



Inverted papilloma with bilateral origin.

A case report and review of the literature



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Inverted papilloma with bilateral origin. A case report and review of the literature

AIM: To show a rare case of inverted papilloma with bilateral origin treated with endoscopic approach

MATERIAL OF STUDY: The authors reported the unusual case of a 71-year-old male patient with a growing mass occupying both nasal cavities. The patient complained bilateral nasal obstruction associated with thick nasal secretions, anterior and posterior nasal drip, snoring and diffuse nasal pain. The biopsy revealed inverted papilloma.

RESULTS: CT Scan and RMI of paranasal sinuses showed a bilateral origin of the tumor with a triple involvement on the left side: middle turbinate axilla, frontal recess posterior wall and ethmoidal sinus roof. Nasal endoscopic surgery approach was performed and the tumoral mass was removed completely.

DISCUSSION: Inverted papilloma is an uncommon primary nasal tumor that presents three typical characteristics: a high rate of recurrence, local aggressive behaviour and association with malignancy. Although bilaterality is very rare, in this case the neoplasia occupied both nasal cavities with extension to left frontal, sphenoid and ethmoid sinuses.

CONCLUSION: The authors, in line with the literature, showed that endoscopic approach represents once again a safe and efficient technique, even for larger tumors.

KEY WORDS: Bilateral origin, Inverted papilloma, Paranasal sinuses

Introduction

Inverted papilloma is a tumor of the nasal cavities and paranasal sinuses, with 3 main characteristics: a relative local aggressive behavior, a high capacity for recurrence and, lastly, a possible association with carcinoma ¹.

According to the World Health Organization (WHO) 2005 classification, inverted papilloma is grouped with the Schneiderian papillomas because arises from the Schneiderian membrane, like oncocyctic and exophytic papillomas. ² On the fourth edition of the World Health Organization classification of head and neck tumors

Schneiderian papillomas were reclassified as sinonasal papillomas ³.

As a group Schneiderian papillomas are uncommon, representing only 0.4-4.7% of all sinonasal tumors and between the different subtypes inverted papilloma is the most prevalent. Inverted papilloma characteristically arises from the lateral nasal wall in the region of the middle turbinate or ethmoid recesses and often extend secondarily into the sinuses, especially the maxillary and the ethmoid, and to a lesser extent, the sphenoid, and the frontal. Although reported to be bilateral in 0-10% of cases, such an occurrence should always arouse the suspicion of septal erosion and perforation from a unilateral disease ^{4,5}.

Pathogenesis is not entirely clear but the exposure to organic solvents and welding fumes seems to have a determining role, while the presence of human papilloma virus (HPV) seems to be more involved in the progression and the recurrence of the disease ^{6,7}.

Patients with inverted papilloma typically present with nonspecific signs and symptoms. The most common is

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unilateral nasal obstruction followed by epistaxis, rhinorrhoea, hyposmia and frontal headache. Pain is a rare presenting complaint and it usually represents a malignancy or a secondary infection ⁸.

Pathological examination is essential to diagnosis ¹ and the radiological assessment has two main objectives: precise determination of the tumor extension and localization of the tumor site. Computed tomography scanning was the standard imaging technique of choice for assessing sinus involvement of inverted papilloma and planning the extent of the surgery. T2-weighted magnetic resonance imaging scans were able to distinguish the tumor (intermediate signal) from the surrounding inflammatory tissues (bright signal) ⁹.

Despite being a benign tumor, there are various staging systems that may be employed, all of them using the extent of disease, measured by radiographic or endoscopic findings, as main parameter ³. Although Krouse's classification is the most popular staging system, other authors like Canada et al. and Han and al. proposed their staging system ¹⁰.

Until the mid-1990s, the gold-standard treatment was surgery on an external approach. Endoscopic treatment for IP was first described in 1992 by Waitz and Wigand, and by Kamel in the same year, and it is now for many authors the new gold-standard ^{1,11,12-15}.

This report describes a particular rare case of bilateral inverted papilloma with a triple origin on the left sinus side.

Case Report

A 71 year-old male was admitted to our hospital complaining bilateral nasal obstruction for 10 years associated with thick nasal secretions, anterior and posterior nasal drip, snoring and diffuse facial pain. He did not complain of epistaxis or visual impairments.

