



# Unusual Locations of Inverted Papilloma

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## Abstract

Inverted papilloma is a tumor found involving the nasal cavity and paranasal sinuses. They are not cancerous but can be locally aggressive. They are most commonly seen involving the lateral wall of nose and maxillary sinus. When the tumor involves the frontal sinus and sphenoid sinus or the post ethmoid cells it presents a surgical challenge. Inverted papilloma almost always occur unilaterally and affect mainly the maxillary sinus. Inverted papilloma arising from the sphenoid and frontal sinuses are rare. Here we present two cases; one involving a 60 year man, with inverted papilloma originating from the frontal sinus and another case where it originated from the sphenoid sinus anterior wall and posterior ethmoidal cell (Onodi cell) and was resected endoscopically.

**Keywords** Inverted papilloma · Frontal sinus · Sphenoid sinus

## Introduction

Inverted papilloma also known as Schneiderian papilloma accounts for 0.5–4% of all sinonasal neoplasm. Inverted papilloma is a benign but a locally aggressive sino-nasal tumor that arises from the Schneiderian membrane, the ectodermal epithelium of the nose and paranasal sinus. It was so named due to its endophytic growth pattern in which projections of the tumour erode into the underlying stroma and bone [1]. Inverted papilloma has a well-known reputation for its tendency to recur after removal and its potential for malignant transformation [2] at an estimated risk of 5–15% [3].

The most common site of tumor origin is the lateral wall of the nasal cavity extending to maxillary sinus followed by ethmoidal sinuses. The frontal sinus is rarely involved with inverted papilloma and represent a significant surgical challenge due to the difficulty in endoscopic visualization and limitation of surgical access in this area [4]. In the report herein, we describe an Inverted papilloma originating in their region of frontal sinus. The sphenoid sinus inverted papilloma is further rare entity and the clinical features are often insidious and subtle with non-specific symptoms, with epistaxis, headache and visual disturbances being the most common symptoms.

## Case Report 1

A 60 yr old male presented in our outpatient clinic with a history of long standing left sided nasal obstruction which was associated with nasal discharge, hyposmia and headache. Past medical and surgical history was insignificant. No previous history of nasal trauma or nasal surgery. Nasal endoscopy showed polypoid mass. Computed Tomography (Fig. 1) was performed first and showed expansile soft tissue lesion involving frontal and ethmoid sinuses extending to nasal cavity. The epicenter was away from the lateral wall of nose. Thickening of bone was seen along the roof of the ethmoid which is more commonly seen in inverted papilloma. According to the age profile we thought patient would

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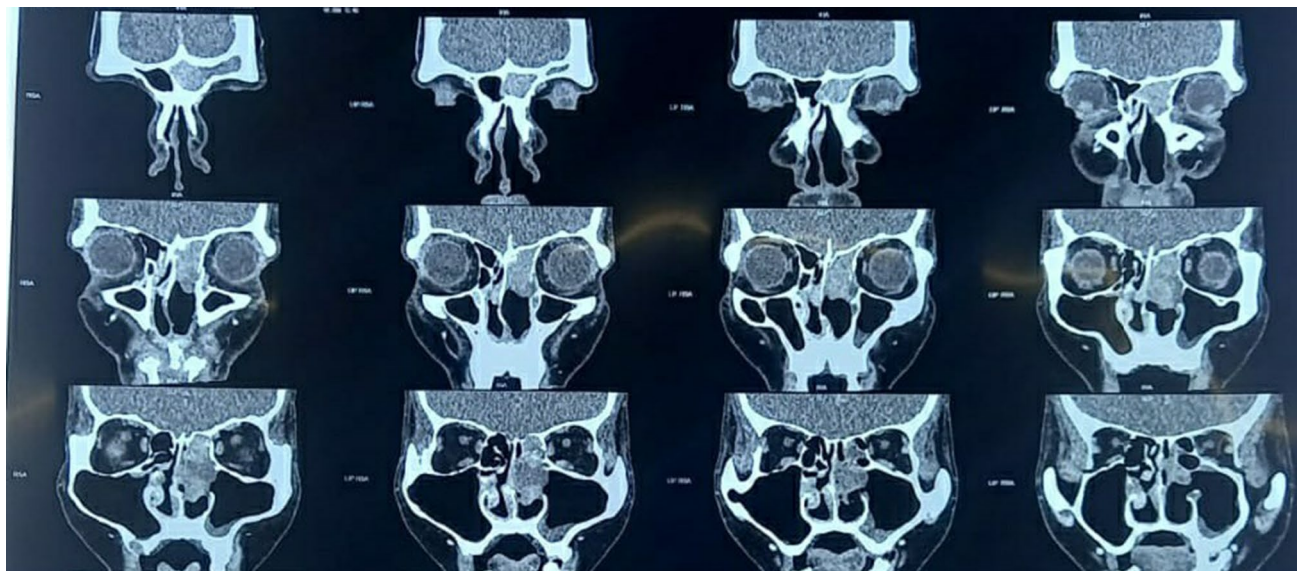
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**Fig. 1** CT SCAN showing soft tissue lesion involving left frontal and ethmoid sinus extending to nasal cavity

