9] as compared with the sinonasal type of inverted papilloma (67 per cent with limited surgery and 10–13 per cent with an aggressive surgical approach).[10] Recurrence after mastoidectomy or tympanomastoidectomy was noted to occur six months to two years after surgery.[9]

The association of inverted papilloma and squamous cell carcinoma is well recognized and varies from 5 to 53 per cent in sinonasal papilloma[6] and 36 per cent in inverted papilloma of the middle ear.[9] There are a few ways in

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which malignancy can be associated with inverted papilloma: (1) synchronous carcinoma in an inverted papilloma; (2) focal areas of carcinoma-in-situ in an inverted papilloma; (3) areas of inverted papilloma inside a squamous cell carcinoma; (4) metachronous carcinoma not associated with the original site of the inverted papilloma; or (5) metachronous carcinoma at the original site of the inverted papilloma (otherwise known as malignant transformation), as we assume occurred in this patient. However, the proven malignant transformation rate is less than 2 per cent.[11] Therefore, it is important to undertake a vigorous histologic examination of inverted papilloma, with complete serial sectioning of the specimen.

Adequate surgical excision of the tumour, irrespective of its origin either in the sinonasal region or in the middle ear, remains the principal treatment choice. Radiotherapy is given only in inoperable cases and in those cases of inverted papilloma associated with malignancy following surgery.

Conclusion

We report a rare presentation of an inverted papilloma, which showed its unique characteristics: local aggressiveness, tendency to recur, association with malignancy and multicentricity. Despite the rarity and scarcity of cases, several papers have reported that inverted papillomas of the middle ear and temporal bone differ pathogenically and epidemiologically from those of the sinonasal region. Recurrence rates and association with malignancy are higher in papilloma of the middle ear than in the sinonasal type. Radical resection is advocated in managing Schneiderian-type papillomas of the middle ear or temporal bone.

- **This report describes a case of multicentric inverted papilloma in the sinonasal region and middle ear of a 54-year-old man. The patient later developed neck metastasis secondary to malignant transformation of the inverted papilloma in the middle ear**
- **Recurrence rates and association with malignancy are higher in papilloma of the middle ear than in the sinonasal type. Radical resection is advocated in managing Schneiderian-type papillomas of the middle ear or temporal bone**

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