

# Sinonasal inverted papilloma involving the temporal bone via the eustachian tube: case report

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## Abstract

**Objective:** This study reports a case of a sinonasal inverted papilloma with spread to the temporal bone via the eustachian tube and subsequent transformation to squamous cell carcinoma.

**Method:** An 81-year-old woman presented with sinonasal inverted papilloma which subsequently spread to the ear. A literature review of inverted papilloma was carried out based on a Pubmed search of studies published between 1987 and 2011, using the key words ‘sinonasal inverted papilloma’, ‘temporal bone inverted papilloma’ and ‘squamous cell carcinoma’.

**Results and conclusion:** Sinonasal and temporal bone inverted papillomas may sometimes be linked through direct spread via the eustachian tube. Inverted papillomas have the potential for malignant transformation; careful monitoring of both the nose and ear is therefore required for inverted papillomas found in the nasopharynx.

**Key words:** Papilloma, Inverted; Temporal Bone; Nasal Cavity; Eustachian Tube; Squamous Cell Carcinoma

## Introduction

Inverted papilloma is a rare, benign epithelial tumour. It has a tendency to recur after resection and there is the potential for malignant transformation. Sinonasal inverted papillomas usually arise from the middle meatus and account for 2–3 per cent of all nasal polyps.<sup>1</sup> Treatment for sinonasal inverted papillomas has shifted from open procedures such as rhinotomy and Caldwell–Luc approaches to endoscopic resection. Isolated temporal bone inverted papillomas are a much rarer clinical entity. A few patients with sinonasal inverted papillomas also develop disease in the middle-ear cavity, and intracranial extension has even been reported.<sup>2–4</sup> In most reported cases there is no direct connection between the different sites of disease. Therefore, it remains controversial whether this reflects the multifocal nature of the tumour or the contiguous spread of disease from the nasal cavity to the ear.<sup>3</sup>

## Case report

An 81-year-old woman presented to our out-patient department with a 12-month history of nasal blockage and blood-stained, purulent discharge from the right nostril. In addition, she felt her hearing was reduced on both sides. She was a non-smoker.

Examination revealed a large polypoidal mass occupying the right nostril, extending to the post-nasal space. There was also possible middle-ear effusion on the right side. Both external auditory canals were normal at this time. A nasal biopsy and examination under anaesthesia showed inverted papilloma without overt evidence of dysplasia or invasion. Computed tomography (CT) of the sinuses (Figures 1 and 2) revealed a massive right-sided polypoidal nasal

mass with no evidence of bony destruction, but extensive opacification of the ipsilateral sinuses was reported. The patient was claustrophobic, which prevented further evaluation using magnetic resonance imaging (MRI) at this point.

On the basis of these findings, the patient underwent endoscopic resection in an attempt to completely excise the inverted papilloma. The patient made a good post-operative recovery. However, when she was reviewed in clinic two months later, she reported increased hearing loss. A polypoidal mass was observed in the right external auditory canal. Excision of the aural polyp was carried out under anaesthesia. A further lesion was found intra-operatively on the superior surface of the soft palate, adjacent to the right eustachian tube opening. Analysis of biopsied tissue from both lesions revealed sinonasal papilloma, without cellular atypia or suggestion of malignancy.

The patient was reviewed in clinic two months later. Recurrence of the right external auditory canal lesion was observed and the palatal lesion had significantly increased in size. She now complained of pulsatile tinnitus and right-sided otorrhoea with occasional blood staining. An urgent CT of the temporal bone (Figure 3) showed soft tissue material in the nasopharynx adjacent to the eustachian tube, which had eroded the bony walls of the right external auditory canal and extended into the right middle-ear cavity and the mastoid air cells.

