Isolated Inverted Papilloma of the Sphenoid Sinus

Shao-Cheng Liu¹, Jih-Chin Lee¹, Jiann-Jy Chen², Yaoh-Shiang Lin¹,³*

¹Department of Otolaryngology–Head and Neck Surgery, Tri-Service General Hospital, National Defense Medical Center, ²Faculty of Medicine, School of Medicine, Fu Jen Catholic University, and ³Medical Affairs Bureau, Ministry of National Defense, Taipei, Taiwan, R.O.C.

Introduction

Inverted papilloma is an uncommon but well-described benign neoplasm involving the mucosal lining of the nasal cavity and paranasal sinuses.¹ The sphenoid sinus is rarely reported as the site of origin.¹–³ Its pathophysiology is characterized by its capacity to be locally destructive and exhibit an endophytic growth pattern with inversion of the surface epithelium into the underlying stroma. Although benign in nature, an inverted papilloma displays peculiar biological behavior, such as its tendency to recur and its association with malignancy. Therefore, the management of this disease is difficult. Since the recurrence rate after an incomplete tumor resection is high,² en bloc tumor extirpation used to be achieved by medial maxillectomy, midfacial degloving approach or through lateral rhinotomy.⁴ With advances in nasal endoscopic techniques, more and more clinical series have shown that the resection of limited lesions via conservative intranasal approaches does not jeopardize the outcome.⁴–⁶ Here, we report a unique case of inverted papilloma confined to the sphenoid sinus in a 42-year-old woman and illustrate our management with excellent outcome.

Case Report

A 42-year-old healthy female presented with a 6-month history of right-side nasal fullness and postnasal drip. Her past medical history was unremarkable. Physical examination and nasal endoscopy revealed an enlarged sphenoid sinus ostium and an irregular, friable, cauliflower-like soft tissue mass occupying the right sphenoid sinus (Figure 1). Ophthalmologic examination was unremarkable. Computed tomography (CT) of the sinuses revealed a homogeneous mass filling the right sphenoid sinus, with lateral expansion of the sinus wall and ostium dilation (Figure 2). There was no erosion of the sella turcica or adjacent bony wall invasion.

By means of endoscopic transnasal sphenoidotomy, the posteroinferior part of the middle turbinate was removed for better access to the sphenoid sinus and to facilitate postoperative inspection and cleaning of the sphenoid sinus by in-office nasal endoscopy. The natural ostium of the sphenoid sinus was localized between the superior turbinate and posterior nasal septum. The anterior wall of the sphenoid sinus was opened, beginning from the medial side of the superior turbinate to the most lateral, inferior, and superior portions of the anterior wall by mushroom forceps.
and a Kerrison punch. The tumor showed nonpedunculate and simultaneous attachment to multiple walls, mainly from the floor and lateral wall. We meticulously removed it as radically as possible. The mucoperiostium was partially included in the resection. The final pathology report confirmed the diagnosis of inverted papilloma (Figure 3). Currently, the patient has had no recurrence after 24 months of follow-up.

Discussion

Isolated sphenoid sinus disease is relatively uncommon, and an isolated inverted papilloma exclusively originating from the sphenoid sinus is therefore a rare clinical finding. The maxillary antrum and the ethmoid labyrinth are the sinuses most frequently affected by inverted papillomas. The sphenoid sinus may often be involved by extension but is rarely reported as the site of origin of this tumor. According to the literature we have reviewed, only 58 cases, including our patient, have been reported. According to the literature we have reviewed, only 58 cases, including our patient, have been reported.7,8

The diagnosis of isolated sphenoid disease is based on history, physical examination, nasal endoscopy, and radiological studies. The differential diagnosis of a lesion isolated in the sphenoid sinus includes chronic sinusitis with polyposis, mucocele, fungal ball, or fungal sinusitis and other neoplasms such as chordoma or meningioma.10,11 Unilateral nasal obstruction, facial pain or pressure, and nasal discharge are the most common presentations of sinonasal inverted papillomas. Although a variety of less commonly encountered

