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Aggressive Inverted Sinunasal Papilloma with Intracranial Invasion: Case Report

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Abstract | **Case Report**

Introduction: Inverted papilloma (IP) is a benign neoplasm of the paranasal sinuses that, despite its non-malignant nature, exhibits aggressive local behavior with the potential for recurrence and, in rare cases, intracranial invasion. Originating in the Schneiderian mucosa, this tumor represents between 0.5 and 4% of primary nasal tumors, primarily affecting men in their fifth and sixth decades of life. The pathogenesis of IP has been linked to multiple factors, including viral factors such as the human papillomavirus (HPV); however, its exact etiology remains under investigation. [1] ***Clinical case:*** We present the case of a 56-year-old male patient with symptoms of nasal airway obstruction and headache, who was diagnosed with an inverted papilloma after rhinoendoscopy and computed tomography revealed invasion of the skull base. A complex surgical intervention was performed, including craniotomy and tumor excision, with a histopathological report confirming the diagnosis. Three months postoperatively, the patient showed no surgical scarring or recurrence. After two years of follow-up, no recurrence was observed, and the patient received adjuvant oncological treatment. ***Discussion:*** Although inverted papilloma is benign, its potential for malignant transformation and characteristics of local invasion are concerning. The incidence is low, and intracranial invasion is rare, with craniofacial resection being the most effective technique in these cases. It is crucial to avoid conservative treatments, given the risk of recurrence. Long-term follow-up is recommended due to the possibility of late recurrences. [3] ***Conclusion:*** Inverted papilloma with cranial invasion, although infrequent, require careful surgical management and long-term follow-up to ensure a favorable prognosis. Endoscopic resection is effective in cases without intracranial extension, while adjuvant radiotherapy may be beneficial in incomplete resections. [5]

Keywords: inverted papilloma, intracranial, nasal tumor.

INTRODUCTION

Inverted papilloma is a locally aggressive benign neoplasm of the paranasal sinuses with a high potential for recurrence and malignant transformation. It was first described by Ward in 1854. Ringertz provided a detailed histology description of this tumor in 1938, and Respler linked its etiology to a viral agent in 1987. [1] It is the most frequent and representative of the three types of intranasal papillomas classified by the WHO. [2]

Several predisposing factors have been proposed, such as atopic processes, chronic inflammation, and occupational exposure, although none of them have a clear association like HPV infection. [2]

Histologically, it originates from the Schneiderian membrane that lines the nasal cavity and paranasal sinuses; strictly speaking, it is not a papilloma but a polypoid change of the nasal mucosa accompanied by severe metaplasia within the polypoid tissue, both of the respiratory epithelium and the glandular ducts, which can even be difficult to distinguish from a low-grade squamous cell carcinoma. [3]

They usually arise from the lateral ethmoid complex of the nasal wall, although the nasopharynx,

oropharynx, middle ear, nasal septum, lacrimal duct, and frontal, sphenoid, and maxillary sinuses have been reported as sites of origin. [4]

Despite being a benign pathology, three characteristics of this tumor define its recurrence rate, local aggressiveness, and potential for malignant transformation: atypia, dysplasia, and carcinoma in situ or squamous cell carcinoma. Local invasion usually causes bone destruction, and intracranial extension is rare in the absence of malignant transformation. If malignant transformation does occur, it often presents a surgical challenge due to the need to reconstruct the anterior skull base and achieve excision of as much tissue as possible, since the prognosis for patients with intracranial extension depends largely on the extent of resection and the degree of dural invasion. [5]

Clinical Case

A 56-year-old male patient reports a clinical picture of approximately 5 years' evolution characterized by bilateral nasal ventilatory insufficiency with right predominance associated with anosmia, clear rhinorrhea and occasional frontal headache treated without response with non-steroidal analgesics.

Physical examination revealed right upper eyelid edema, without impairment of eye movements, and preserved visual acuity. Rhinoendoscopy revealed a pinkish-yellow mass with irregular borders in both nasal cavities, originating from the medial wall of the maxilla and middle meatus.

Results

In computed tomography of the craniofacial complex, occupation by isodense soft tissue density of the frontal sinus, anterior and posterior ethmoid cells, maxillary and bilateral sphenoid.