Due to concerns that the tumour may be malignant and had eroded through the tegmen, the patient was referred to Addenbrooke’s Hospital which offered skull base and neurosurgery facilities. A repeat biopsy of the post-nasal space, external auditory canal and eustachian tube was undertaken and a histological diagnosis of invasive non-keratinising

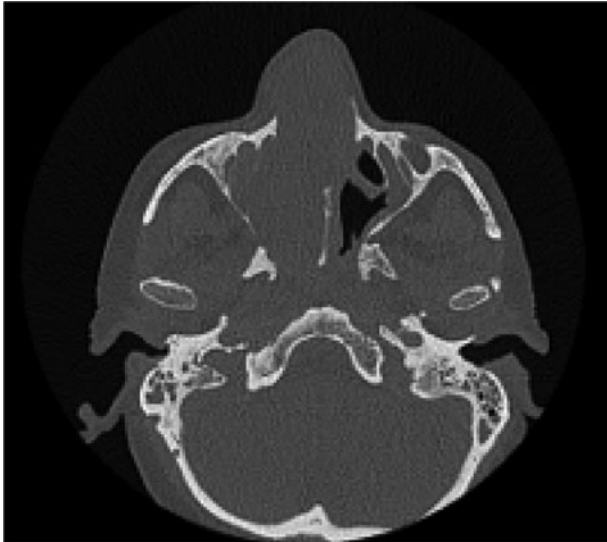


FIG. 1

Pre-operative axial computed tomography image of the sinuses demonstrating right-sided sinonasal papilloma extending to the post-nasal space.

papillary squamous cell carcinoma was made. Immunohistochemistry showed that the lesional cells were positive for cytokeratins 5 and 6, p63 and p16, suggesting human papilloma virus infection as a possible aetiology. There was no residual inverted papilloma. Samples from all three sites had similar histology. Computed tomography and MRI scans (Figure 4) showed no brain invasion or distal metastasis. The sinonasal papilloma may have spread from the post-nasal space to the external auditory canal via the eustachian tube, prior to malignant transformation. Extensive tumour involvement of the temporal bone and the post-nasal space rendered the patient unfit for surgical intervention, and she was commenced on palliative radiotherapy.

### Discussion

Inverted papilloma is a rare, benign epithelial tumour, which has the potential to induce local bony remodelling and to recur after resection. Sinonasal inverted papillomas account for 2–3 per cent of all nasal polyps.<sup>1</sup> Three variants of



FIG. 2

Pre-operative coronal computed tomography image of the sinuses showing right intranasal soft tissue mass.

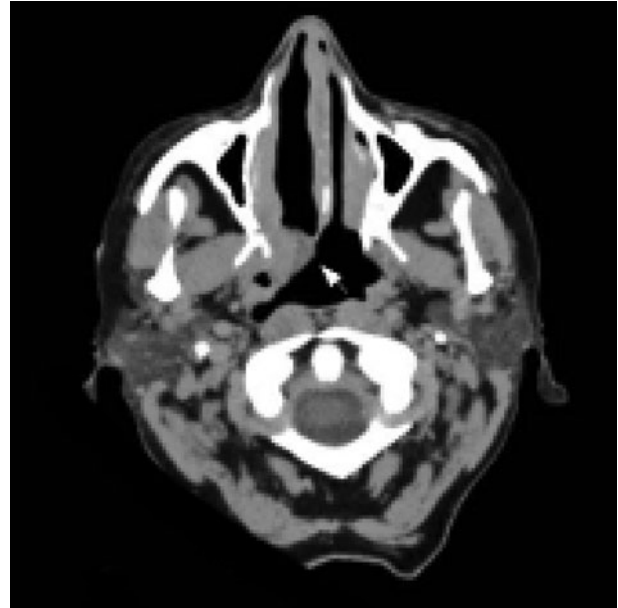


FIG. 3

Axial computed tomography image of the petrous temporal bones (post functional endoscopic sinus surgery) showing recurrence of sinonasal papilloma near the eustachian tube orifice (arrow) and filling the external auditory meatus.

inverted papilloma have been reported: sinonasal inverted papillomas, primary temporal bone inverted papillomas and a combination of both. There are fewer than 20 reports of temporal bone inverted papillomas in the literature; of these, 10 describe cases with concurrent sinonasal and temporal bone involvement.<sup>2–4</sup>

The most common symptoms associated with temporal bone inverted papillomas are hearing loss, otalgia and otorrhoea. Sinonasal inverted papillomas are now usually treated via endoscopic approaches, with reported recurrence