The otorhinolaryngology exam revealed a mass that completely occluded the nasal cavity bilaterally, mucosal pale-

ness associated with mucopurulent secretion and posterior nasal drip.

Under local anaesthesia a biopsy of the mass was performed, and the sample was sent for histopathological examination. The result was sinonasal inverted papilloma.

Radiological assessment was also performed. The CT scan showed sinonasal polyposis involving frontal, maxillary, ethmoid and sphenoid sinuses bilaterally, and osteomeatal complexes obstruction without bony lesion. The Magnetic Resonance Imaging, with gadolinium, of the nose and paranasal sinuses showed a large polypoid mass occupying both nasal cavity with extension to left frontal, sphenoid and ethmoid sinuses, with associated mucosal hypertrophy of right maxilla that was filled with fluid; the mass showed a heterogeneously high signal in T2 and low signal in T1, with heterogeneous enhancement and no local invasion or cavernous extension, but penetration of the the mild part of the nasal septum.

The patient then underwent nasosinus endoscopic surgery that showed a bilateral location of the disease and a triple origin on the left side: middle turbinate axilla, frontal recess posterior wall and ethmoidal sinus roof. We performed the debulking of the mass using the microdebrider, bilateral type one medial wall maxillectomy, Draf type III frontal sinusotomy with free inferior turbinate flap. Then the posterior ethmoid roof and bone surrounding the paraclival internal carotid artery was drilled after removing the pterygoid process.

The submitted material has been subjected to histopathological analysis and the report confirmed inverted papilloma diagnosis and with no malignancy sign.

Antibiotic-antihistamine therapy was subsequently set and after three days the whole intra nasal pack was removed.

Histopathological report confirmed inverted papilloma diagnosis and didn't show any malignancy sign.

Two weeks after surgery, paranasal sinus CT scan was performed, and it didn't show any sign of tumor residual (Figs. 1, 2).



Fig. 1: Endoscopic view of left nasal cavity: directly vision, after removal of middle turbinate head and access on sphenoidal sinus.

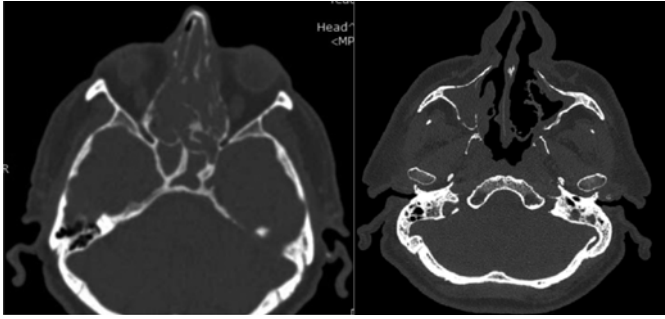


Fig. 2: Paranasal sinuses CT scan, axial section. Before and after the surgery.

Discussion

Inverted papilloma is the most common benign sinonasal tumour, with an incidence of 0.6-1.5 cases per 100 000 inhabitants per year¹⁶. The name inverted is justified by the endophilic growth of the superficial epithelium inside the adjacent stroma.¹⁷

Nasal polyps are 25 times more frequent than inverted papilloma, presenting respiratory mucosa with stromal edema and eosinophilic infiltrate which are the main characteristics for differential diagnosis¹⁹.

Inverted Papilloma is associated with malignancy, presenting a malignant transformation rate of 1.9-15%. Squamous Cell Carcinoma is the predominant histologic type developed but mucoepidermoid carcinoma and sinonasal undifferentiated carcinoma have also been described. Sbrana et al. study showed a malignant transformation rate of 13.64% on a total of 69 inverted papilloma surgeries, and all these patients were staged T3 or T4 in Krouse staging system which suggests that the extension of the tumor might have an association with malignant transformation^{20,21}.

The role of some risk factors in the tumor recurrence is still being studied: some authors showed that inverted papilloma recurrence is strongly related to specific risk factors, including incomplete surgical removal, stage of disease, site of the lesion, surgical technique, and malignancy rate; other studies, however, showed that the rate of recurrence did not correlate with any of the following: tumor stage, surgical approach, presence of squamous cell carcinoma, whether the surgery was primary or revision, the presence and location of focal hyperostosis on computed tomography. No statistical significance was found to associate smoking with recurrence²²⁻²⁴.