![Figure 1. Intraoperative endoscopy shows an irregular, friable, cauliflower-like mass arising from the right sphenoid ostium.](image1)

![Figure 2. Computed tomography scan at the level of the sphenoid sinus: (A) coronal view; (B) axial view. A homogeneous mass filled the right sphenoid sinus, with lateral expansion of the sinus wall and ostium dilation (arrow). There was no erosion of the sella turcica or adjacent bony wall invasion.](image2)

![Figure 3. Inverted papilloma with endophytic or “inverted” growth pattern, composed of thickened squamous epithelial nests growing downward into the myxoid stroma (hematoxylin & eosin, 40×).](image3)
symptoms have been reported, including epistaxis, anosmia, occipital/frontal headaches, retro-orbital pain, epiphora, proptosis, diplopia, and even auditory symptoms such as hearing loss and tinnitus. The clinical presentation of inverted papillomas confined to the sphenoid is often nonspecific. By review of relatively large case series studies, a predominance of neurological and visual symptoms in symptomatic patients with sphenoid inverted papillomas are demonstrated. Headache seems to be the most common presenting symptom but was absent in our case. As for imaging findings, inflammatory etiologies, which bear the greatest responsibility for isolated sphenoid sinus lesions, often present with a spectrum of pathology ranging from mucosal thickening to complete opacification. Evidence of adjacent bony erosion secondary to bony remodeling of the sinus walls because of pressure atrophy is useful in diagnosing sinonasal inverted papilloma; however, this manifestation was absent in our case (Figure 2). Therefore, the diagnosis in our patient depended mainly on endoscopic features and pathological findings.

Grossly, inverted papillomas tend to be firm, opaque, polypoid, quite bulky, and more vascular than inflammatory polyps. Histopathologically, inverted papillomas exhibit a hypercellular epithelial surface that invades the underlying stroma instead of proliferating outward. The basement membrane remains intact and distinct from the underlying stroma. There is an obvious lack of eosinophilia and edematous mixoid stroma, further distinguishing inverted papillomas from ordinary inflammatory nasal polyps. Some of these features were seen on pathological examination of our patient’s specimen and are depicted in Figure 3.

Proper management is crucial because of the erosive behavior of inverted papilloma, and the small but non-negligible possibility of malignancy and well-documented tendency to postoperative recurrence. Surgical approaches to access the lesion in the sphenoid sinus include various transcranial, transfacial, and transnasal approaches. Endoscopic surgery has gained great popularity in management of paranasal sinus disease, although it does have limits in patients with a tumor that requires more extensive or radical excision. For this purpose, some reports advocate radical excision utilizing endoscopic techniques in conjunction with open approaches for complete tumor extirpation. However, if the sphenoid sinus is the only sinus requiring investigation, we believe that endoscopic sinus surgery alone, which allows a more anatomically directed approach to the sphenoid sinus, can offer equally good results in selected patients. Surgical violation of other sinuses or the nasal septum can be avoided. In a previous report, bilateral sphenoidectomy was performed after anterior and posterior ethmoidectomy. The widening of the anterior wall of the sphenoid sinus is achieved over the lateral side of the superior turbinate, and the lower half to two-thirds of the superior turbinate is cut. In our patient, since the lesion was confined to the unilateral sphenoid sinus, we preserved the unaffected sphenoid sinus mucosa. Unilateral sphenoidotomy was performed over the medial side of the superior turbinate without ethmoidectomy or superior turbinate excision. We did cut the posteroinferior part of the middle turbinate for better sphenoid sinus access and facilitation of postoperative in-office nasal endoscopic follow-up. Because the sphenoid sinus is closely related to several vulnerable neural and vascular structures, complete tumor extirpation including the surrounding bony wall was not possible. Although we took the most radical approach possible, a longer follow-up than normal is necessary to monitor recurrences.

References