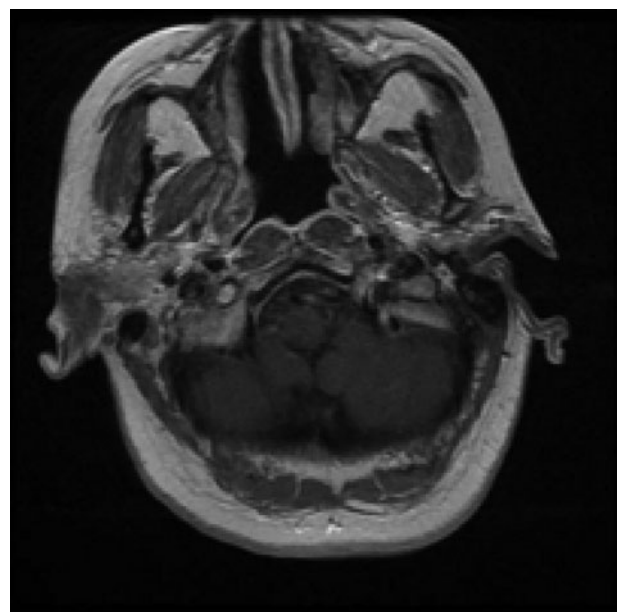


FIG. 4

Post-operative axial magnetic resonance image of the head showing soft tissue filling the external auditory meatus.

rates of 8–16 per cent. The much rarer temporal bone inverted papilloma is treated by resection with adjuvant radiotherapy. Malignant transformation of sinonasal inverted papilloma has been reported in 3.6 per cent to 11.8 per cent of cases,<sup>5,6</sup> with the median time from presentation to transformation being 52 months, ranging from 6 to 180 months.<sup>5</sup> Our case transformed within nine months, although histology at presentation did not demonstrate any high-risk features such as cellular atypia, hyperkeratosis or dysplasia.

It has been speculated that sinonasal and primary temporal bone inverted papillomas may be distinct clinical entities. The limited number of cases indicate that sinonasal inverted papillomas are more common in men (with a male to female ratio of 4:1) whereas primary temporal bone inverted papillomas are more common in women (a female to male ratio of 3:1).<sup>3</sup> At least one primary temporal bone inverted papilloma was shown to be progesterone receptor positive.<sup>7</sup> The rate of human papilloma virus positivity in temporal bone inverted papilloma is also much lower (12 per cent) compared with sinonasal inverted papilloma (30 per cent). However, this applies only to isolated primary temporal bone inverted papillomas. In patients with both sinonasal and temporal bone inverted papillomas, the sinonasal variant preceded temporal bone involvement in 9 out of 10 cases reported.<sup>4</sup>

- **Sinonasal inverted papilloma is a benign tumour that can invade the middle-ear cavity via the eustachian tube**
- **It is important to consider ear examination on both initial assessment and later follow up of sinonasal inverted papillomas**
- **Malignant transformation of inverted papilloma is a rare but clinically important occurrence**

Sinonasal inverted papilloma may involve the temporal bone due to multicentric growth of the tumour, as suggested by Kaddour and Woodhead.<sup>8</sup> These authors reported a patient who had sinonasal and temporal bone tumours nine years apart, without malignant transformation. Alternatively, it has been speculated, but not directly observed, that sinonasal inverted papillomas may spread to the temporal bone via the eustachian tube. However, only one other case report has described involvement of the eustachian tube, and that patient already had inverted papillomas in the nasal cavity, middle-ear cavity and the eustachian tube on presentation; therefore, it could not be ruled out that multiple tumours had arisen independently in the two sites.<sup>9</sup> In our patient, there was only a short period of time between the original diagnosis of sinonasal inverted papilloma and inverted papilloma in the middle ear with early involvement of the eustachian tube, which would suggest that direct spread of the

tumour was most likely. Our case demonstrates that a proportion of sinonasal inverted papillomas are capable of spreading to the temporal bone via the eustachian tube at a time when they are histologically benign, although in our case later malignant transformation did occur.

## Conclusion

We report a rare case of sinonasal inverted papilloma which spread to the middle-ear cavity prior to malignant transformation. The report demonstrates that this benign but locally aggressive tumour can invade the middle-ear cavity, most likely through the eustachian tube orifice. It is therefore important to ensure careful clinical assessment of the middle-ear cavity when managing patients with sinonasal inverted papillomas.

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