CASE REPORT

Pituitary apoplexy associated with an inverted papilloma of sphenoid sinus

Sang-Jek Liew a, Jung-Tung Liu a, Chao-Yu Shen b, Wei-Chean Tan a,*

a Department of Neurosurgery, Chung Shan Medical University Hospital, Taichung 402, Taiwan
b Department of Medical Imaging, Chung Shan Medical University Hospital and School of Medicine, Chung Shan Medical University, Taichung 402, Taiwan

Received 29 July 2011; received in revised form 12 September 2011; accepted 11 December 2011
Available online 30 October 2012

KEYWORDS
inverted papilloma; pituitary apoplexy; sphenoid sinus

Summary A 42-year-old man had a severe progressive headache and visual field defects due to pituitary apoplexy. Transsphenoidal resection of a pituitary tumor (measuring approximately 8.7 cm³) resulted in the complete relief of his symptoms. Histological examination revealed the presence of a pituitary adenoma and an inverted papilloma of the sphenoid sinus. This communication emphasizes the following: (1) mucosal thickening and an inverted papilloma of the sphenoid sinus can be a precipitating factor in pituitary apoplexy; (2) an inverted papilloma of the sphenoid sinus may increase intrasellar pressure and therefore increase the risk of acute pituitary apoplexy; (3) if imaging shows thickening of the sphenoid sinus mucosa, then a partial resection of the sphenoid sinus mucosa for histological evaluation should be performed during the transsphenoidal resection of the pituitary tumor.

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1. Introduction

In 1898, the neurologist Pearce Bailey first described pituitary apoplexy that was caused by a hemorrhage into a pituitary adenoma. 1 In 1905, Bleibtreu 2 reported a similar case. The disorder remained obscure as a clinical entity until 1950 when Brougham et al 3 described it as a clinicopathological syndrome in five patients postmortem. Arita et al 4 recently described thickening of the sphenoid sinus mucosa as a radiological finding during the acute stage of pituitary apoplexy. Liu and Couldwell 5 observed this thickening on MRI scans of patients with pituitary apoplexy. Since this type of thickening has never been pathologically evaluated, it cannot be determined whether the thickening is the result of swelling only, the result of a neoplastic
lesion, or is directly associated with the mucosal thickening of the sphenoid sinus. In the present report, we describe a case of pituitary macroadenoma causing pituitary apoplexy that was associated with an inverted papilloma of the sphenoid sinus. To the best of our knowledge, this is the first case reported in the medical literature.

2. Case report

2.1. History and examination

A 42-year-old man presented with a 2-year history of hypothyroidism. He had a severe progressive headache, dizziness, nausea, and vomiting for 3 days. His neurological examination was unremarkable, except for visual field defects. A computed tomography (CT) scan revealed a pituitary tumor with thinning of the sellar floor (Fig. 1A). Magnetic resonance imaging (MRI) revealed a homogeneously enhanced pituitary mass (2.5 cm in length and 8.7 cm³ in volume) that was compressing the optic chiasm and sellar content, and was bleeding into the pituitary mass (Fig. 1B). Also present were thickening of the sphenoid sinus mucosa (approximately 1.3 mm) (Fig. 1C) and a lesion of the sphenoid sinus with focal contrast-enhancement (6%, 2.3 mm²) (Fig. 1C and D). Clinical evaluation and the presence of the lesion suggested apoplexy due to a nonfunctioning pituitary macroadenoma.

Figure 1  (A) A brain computed tomography image shows pituitary tumor apoplexy and thinning (arrow) of the sellar floor. (B) The postcontrast sagittal T1-weighted magnetic resonance imaging (MRI) scan shows a huge cystic lesion with fluid–fluid level (arrow) in the suprasellar region. The lesion is compressing the optic chiasm (arrowheads). (C) The postcontrast coronal T1-weighted MRI image shows thickening (greater than 1 mm) and enhancement (arrowheads) of the roof of sphenoid sinus mucosa. The focal sphenoid sinus mucosa lesion (arrow) is enhanced. (D) Postcontrast sagittal T1-weighted image shows a focal contrast-enhanced lesion (6%, 2.3 mm²) (arrow) of the sphenoid sinus mucosa.
2.2. Treatment

The patient underwent a transsphenoidal resection of the tumor. Before opening the sellar floor, the red, swollen sphenoid sinus mucosa was noted to have an irregular margin (Fig. 2). It was partially resected for histological evaluation. The sellar floor was thin and therefore easy to drill. A window cut through the sellar floor revealed an intrasellar tumor and hematoma. Glucocorticoids were administered before and after the operation.