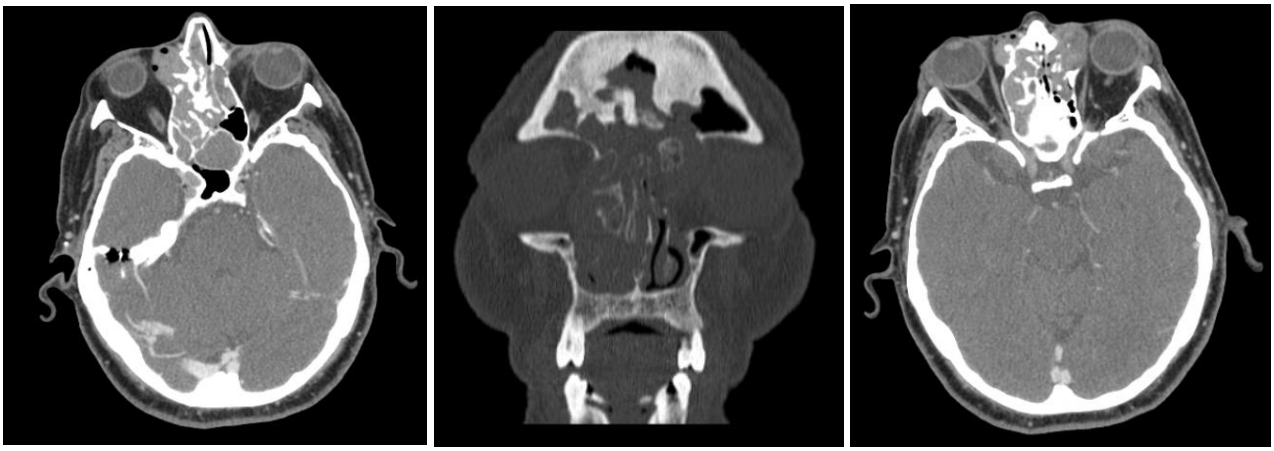
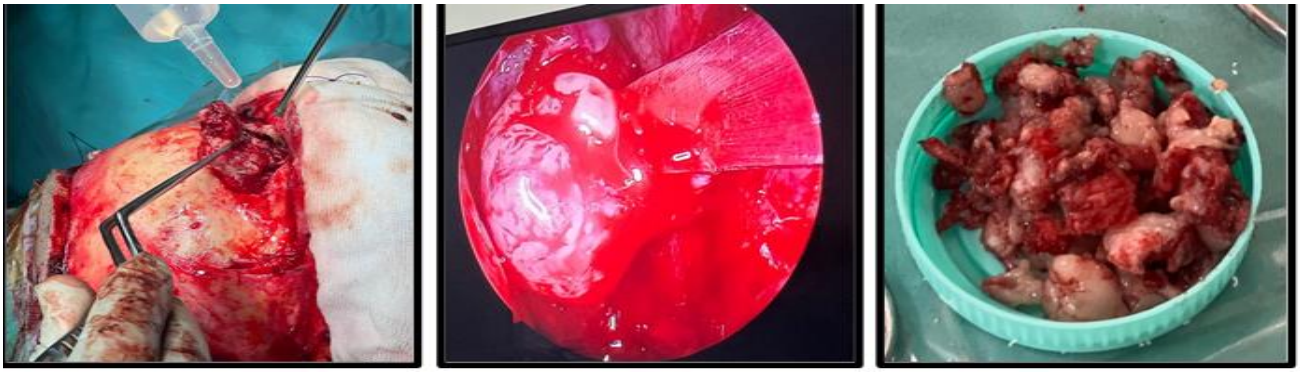


Figure 1: Computed tomography of the craniofacial complex shows soft tissue density occupation in both nasal fossae, predominantly on the right, maxillary sinus, anterior and posterior ethmoid, frontal and bilateral sphenoid with erosion of the lamina papyracea and invasion of the anterior skull base

A biopsy of the tumorous lesion was performed, reporting it as an inverted Schneiderian papilloma (HFA). Subsequently, surgical intervention was performed in conjunction with the neurosurgery service. They made an incision with a bicoronal approach, a musculocutaneous flap was raised, the pericranium was debrided, and a craniotomy of approximately 5cm x 4cm was performed on the external table of the frontal sinus. The sinus was exposed, and a lobulated tumor with a grayish coloration and solid areas was observed. Excision of the tumor was completed, infiltrated bone tissue was drilled, a flap was created using fascia from the temporalis muscle and pericranium, the craniotomy was repositioned, and it was closed in layers. An endoscopic intervention was performed through the right nasal cavity with a 30-degree endoscope. A grayish tumor with calcified areas was identified and excised in blocks. The inferior turbinate was identified and preserved. A bony component of the tumor was evident at the level of the middle meatus, so it was decided to perform Uncinectomy, maxillary antrostomy; right turbinectomy is decided upon due to tumor invasion, preserving the middle turbinate axilla as an anatomical reference; anterior and posterior ethmoidectomy and sphenoidectomy; a tumor occupying the orbital region is resected; the skull base is clear; the frontal floor has fascia previously placed by neurosurgery; through the left nasal cavity, an anterior ethmoidectomy is performed, revealing a clear skull base, a clear posterior ethmoid and sphenoid; hemostasis is confirmed. Tumor samples of different characteristics are sent for pathological examination.



