



Inverted Papilloma of the Bulla Ethmoidalis: An Uncommon Epicenter for a Benign Yet Aggressive Tumor

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Authors' contributions

This work was carried out in collaboration between both authors. Author AZBKZ clerked and examined this patient and wrote this manuscript. Author KABMR proof read this manuscript and perform surgery for this patient. Both authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Inverted papillomas are benign tumours known for their locally aggressive behaviour, characterised by local tissue destruction, a high tendency to recur if resection is incomplete, and a risk of carcinomatous evolution into squamous cell carcinoma. Although most cases arise from the nasal wall and paranasal sinuses, inverted papillomas originating from the bulla ethmoidalis are exceedingly rare. We report the case of a 67-year-old man who presented with progressive right-sided nasal obstruction and rhinorrhoea. Imaging and histopathological findings confirmed the diagnosis of inverted papilloma arising from the right bulla ethmoidalis. Surgical excision was performed, and complete resection was achieved. The patient had an uneventful postoperative

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recovery, and follow-up will be crucial for monitoring potential recurrence. This case highlights the importance of early recognition, appropriate imaging, and thorough surgical planning in managing inverted papillomas, particularly in rare locations such as the bulla ethmoidalis, to ensure optimal outcomes and reduce the risk of recurrence and malignancy.

Keywords: *Inverted papilloma; bulla ethmoidalis; mulberry-like mass; focal hyperostosis.*

ABBREVIATIONS

CT : Computer Tomography

MRI : Magnetic Resonance Imaging

1. INTRODUCTION

Inverted papillomas, also known as Schneiderian papillomas, are benign sinonasal tumours that commonly arise from the lateral nasal wall and middle meatus, but they may develop from any part of the Schneiderian membrane in the nose and paranasal sinuses (Murphy et al., 2010; Thompson, 2015). Although inverted papillomas are typically benign, they are characterised by locally aggressive behaviour, a high potential for recurrence, and an association with malignant transformation into squamous cell carcinoma (Lisan et al., 2016).

2. CASE PRESENTATION

A 67-year-old non-smoker with underlying hypertension and hyperlipidaemia presented with progressive right nasal blockage and rhinorrhoea

for the past 2 months. He had a history of nasal surgery at a private centre 12 years ago.

Anterior rhinoscopy and endoscopy revealed a grade IV right nasal cavity mass arising from the osteomeatal complex. A biopsy was taken from the right nasal cavity mass, and histopathological study showed invagination of superficial epithelium into underlying stroma, which suggests inverted sinonasal papilloma. Computed tomography (CT) showed a soft tissue density mass arising from the right middle turbinate, extending to the posterior choana, with minimal calcification seen at the superior margin of the mass at the anterior ethmoidal air cells' origin. The right osteomeatal complex was patent, and there was no mass effect on surrounding structures.

The patient underwent endoscopic right medial maxillectomy and right ethmoidectomy, which revealed a pale, glistening polypoidal, mulberry-like mass arising from the right bulla ethmoidalis (Figs. 1, 2). Pathological examination of the excised mass confirmed the diagnosis of inverted sinonasal papilloma.



Fig. 1. Intraoperative nasal endoscopy shows mulberry liked mass arising from right bulla ethmoidalis