2.3. Histological examination

Histological examination of the pituitary adenoma revealed sheets or cords of uniform polygonal cells and little connective tissue (Fig. 3A). Histological examination of the sphenoid sinus mucosa revealed a small area of an inverted papilloma (Fig. 3B).

3. Discussion

The incidence of pituitary apoplexy in published series varies between 0.6% and 10%; it is usually less than 5%, with a mean of 2% for all surgically resected adenomas. In 60–80% of cases, pituitary apoplexy occurs spontaneously in previously asymptomatic patients and occurs in patients who have never been diagnosed as having an adenoma. Approximately 25–30% of patients presenting with pituitary apoplexy have precipitating or predisposing factors such as closed head trauma; hypotension (regardless of the cause); hypertension; history of irradiation; cardiac surgery; anticoagulant therapy; dopamine agonist treatment; and dynamic testing of pituitary function.

An inverted papilloma is a primary benign epithelial tumor of the nose and paranasal sinus. It is uncommon in the general population; however, inverted papillomas account for 0.5–5% of primary nasal tumors. An inverted papilloma of the sphenoid sinus occurring in correlation with precipitating factors or as a cause of pituitary tumor apoplexy has never been reported in the literature.

The term “inverted papilloma” refers to its histological appearance as an endophytic (i.e., downgrowth) of the epithelium into the stroma below the basement membrane boundary that separates the epithelial component from the underlying connective tissue stroma. An inverted papilloma has three characteristics: (1) a tendency to recur; (2) the ability to destroy or remodel bone; and (3) a tendency to be associated with malignancy. Pressure due to the inverted papilloma growth can cause thinning or destruction of the bony nasal wall and can cause decalcification or deformation that displaces neighboring structures. Osteolysis may signal the presence of malignancy. We have found it remarkable in our present case that an inverted papilloma of the sphenoid sinus may be a causal factor in thinning the sella turcica and may increase intrasellar and venous flow pressure, thereby causing pituitary apoplexy.

Mucosal thickening and an inverted papilloma of the sphenoid sinus may be a precipitating factor of pituitary tumor apoplexy. As Arita et al proposed, thickening of the sphenoid sinus mucosa may also be caused by a sudden increase in the intrasellar pressure, leading to obstruction of the out-to-in transsellar venous flow and consequently...
venous congestion. Increased intrasellar pressure caused by pituitary vessel hypotension and ischemia has also been implicated in acute pituitary apoplexy.\textsuperscript{7} Zayour et al\textsuperscript{10} found an association of pituitary apoplexy with a rapid increase in intrasellar contents. The impact of this rapid increase on intrasellar pressure is unknown. Zayour et al\textsuperscript{10} also reported that patients with pituitary apoplexy had large macroadenomas (mean size, 2.3 $\pm$ 0.4 cm; range, 1.5 to 3.5 cm; median, 2.5 cm) and a remarkably high intrasellar pressure (mean pressure, 44.1 $\pm$ 11.1 mmHg; range 25–58 mmHg; median, 47 mmHg). In our patient, the cause of sphenoid sinus mucosal thickening was an inverted papilloma of the sphenoid. Therefore, we suspected that the inverted papilloma increased the intrasellar pressure and increased the risk of acute pituitary apoplexy.

Other important characteristics of inverted papillomas are that they are associated with squamous cell carcinomas; they have a variable incidence of malignant transformation (5–21%); and they tend to have a local recurrence.\textsuperscript{9} All mucosa with an inverted papilloma should be completely removed by the endoscopic endonasal route.\textsuperscript{9} Thus, we conclude that sphenoid sinus mucosal thickening and a sphenoid sinus inverted papilloma could be a precipitating factor of pituitary apoplexy. Inverted papilloma of the sphenoid sinus may increase the intrasellar pressure and therefore increase the risk of acute pituitary apoplexy. During transsphenoidal resection of a pituitary tumor, partial resection of the sphenoid sinus mucosa for histological evaluation is recommended if imaging shows thickening of the sphenoid sinus mucosa.

References