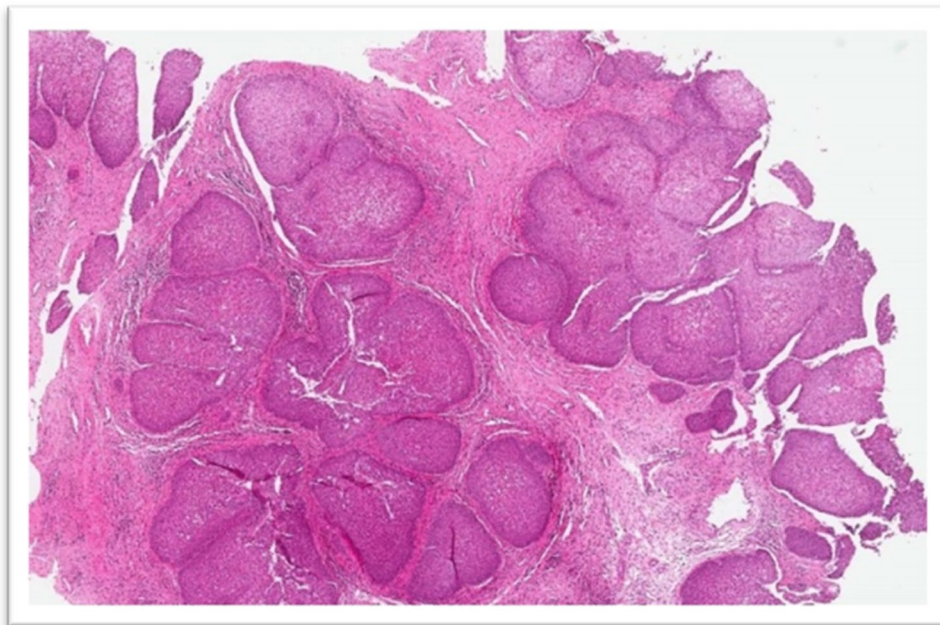
be having Aesthesioneuroblastoma (more common arising from the roof of ethmoid) but the biopsy report came as inverted papilloma.

Tumour removal was done endoscopically. The frontal sinus on the left had a challenging anatomy as seen in the CT scan. we followed the mass to its origin which was arising from mucosa of posterior table of frontal sinus and the interfrontal septum, Draf 3 frontal sinusotomy was done to do wide exposure of posterior table.

Histologic sections (Fig. 2) show an endophytic (inverted) pattern that grows downward into the underlying

stroma. The epithelium consists of hyperplastic squamous epithelium and ciliated columnar cells admixed with scattered mucocytes. The cells have a bland appearance and uniform nuclei. No cytologic atypia, increased mitotic activity or necrosis seen. Mixed inflammatory cells are dispersed within the epithelium and stroma [5].

**Fig. 2** Histopathology of inverted papilloma

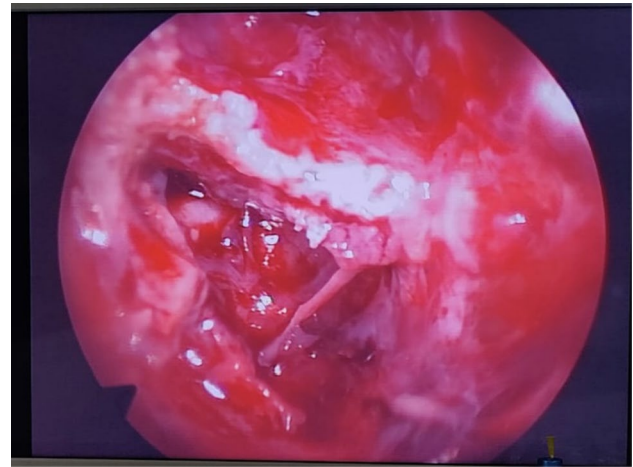


## Case Report 2

The patient, a 55-year-old woman, presented to our outpatient clinic with a 6-month history of epistaxis and headache. Then the patient was advised CT PNS. On doing CT scan a lesion was found involving sphenoid sinus and a onodi cell on the right side (Fig. 3). DNE revealed a fibro-papillomatous mass in the nasal cavity. Biopsy was taken and sent for HPE. Histopathologic study suggested inverted papilloma. The lesion was completely resected through a trans nasal endoscopic approach. On endoscopic removal we found that the lesion was arising from the anterior wall of sphenoid and the posterior ethmoid air cell. She had a onodi cell on the right and the optic nerve was lying free in it (Fig. 4). Drilling the underlying osteitic bone was a real challenge in the patient but we somehow managed it with a diamond drill keeping in mind the vital structures around. The maxillary sinus was free of tumor and we did not require to do the endoscopic denkers. Postoperatively, the patient did well and there was no evidence of recurrent disease on follow up.