Fig. 2. Excised mass from right bulla ethmoidalis

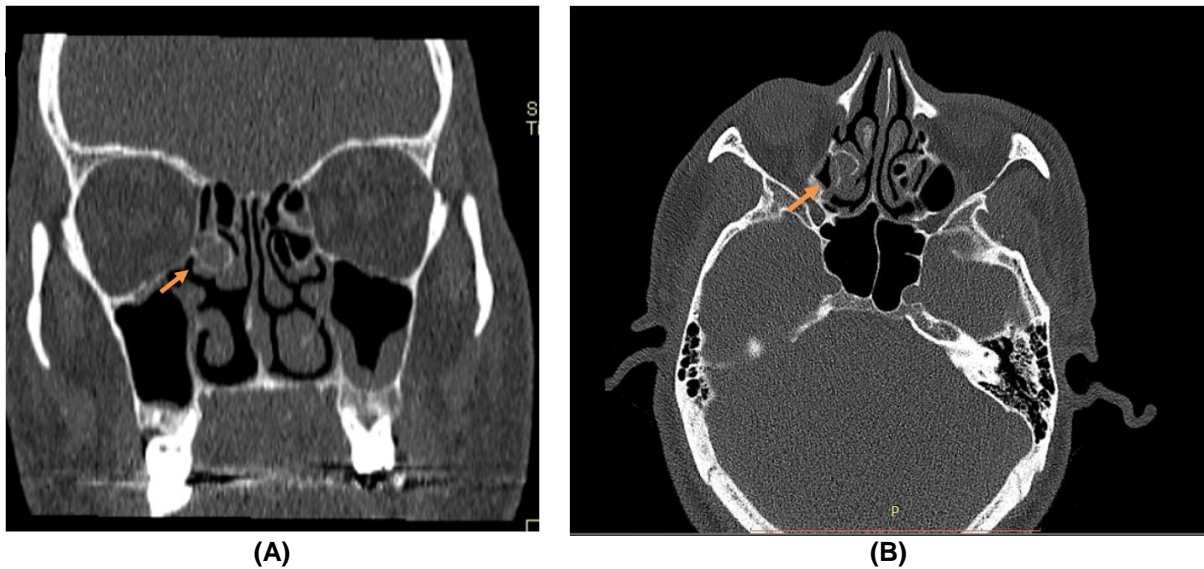


Fig. 3. (A) Coronal view (B) Sagittal view of CT Paranasal Sinus bone window setting showing mass at the anterior ethmoidal air cells with focal hyperostosis

3. DISCUSSION

Inverted papillomas are benign neoplasm originating from the Schneiderian membrane, the mucosa lining the nasal cavity and paranasal sinuses (Murphy et al., 2010). According to the World Health Organization (WHO) classification, nasal papillomas can be divided into three types: inverted, exophytic, and oncocytic papillomas (Nasal cavity, paranasal sinuses, nasopharynx, 2022).

A study by Schneyer et al. identified 90 attachment sites in a cohort of 83 patients (Nasal cavity, paranasal sinuses, nasopharynx, 2022).

For inverted/cylindrical papillomas, the maxillary sinus (42%) is the most common site of attachment, followed by the ethmoid sinus (18%), nasal cavity (15%), middle/superior turbinate (12%), sphenoid sinus (1.5%), and cribriform plate (1.5%). Fungiform papillomas have been described as originating from the septum or nasal vestibule, whereas inverted and cylindrical papillomas are more commonly found in the paranasal sinuses or lateral nasal wall.

The exact pathogenesis of inverted papilloma remains unclear, but several theories have been proposed. Metaplasia of the respiratory

epithelium into a stratified squamous epithelium is the most widely accepted theory. This metaplastic process is thought to be induced by irritant exposure, chronic inflammation, or viral infections like human papillomavirus (HPV) (Schneyer et al., 2011).

Radiologic assessment plays an important role in the preoperative evaluation of inverted papilloma, as it allows for precise determination of tumour extension, location, and surgical mapping for excision. There is insufficient evidence to suggest one specific modality as providing optimal imaging for inverted nasal papilloma. CT scanning is the initial modality of choice to evaluate the extent of the disease (Kakkad et al., 2023; Karkos et al., 2009), although the appearance on CT is non-specific. Inverted papillomas may present on CT as isodense unilateral homogeneous lesions, generally centred on the meatus nasi medius. Microcalcifications within the lesion are found in about 20% of cases, and bone lysis may suggest malignancy. Lee et al. found a strong correlation (89%) between focal hyperostosis seen on CT bone window settings and the surgically identified attachment site of inverted papillomas (Lee et al., 2007). Focal hyperostosis is an eccentric bone thickening and sclerosis that involves only a limited portion of the wall of a given paranasal sinus. In this case, calcification was observed at the superior margin of the mass at the anterior ethmoidal air cells (Fig. 3). Apart from CT scan, magnetic resonance imaging (MRI) also aids in managing inverted papilloma. The main role of MRI is to differentiate inverted papilloma and malignant tumours from inflammatory disease and accurate assessment of spread (Roobottom et al., 1995)

Surgery is the treatment of choice, either through an endonasal endoscopic or external approach, depending on the extension of the disease, tumour characteristics, and tailored according to the patient's condition (Busquets & Hwang, 2006; Han et al., 2001). Postoperative follow-up is critical for early detection of local relapse, which can occur early but may also present later. The seriousness of this pathology lies in the risk of carcinomatous evolution, which may be diagnosed at the outset or at recurrence during follow-up.

4. CONCLUSION

Although inverted papillomas comprise only 0.5–7% of all primary nasal tumours (Barnes, 2002),

they should be suspected in cases of unilateral polyps arising from the lateral nasal wall, particularly in male patients in the fifth to seventh decades of life (Buchwald et al., 1995). Diagnosis relies on a combination of imaging studies and histopathological examination. The mainstay of treatment for inverted papilloma, regardless of its site of origin, is surgical excision. Complete resection is essential to minimise the risk of recurrence and malignant transformation. In conclusion, while inverted papilloma is most commonly found on the lateral nasal wall and middle meatus, it can arise from various rare sites. Early recognition, appropriate imaging, and thorough surgical planning are key to achieving a favourable outcome.

CONSENT

All authors declare that written informed consent was obtained from the patient and patient agreed for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial office/Chief Editor/Editorial Board members of this journal whenever needed.

ETHICAL APPROVAL

It is not applicable.

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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