CASE REPORT

Rhinology, 40, 220-222, 2002

A rare location of bilateral inverted papilloma of the nose and paranasal sinuses*

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SUMMARY

Inverted papilloma is a rare benign sinonasal tumour, characterized by a potentially invasive nature. The lateral nasal wall represents the most common site of origin, whereas paranasal sinuses are involved by extension quite frequently. In contrast, primary sinus inverted papillomas have rarely been reported. Although the midfacial degloving approach has historically been the procedure of choice, recent technological advances have rendered endoscopic sinus surgery a safe procedure with equivalent success rates and low probability of papilloma recurrence.

We present a rare case of bilateral inverted papilloma arising from the sphenoid septum and extending towards both the sphenoid and posterior ethmoid sinuses and the posterior section of both nasal cavities, while it slenderizes the sella turcica by compression and elevates the pituitary gland. The bilateral inverted papilloma was successfully removed with a transnasal endoscopic procedure. There is no evidence of recurrence for a follow up period of 1.5 years postoperatively. We conclude that it is a rare entity, which can be treated successfully with endoscopic sinus surgery in the hands of an experienced otorhinolaryngologist.

Key words: inverted papilloma, sphenoid sinus, solitary lesion

INTRODUCTION

An interesting case of a bilateral inverted papilloma was encountered in our department. The mass seemed to arise from the sphenoid septum and extended towards both the sphenoid and posterior ethmoid sinuses and the posterior section of both nasal cavities, while it slenderized the sella turcica by compression and elevated the pituitary gland. Although the midfacial degloving approach has historically been the procedure of choice, recent technological advances have rendered endoscopic sinus surgery a safe procedure with equivalent success rates and low probability of papilloma recurrence. The papilloma was successfully removed with a transnasal endoscopic procedure. There is no evidence of recurrence for a follow up period of 1.5 years postoperatively. To our knowledge, this is the very first case of bilateral inverted papilloma of such origin and location treated endoscopically.

CASE REPORT

Patient

A 52-year old male presented to our clinic with a 3-year history of nasal fullness, purulent nasal discharge and postnasal drip. He also complained of several bouts of retrobulbar headache radiating to the occiput. He reported a negative past medical and surgical history. His habits include a 25-year history of smoking (1 pack/d).

On physical examination, anterior rhinoscopy and endoscopy with the 30° 4-mm rigid endoscope revealed accumulated purulent secretions in both nasal cavities and large polypoid structures occupying and obstructing both posterior nasal choanae. The latter seemed to emerge through the posterior ethmoid cells or, possibly, the sphenoid sinus.

Computed tomography (Figure 1) and magnetic resonance imaging (Figure 2) of the nose and the paranasal sinuses showed homogeneous opacification of both the sphenoid sinuses and posterior ethmoid cells indicative of large polypoid masses, while extending and obliterating the posterior section of both nasal cavities, particularly the right one. The anterior wall of the sphenoid sinuses was eroded, thus enabling protrusion of the mass through both nasal cavities. The mass slenderized the sella turcica by compression and elevated the pituitary gland. The carotid ostia, cavernous sinuses and meningeal sheaths appeared intact.

Presented in the IX Congress of the International Rhinologic Society "The Nose 2000…And Beyond", Washington, DC, USA, 20-23 Sept., 2000