## Discussion

Inverted papilloma is an uncommon, noncancerous tumor often discovered within sinuses or the nasal cavity. It can notably emerge from the lateral region of the nose. This unnatural growth was first ever documented by Ward in 1854. One of the primary areas you might find this type of tumor nesting is within the middle meatus area positioned on the side-wall of the nasal region, from where it can threaten to move into its neighbouring paranasal sinuses, especially



**Fig. 4** Intraoperative pic showing the exposed right optic nerve

nearing the maze-like ethmoidal structure or the hollow void of the maxillary antrum. However, keep in mind that if the condition becomes more progressive, it can even reach the frontal sinus, or the butterfly-shaped sphenoid.

Hyams presents the following incidences in terms of origin: maxillary sinus in 64% of the cases, ethmoidal in 25%, frontal in 8% and sphenoid in 4% [4–6]. Waitz e Wigand report the involvement of some paranasal sinus in 88% of the cases, and the anterior ethmoidal was the most frequent with 71% and the sphenoidal, the least frequent, with 10% [6]. Many authors report the sphenoid sinus as the least frequently affected by the inverted papilloma, with very few reports of involvement of this sinus alone [7].

Unilateral nasal obstruction is the most frequently reported manifestation of this disease. A lump that is fleshy,



**Fig. 3** CT Scan showing soft tissue lesion involving the anterior wall of sphenoid sinus, onodi cell and extending to right nasal cavity



pink, reddish-brown or grey, papillary or polypoid mass is often observed. Nasal obstruction may be accompanied by intermittent unilateral epistaxis, headache, and nasal discharge. These symptoms may persist for months to years. Imaging usually indicates changes in the nasal sinus on the same side as the symptoms. The most frequent observation on CTScan is opacification with low density. Bone destruction suggests a possible malignancy. If intracranial invasion is evident, an MRI scan is required to assess the characteristics of the neoplastic process. Typically, involvement of the frontal sinus is assessed during surgery.

It's possible to tackle frontal sinus effectively through modified endoscopic methods like Draf IIB or III. When the issue arises from the frontal recess level, a Draf type IIB frontal sinusotomy is ideal. For those who have inverted papilloma emerging from the mucosa of the frontal sinus's posterior wall, opting for a Draf type III frontal sinusotomy is crucial in creating a wide posterior wall exposure. Extensive mucosal involvement, particularly with hyperpneumatized frontal sinuses or a large pneumatized supraorbital air cell, necessitates a combined approach. Our particular case involved a mass that extended to the posterior table of the frontal sinus, resulting in a wide exposure necessitating a draf 3 frontal sinusotomy. The treatment was successful in clearing the disease, and post-operative follow-up revealed the patient to be healthy with no evidence of recurrence.

It is important to differentiate unilateral lesions of the sphenoid sinus from other inflammatory diseases or tumors. Since it can often occur, it should be differentiated from hemangioma, fibro-osteoma, schwannoma, pseudotumour, plasmacytoma, myxofibroma and osteochondroma which are all benign lesions [9]. When there are changes in the sphenoid sinus that are inflammatory a bilateral opacification with the lesion or the involvement of neighboring structures (posterior ethmoid cells) is usually presented. These changes appear as pathological masses on CT scans with smooth and clear contours as well as liquid components. In 2009, Guillemaud et al. conducted a systematic review and case study on nine patients who were diagnosed with an inverted papilloma in their sphenoid sinuses. This would make them the largest reported series in English literature [10]. Another case was reported by Rabelo et al. in 2015 [11]. Inverted papilloma occurring in the sphenoid sinus is a rare condition [12]. Its clinical presentation often involves gradual symptoms such as epistaxis, headaches and visual disturbances. CT scans often show that 70% of cases involve erosion of the bone [13]. Due to its proximity to structures like the internal carotid artery, cavernous sinus and cranial nerves II, III, IV, V and VI it is crucial to identify the clinical features of sphenoid sinus inverted papilloma early on to prevent severe complications [8, 10]. For years the standard treatment for inverted papilloma has been maxillectomy and complete tumor removal through lateral rhinotomy or

a midfacial degloving approach [14]. However, in years an invasive method using endoscopic transnasal approach has become more popular. This approach offers benefits such as time and reduced postoperative complications. Additionally, it minimizes deformities. Allows for shorter hospital stays, after surgery [3, 11]. Our patient underwent endoscopic resection without any intraoperative or postoperative complications [15, 16].

## Conclusions

Inverted papilloma arising from frontal sinus is very rare. If it occurs it can be treated either with endoscopic approach or osteoplastic flap approach depending on the extent with good results. The ideal management should always target a complete excision of all the affected mucosa.

Inverted papilloma involving the sphenoid sinus is also a rare entity. Nonspecific symptomatology and clinical presentation make this kind of tumor a diagnostic and therapeutic challenge for otolaryngologists. CT scan, and DNE are done preoperatively to evaluate. Complete endoscopic removal with drilling of the underlying osteitic bone is always the treatment of choice.

Close follow-up is required to detect possible recurrences and malignant transformation.

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## Declarations

**Conflict of interest** The authors declare that they have no conflicts of interest to disclose.

**Ethics Approval** This work was granted permission approval from Ethics committee of the concerned Institute.

**Informed Consent** Written Informed consent for publishing in Springer Nature was taken from both the patients.

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