CASE REPORT

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Oncocytic schneiderian papilloma confined to the sphenoid sinus detected by FDG-PET*

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SUMMARY

We report a 55-year-old man with oncocytic schneiderian papilloma confined to the sphenoid sinus, which was initially detected by positron emission tomography with 18F-fluorodeoxyglucose (FDG-PET) at a very early stage. Based on CT and MRI findings, we suspected that the tumor was most likely benign; however, positive uptake in FDG-PET suggested malignancy. The patient underwent endoscopic resection of the tumor, and the histopathological diagnosis turned out to be oncocytic schneiderian papilloma. FDG-PET is thought to be a powerful tool to search for malignant lesions, but the present case demonstrates the fallibility of this technique. This should be taken into consideration when interpreting FDG-PET images.

Key words: sphenoid sinus, positron emission tomography, 18F-fluorodeoxyglucose, oncocytic schneiderian papilloma, oncocytoma

INTRODUCTION

Isolated sphenoid sinus disease is a relatively uncommon entity. Patients with sphenoid sinus diseases may manifest various clinical symptoms and signs including headache, ocular symptoms, cranial nerve palsies, and nasal symptoms. However, small lesions confined to the sphenoid sinus are generally asymptomatic, making it difficult to detect the disease at an early stage.

Positron emission tomography with 18F-fluorodeoxyglucose (FDG-PET) is a recently established imaging device that exhibits powerful potential in the diagnosis of tumorous lesions. We herein report a case of oncocytic schneiderian papilloma that was detected by FDG-PET at a very early stage, and review the clinical and radiological characteristics of this disease. To our knowledge, this clinical report describes the first case of asymptomatic benign tumor of the sphenoid sinus detected by FDG-PET.

CASE REPORT

A 55-year-old man underwent a total cystectomy combined with lymph node dissection for a bladder carcinoma in December 2003. His postoperative clinical course had been uneventful; however, upon a periodic checkup in January 2005, positive FDG uptake (mean standardized uptake value=11.26, maximum standardized uptake value=18.86) around the sphenoid sinus was revealed in an FDG-PET image (Figure 1). The patient was then referred to our department for further examination. When he was first seen by us on February 4, 2005, otorhinolaryngological findings were normal, and no cervical lymph nodes were palpable. He did not complain of any related symptoms such as headache, visual disturbance, or nasal symptoms. His chest X-ray and laboratory data were unremarkable. CT and MRI revealed a 15 x 10-mm well-defined soft tissue mass in the left sphenoid sinus without bone destruction (Figures 2 and 3). The mass was well enhanced

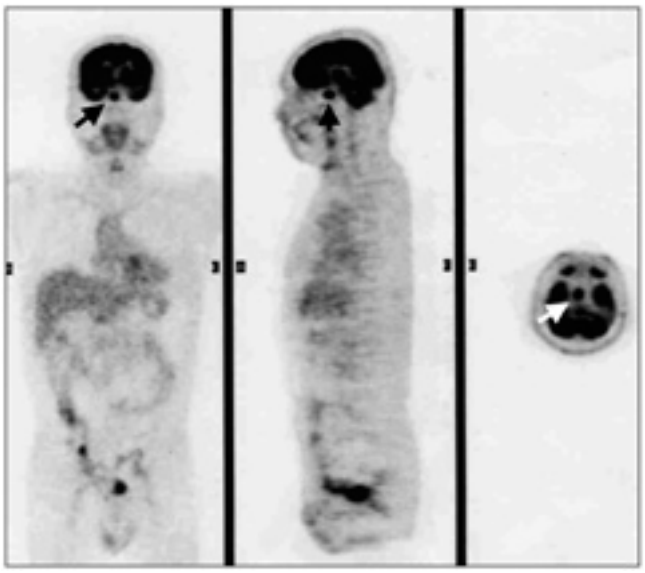

Figure 1. FDG-PET images showing increased FDG uptake (arrows) around the sphenoid sinus.