Surgical procedures of sinonasal inverted papilloma are planned according to the origin of the tumor rather than its extent. Origin targeted surgery and proper management of the site of attachment are the key to achieve a complete surgical resection of sinonasal inverted papilloma²⁵. Endoscopic surgery is the gold standard treatment for the vast majority of inverted papillomas. Depending on the expertise of the surgical team, the

use of enhanced endoscopic procedures, such as medial maxillectomy, Draf 2 or 3, and expanded sphenoidotomy, may allow more than 90% of the cases to be treated successfully²⁶.

The development of new techniques and more angulated instruments for endoscopic surgery could potentially increase the access to more difficult areas, such as the frontal sinus and the anterior and lateral maxillary sinus walls, further improving the surgical outcomes²⁷.

Radioterapy is a safe and valuable treatment option for histologically overt benign inverted papillomas. It is indicated when there is a high risk of tumor recurrence after surgery and in inoperable tumors²⁸.

Conclusions

Many cases about inverted papilloma have been described in literature and bilateral origin is extremely rare. In our case, the inverted papilloma, not only showed a bilateral origin, but on the left side it had a triple origin site extending from the nasal cavity to sphenoidal sinus. We can say that endoscopic approach represents once again a safe and efficient technique, also for very expanded tumors.

Riassunto

Il papilloma invertito è un tumore delle cavità nasali e dei seni paranasali che presenta tre caratteristiche: una discreta aggressività locale, una marcata capacità di ricorrenza e una possibile associazione con tumori maligni, spesso carcinomi squamocellulari. Secondo la nuova classificazione WHO dei tumori testa e collo, rientra, con il papilloma oncocitico ed il papilloma esofitico, tra i "papillomi dei seni paranasali" che, nel loro insieme, rappresentano una bassa percentuale (0.4-4.7%) di tutti i tumori dei seni paranasali. La bilateralità è molto rara mentre il tasso di trasformazione maligna si attesta intorno al 13-14%. Si manifesta, di solito, con ostruzione nasale monolaterale, epistassi, rinorrea, iposmia e cefalea frontale. L'esame istopatologico è essenziale per la diagnosi mentre le indagini strumentali, come la TC del Massiccio Facciale, permettono di identificare la sede della lesione e di valutare l'espansione e l'eventuale erosione ossea del tumore. Il trattamento di scelta prevede l'escissione di tutta la massa tumorale spesso per via endoscopica mentre il trattamento radioterapico adiuvante è riservato ai casi in cui sono presenti focolai di carcinoma. Il caso clinico esaminato riguarda un paziente di 71 anni giunto in ospedale per ostruzione nasale associata a rinorrea mucosa, russamento e diffuso dolore facciale. L'esame otorinolaringoiatrico ha mostrato la presenza di una massa oblitterante entrambe le cavità nasali che, in seguito a biopsia, risultava essere un papilloma invertito. Attraverso tecniche di imaging quali TC e Risonanza

Magnetica del Massiccio Facciale è emerso che il tumore originava da entrambe le fosse nasali, a destra a livello dell'ascella del turbinato medio mentre a sinistra aveva una tripla sede di origine: ascella del turbinato medio, parete posteriore del recesso frontale e tetto dell'etmoide. Il paziente, sotto anestesia generale e per via endoscopica, è stato sottoposto a rimozione della massa tumorale tramite "debrider", a maxillectomia mediale di tipo I sinistra, a senectomia frontale "Draf III" e ad asportazione della massa dal tetto etmoidale fino al livello della porzione paraclivale dell'arteria carotidea interna. Il materiale asportato è stato analizzato e l'esame istopatologico ha confermato la diagnosi di papilloma invertito escludendo comunque segni di malignità. Due settimane dopo l'intervento, il paziente è stato sottoposto ad un'ulteriore TC del Massiccio Facciale che non ha mostrato nessuna presenza di residui tumorali. La chirurgia endoscopica nasale si è dimostrata, ancora una volta, una strategia efficace nel trattamento anche di tumori di grosse dimensioni.

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