A delayed anatomopathological report was received which confirms the presence of sinus mucosa completely replaced by proliferation of inverted Schneiderian papilloma (HFA), with foci of dysplasia.

A follow-up CT scan was performed at 3 months, which showed a surgical scar; the patient was undergoing cancer treatment.

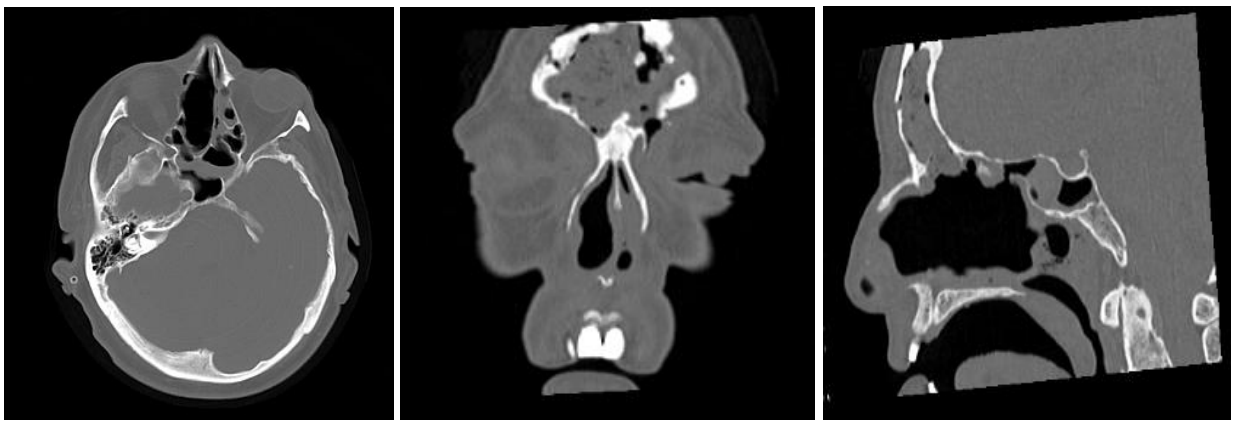


Figure 2: Computed tomography of the craniofacial complex, axial, coronal and sagittal cuts, shows surgical scar

After 2 years of clinical follow-up, imaging studies and nasofibroscopy; the patient does not present a recurrence, and received adjuvant oncological treatment.

DISCUSSION

Inverted papilloma is a benign neoplasm of the Schneiderian epithelium of the upper airways with an incidence of 0.6/100,000 people per year, comprising approximately 0.5 to 4% of primary nasal tumors. It generally presents between the fifth and seventh decades of life with a male-to-female ratio of 2:1 to 8:1. [2]

Histologically, it derives from the mucosa of the nasal cavity. Its diagnosis is made by demonstrating the characteristic inversion of epithelial hyperplasia, with its intact basement membrane, intercalated mucocysts, and certain degrees of cellular atypia. Magnetic resonance imaging and computed tomography of the facial skeleton are useful for determining the extent of tumor invasion and for surgical planning. [3-4]

Intracranial invasion is extremely rare. In a study by Miller et al., 1469 cases were reviewed and only five showed extension into the anterior cranial fossa. [6]

In cases of inverted papillomas with intracranial extension, craniofacial resection is the technique described with the best reported results. However, it is difficult to evaluate the efficacy of craniofacial resection and compare it with other treatment modalities due to the limited number of cases with long-term follow-up and the small number of reported cases. [7]

The persistence of residual intracranial cells will almost invariably lead to recurrence and dural invasion; therefore, conservative treatment should be avoided and reserved only for patients with unresectable lesions or severe comorbidities that limit life expectancy. Adjuvant radiotherapy appears to offer some benefits, particularly in advanced cases or those with incomplete resection. [8,9]

The administration of corticosteroids as preoperative medical therapy can help decrease mucosal inflammation and minimize intraoperative bleeding. Adjuvant chemotherapy may be used in established cases of malignancy. [10]

Long-term follow-up for a minimum of 5 years is recommended, including endoscopy with exploration of the nasal cavity and paranasal sinuses, given the

capacity of this condition to recur even at intervals greater than 5 years after treatment. [9-10]

CONCLUSION

Benign inverted papillomas with cranial invasion represent a rare variant. The prognosis depends on dural invasion in addition to achieving complete resection. Craniofacial resection has proven effective in terms of complete disease elimination and follow-up free of recurrence. Endoscopic resection is the standard treatment for inverted papillomas in the absence of intracranial extension, with various techniques described. Adjuvant radiotherapy appears to offer some benefits, particularly in cases of incomplete resection.

Conflicts of Interest: The authors declare that there is no conflict of interest regarding the publication of this paper.

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