* Received for publication: November 19, 2001; accepted: July 17, 2002

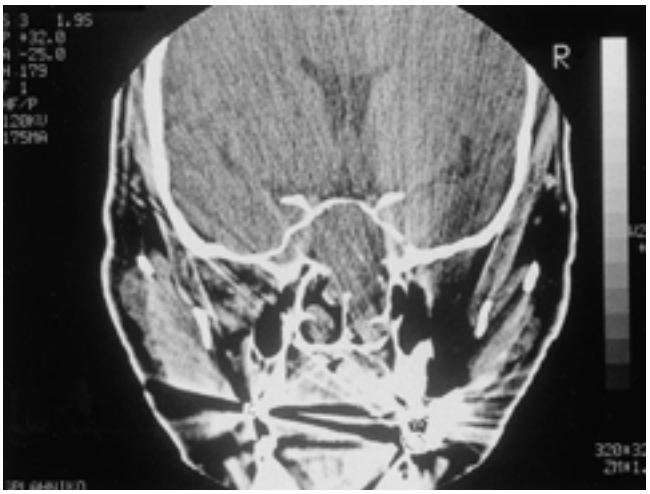

Figure 1. Coronal CT view of a large opacifying lesion occupying the entire sphenoid area and elevating the sella turcica by compression.

Due to the suspicious macroscopic appearance and radiographic evidence, biopsy specimens were taken under local anaesthesia from both nasal cavities. The pathology report came back with the diagnosis of bilateral inverting papilloma without any evidence of malignant transformation.

The patient was scheduled for endoscopic removal of the papilloma. Under direct visualization with the use of a 0° and a 30° 4-mm rigid endoscope, the mass was meticulously removed with Blakesley-Weil forceps at a piece-meal fashion. Extreme care was taken to avoid disruption of the superior, posterior and lateral walls of the sphenoid sinus, which are in contact with vital structures. A curved curette was used to gently remove the mass from the most lateral areas of the sphenoid cavity. The vomer was partially excised, in order to facilitate removal of the mass. The nasal cavities were packed upon completion of the procedure. The patient tolerated the operation without preoperative and postoperative complications. He had an uneventful hospital stay and was finally discharged in good condition on the following day.

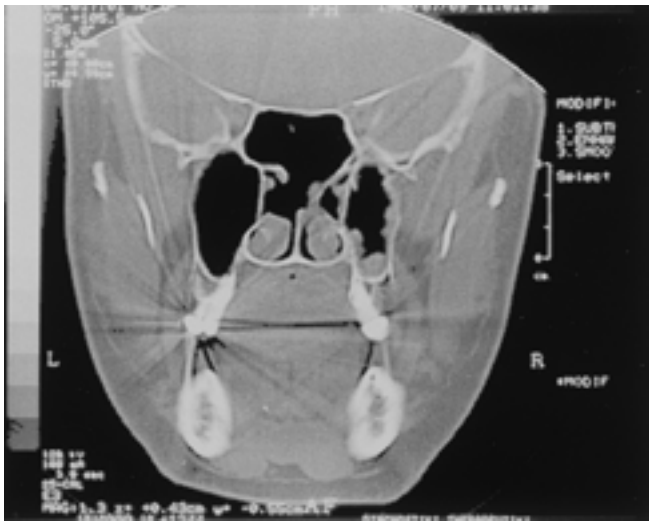

Figure 3. Postoperative coronal CT view of the same patient, showing complete removal of the mass without any signs of recurrence.

Figure 2. Axial T2-weighted MRI view of the sphenoid sinuses, demonstrating their complete opacification from the mass.

Computed tomography scan (Figure 3) and magnetic resonance imaging on the first postoperative year did not show any recurrence. As up today, the patient is free of disease. The final pathology report of the removed tumour was negative for malignancy.

DISCUSSION - CONCLUSIONS

Inverted papilloma is a locally aggressive sinonasal tumour that arises from the outlining Schneiderian membrane when it inverts into the underlying stroma (Batsakis, 1979). It has a male predominance and usually appears in the fifth and sixth decades of life (Lawson et al., 1989). Although classified as benign, it has a notorious reputation for high recurrence rate and malignant transformation, the latter ranging from less than 2% to 53% (Lampertico et al., 1963; Hyams et al., 1971; Dolgin et al., 1992). According to several studies, the reported average recurrence ranges from 7% after radical extranasal excision to 59% after non-endoscopic endonasal excision (Sham et al., 1998). Paranasal sinuses may often be involved by extension, since the tumour originates mainly from the lateral nasal wall, but they are rarely reported as the primary sites. Reviewing the whole literature on primary inverted papillomas, one can hardly find any located exclusively in the sphenoid sinus. To our knowledge, it is the first case of primary inverting papilloma emerging from the septum of the sphenoid sinus and extending bilaterally anteroposteriorly.