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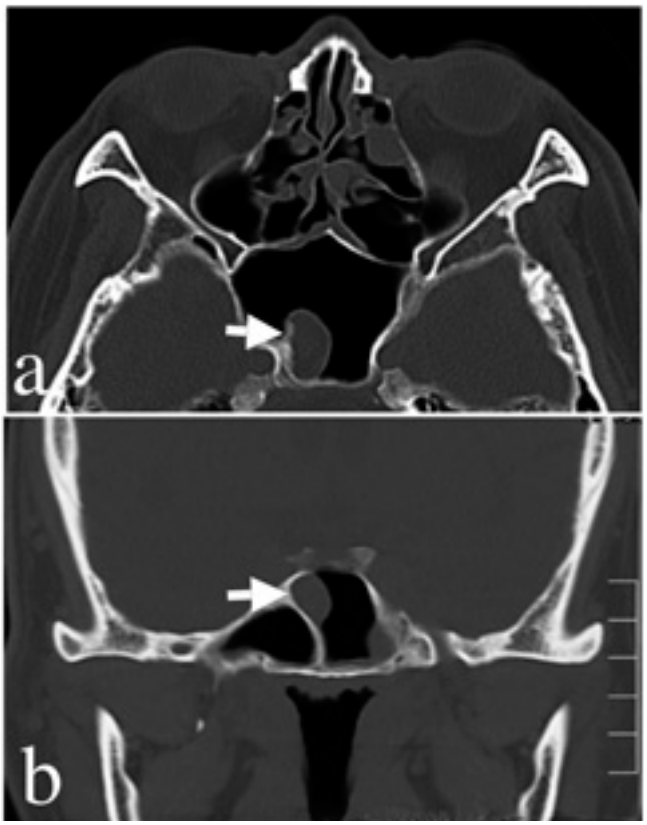

Figure 2. CT scans. (a) Axial scan shows a soft tissue mass of the sphenoid sinus (arrow). (b) Coronal scan reveals a sphenoid sinus mass (arrow).

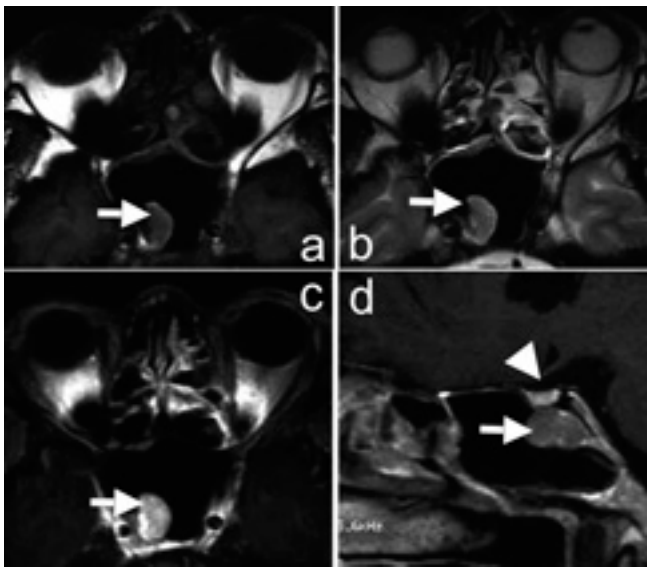

Figure 3. MR images. A-C, Axial T1-weighted (a), T2-weighted (b), and enhanced (c) MR images depict a well-defined and well-enhanced soft tissue mass in the left sphenoid sinus (arrows) (d) Right sagital enhanced MR image shows a sphenoid sinus mass (arrow) and the adjacent pituitary gland (arrowhead).

and showed hypointensity and heterogeneous isointensity in the T1- and T2-weighted MRI, respectively. Based on these CT and MRI findings, we suspected that the tumor was most likely benign; however, the high FDG uptake at the same site shown in FDG-PET suggested the possibility of malignancy. Endoscopic exploration of the sphenoid sinus via the left ethmoid sinus was then performed on November 21. The nasal cavity was normal, and the mucosa of the ethmoid sinus was slightly edematous. In the left sphenoid sinus, there was a little-finger-sized polypoid and hypervascular tumor arising from the posterior wall (Figure 4a). Bone defect was not observed. The tumor was carefully detached and completely removed from the posterior wall of the sphenoid sinus.

Microscopically, the resected tumor was revealed to be composed of multilayered oncocytic columnar or transitional epithelium with an exophytic or inverted growth often forming microglandular or microcystic structures supported by a relatively delicate fibrovascular stroma (Figure 4b). The tumor was histopathologically diagnosed as an oncocytic schneiderian papilloma.

The patient's postoperative clinical course was uneventful. He was discharged on the 5th postoperative day and is currently free from disease 10 months after surgery.

DISCUSSION

The sphenoid sinus is located right under the skull base and adjoins a variety of vital anatomical structures such as the dura mater, pituitary gland, cavernous sinus, pterygoid canal and nerve, internal carotid artery, and cranial nerves II, III, IV, V, and VI. Diseases of the sphenoid sinus may damage these structures, leading to consequent symptoms: headache is the most common presenting symptom in patients with isolated sphenoid sinus diseases, while visual disturbance, cranial nerve deficits, and epistaxis are next most common [1,2]. On the other hand, small lesions confined to the sphenoid sinus, as in the present patient, are generally asymptomatic, hardly visible on paranasal sinus x-rays or even by using nasopharyngeal fiberscopy, and therefore, rarely detected at an early stage. However, because of the recent advance of imaging modalities, there have been increasing cases of such asymptomatic lesions that happened to be discovered by physicians other than otolaryngologists.

Inflammatory diseases are most common in the sphenoid sinus, whereas benign and malignant tumors are relatively minor [1,2]. Lawson et al., in their retrospective analysis, found 10 benign and 18 malignant tumors out of 132 cases of isolated sphenoid sinus diseases [1]. In another report of 122 cases of isolated sphenoid sinus diseases, 8 benign and 8 malignant tumors were included [2]. According to these reports, inverted papilloma is the most common benign tumor of the sphenoid sinus [1,2]. Other benign tumors arising in the sphenoid sinus such as fibro-osteoma, schwannoma, plasmacytoma, hemangioma, pseudotumor, myxofibroma, and osteochondroma have

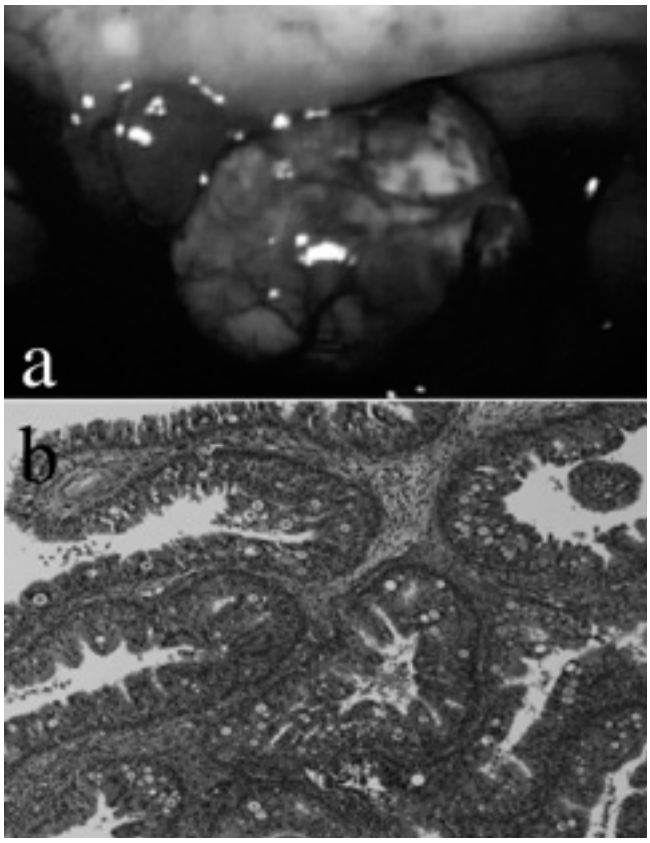

Figure 4. Perioperative endoscopic view (a) and photomicrograph of the tumor (b). (a) Endoscopic view (30°) via a left transethmoidal route shows a polypoid and hypervascular mass. (b) The tumor is composed of multilayered oncocytic columnar or transitional epithelium with an exophytic or inverted growth, often forming microglandular or microcystic structures supported by a relatively delicate fibrovascular stroma. (H&E staining; original magnification x 100).