Sham et al. (1998) have summated the options of interventional treatment into four categories:

1. Non-endoscopic endonasal approaches e.g. polypectomy or sphenoidectomy
2. Limited extranasal approaches e.g. Caldwell-Luc or external ethmoidectomy
3. Radical extranasal approaches e.g. lateral rhinotomy or midfacial degloving with en-bloc resection of the lateral nasal wall
4. Endoscopic endonasal excision

The efficacy of number 4 in diminishing tumour recurrence has been questioned. In fact, the preference of open procedures to endoscopic endonasal excision has consistently been advocated. The justification for this opinion stems from the belief that there is better access to the paranasal sinuses with open techniques, thus enabling better removal of the mass and ensuring that no tumour remnants are left behind, which would later lead to recurrence. For these reasons, the traditionally recommended by many treatment of choice is medial maxillectomy and en bloc tumour resection through a lateral rhinotomy or a midfacial degloving approach (Cummings et al., 1970; Vrabec, 1975; Segal et al., 1986; Phillips et al., 1990).

The recent advances of endoscopic sinus surgery have tempted some surgeons into attempting endoscopic removal of the tumour with comparable success rates (Sham et al., 1998; Waitz et al., 1992; Stankiewicz et al., 1993; McCary et al., 1994; Kamel, 1992). However, Wigand's results of 17% recurrence rate have been questioned on the basis of bias attributed to selection of patients with smaller and less aggressive lesions (Bielamowicz et al., 1993). Kamel (1992) concluded that lesions involving the maxillary sinus should be treated with transnasal medial maxillectomy, leaving the endoscopic procedure to be effective for the remaining paranasal sinuses. Stankiewicz et al. (1993) suggested application of endoscopic approach for limited inverting papilloma located in the ethmoid sinuses, the sphenoid sinus, the lateral nasal wall and the medial wall of the maxillary sinuses. Among others, Sham et al. (1998) summarized that the endoscopic approach is inadvisable for removal of tumours in endoscopically inaccessible areas of the maxillary, frontal or sphenoid sinuses, as well as for those extending to the orbit or intracranial structures.

With all of the above taken into consideration, we decided to attempt endoscopic removal of a bulky, infiltrative lesion occupying the entire sphenoid and partial ethmoid sinuses bilaterally and obliterating both posterior nasal choanae. However, the patient had already been informed of the possibility of converting the endoscopic procedure to an open one, should inability to adequately access and completely remove the tumour appear. The mass seemed to emerge from the sphenoid septum, as it was easily mobilized from the roof and posterior walls of the unified sphenoid sinuses as one single chamber. The lateral, anterior and posterior walls of this single cavity were completely resorbed, thus permitting expansion of the mass to the posterior ethmoid sinuses and nasopharynx, especially to the right side. The mass was excised at a piece-meal fashion with Blakesley-Weil forceps. To enable better access and removal of the mass, the vomer was partially excised. As success of any intervention is dependent upon complete removal of the tumour and not particularly in the type of operation performed, we were extremely meticulous not to leave any remnants behind and to carefully strip off the underlying diseased mucosa.

There is no doubt that most recurrences occur early in the postoperative course (Waitz et al., 1992). The fact that there is no proven recurrence after one postoperative year provides good evidence of a successful outcome. However, a 3-year follow-up with careful endoscopy and imaging would be sufficient to fortify the success of the procedure.

The idiosyncratic clinical behaviour of inverting papilloma renders it a therapeutic challenge for every rhinologist. In our case, the rarity of its location and extent made it a unique and thrilling experience.

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