sporadically been reported [1,2]. Although oncocytic schneiderian papilloma involving both the sphenoid and ethmoid sinuses has been described previously [3], the present patient is the first case that was diagnosed while the tumor was still confined to the sphenoid sinus.

The schneiderian membrane, the mucous membrane that lines the nasal and paranasal sinuses, gives rise to three morphologically distinct variants of sinonasal papillomas [4]: fungiform papilloma (exophytic papilloma), inverted papilloma (inverting papilloma), and oncocytic schneiderian papilloma (cylindrical cell papilloma or columnar cell papilloma). Oncocytic schneiderian papilloma is the rarest of the three variants, comprising 3.2-5.0% of all sinonasal papillomas, and occurs predominantly in the maxillary sinus of elderly people [4,5]. In contrast to the male predominancy of fungiform and inverted papillomas, this tumor equally affects both sexes [4]. Histologically, oncocytic schneiderian papilloma shows multilayered eosinophilic columnar epithelium with small mucous-containing cystic structures, and it exhibits both exophytic and inverted growth patterns [4]. This tumor is often confused with well-differentiated adenocarcinoma, but the latter contains only monolayered epithelium [5]. Histopathological findings of the present case were compatible with those of oncocytic schneiderian papilloma.

Clinically, it is well recognized that sinonasal papillomas, particularly inverted papillomas, often show repetitive local recurrence and have a risk of malignant transformation [3,4]. Oncocytic schneiderian papillomas show equally aggressive biological behavior to inverted papillomas: The local recurrence rates of oncocytic schneiderian papilloma and inverted papilloma are 33-40% and 15-46%, respectively [3-6]. The incidences of malignant transformation of the two tumors are 4-17% and 9-13%, respectively [3-6]. The most common form of malignancy arising in oncocytic schneiderian papillomas is squamous cell carcinoma [4,6]. Patients with this tumor should, therefore, be under careful and long-term follow-up.

The performance of FDG-PET in the differential diagnosis of benign from malignant lesions has been elucidated to a certain extent. A number of authors have shown significant differences in FDG uptake between benign and malignant lesions. Ninomiya et al. [7] analyzed FDG-PET images of patients with sinonasal diseases prior to surgical treatment and found that the standardized uptake value, the ratio of radioactivity in the region of interest to average radioactivity throughout the body, of squamous cell carcinomas (ranging 10.40-19.36 with a median of 12.94) was significantly higher than that of inverted papillomas (ranging 2.3-5.2 with a median of 3.49). However, their report also documented that some other malignancies showed relatively low uptake values [7]. Although FDG uptake of benign tumors and inflammatory lesions is generally considered to be low, there are occasional exceptions. Strauss described that FDG accumulates in some non-specific inflammatory tissue as in malignancies [8]. It was also reported that oncocytic tumors such as oncocytma, Warthin's tumor, Hurthle cell adenoma, and oncocytic papillary neoplasms exhibit high FDG uptake even though they are benign [9-12]. In the present case, the sphenoid lesion had shown high FDG uptake, implying malignancy, but the surgical specimen was histopathologically diagnosed as a benign tumor, an oncocytic schneiderian papilloma.

The reason why oncocytic tumors show high FDG uptake is unclear. It has been known that the uptake pattern of FDG resembles that of Tc-99m pertechnetate, which also accumulates in oncocytic tumors [13,14]. Abundant mitochondria present in oncocytic tumor cells may suggest their vigorous energy metabolism, and this may explain their anomalous uptake of the tracers [9]. Although FDG-PET is thought to be a powerful tool to search for malignant lesions, we must be aware that this technique may be fallible in some cases. This should be taken into consideration when interpreting FDG-PET images.

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