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CASE REPORT

Primary Benign Intraosseous Meningioma on ^{18}F-FDG PET/CT Mimicking Malignancy

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Abstract We present a case of primary benign intraosseous meningioma in the sphenoid bone mimicking malignancy. A 44-year-old female patient who had a protruding right eye and headache came to our hospital. MRI showed a large, destructive, heterogeneously well-enhancing soft tissue mass in the right sphenoid bone suggesting malignancy. ^{18}F-FDG PET/CT showed a hypermetabolic mass in the same site with an SUV_{max} of 9.1 The pathological diagnosis by surgery revealed that this tumor was a WHO grade I transitional meningioma. This case suggests that primary benign intraosseous meningioma may show high ^{18}F-FDG uptake mimicking a malignancy.

Keywords Benign meningioma · Sphenoid bone · PET/CT · ^{18}F-Fluorodeoxyglucose

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Introduction

Primary intraosseous meningioma is a very rare benign tumor, mainly originating from the cranial bones, and is also found in the skin, nasopharnyx or neck [1, 2]. This tumor can be clinically and radiologically confused with metastatic cancer and other malignant bone tumors. Only one case report has dealt with ^{18}F-fluorodeoxyglucose (^{18}F-FDG) positron emission tomography/computed tomography (PET/CT) in a primary intraosseous meningioma of the calvarium showing high ^{18}F-FDG uptake [3]. We present a case of primary benign intraosseous meningioma of the sphenoid bone that was confused with malignancy on ^{18}F-FDG PET/CT.

Case Report

A 44-year-old female patient who had a protruding right eye and headache came to our hospital for further evaluation. She had been previously healthy based on the past medical history. Contrast-enhanced MRI showed a large, heterogeneously well-enhancing soft tissue mass in the right sphenoid bone destroying the right greater and lesser wing of the sphenoid bone, replacing bone marrow and involving the right orbital roof with invasion of the right supraorbital muscle complex and right lateral rectus muscle, which suggested a malignancy (Fig. 1). Subsequently, ^{18}F-FDG PET/CT was carried out for further evaluation. PET/CT was performed using a Discovery STe scanner (GE Healthcare, Milwaukee, WI, USA) 60 min after the intravenous injection of 370 MBq ^{18}F-FDG. PET/CT showed a large hypermetabolic mass involving the right retrobulbar area and right sphenoid bone with a maximum standardized uptake value (SUV_{max}) of 9.1. However, no significant abnormal ^{18}F-FDG uptake suggesting a malignancy was found in other sites (Fig. 2). The histophathologic examination by an incisional biopsy of the right orbital mass

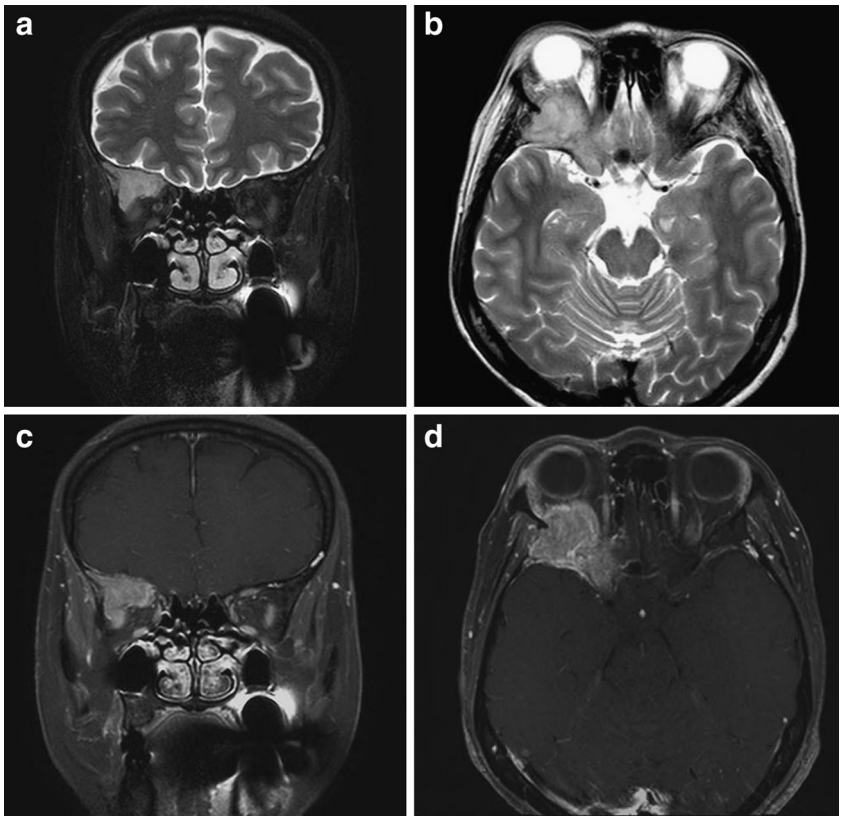

Fig. 1 MRI images of the patient. Axial (**a**) and coronal (**b**) T2-weighted images show a hyperintense lesion in the right sphenoid bone with invasion of the right supraorbital muscle complex and right lateral rectus muscle. Axial (**c**) and coronal (**d**) gadolinium-enhanced T1-weighted images show a large, heterogeneously well-enhancing soft tissue mass in the right sphenoid bone destroying the right greater and lesser wing of the sphenoid bone

showed WHO grade I meningioma. After that, craniotomy with tumor removal was performed. The final diagnosis was primary benign intraosseous WHO grade I transitional meningioma (Fig. 3). After surgery, the patient was clinically followed up for 6 months without evidence of recurrence.

Discussion

Meningioma is the most frequent benign intracranial tumor [4]. Most meningiomas originate from intradural lesions located in the subdural space. However, extradural meningioma does not originate from the dura mater. Primary intraosseous meningioma is a kind of extradural meningioma that arises in bone [5]. The point of origin of extradural meningioma is controversial. Because meninges originate from mesenchymal cells, meningioma could theoretically develop in any site where mesenchymal multipotent precursor cells exist. Some researchers thought that extradural meningioma originated from ectopic arachnoid cap cells or ectopic meningocytes in the cranial sutures during molding at birth. Others suggested that it was related to a trauma history or old skull fracture [6]. However, there was no previous history of trauma in our case.

Extradural meningioma that arises from the skull has been referred to as intraosseous, intradiploic or calvarial meningioma [7]. Primary intraosseous meningioma constitutes about 2 % of all meningiomas [8]. The skull base and convex diploe are two major sites for primary intraosseous meningioma [9, 10]. Primary intrasosseous meningioma on the sphenoid bone corresponds to 15–20 % of all cranial meinigiomas.

Usually primary osseous meningiomas have been shown to be benign and slow growing. However, the possibility of malignant transformation in the primary intrasosseous meningioma is higher than that in the intracranial meningioma [2, 6]. Primary intraosseous meningioma in the sphenoid bone can involve the adjacent orbit and cavernous sinus. Therefore, complete resection of the tumor is always recommended whenever possible [11]. Histopathologically, meningothelial meningioma is a most common subtype of primary intraosseous meningiomas. However, other histologic subtypes include fibroblastic, chordoid, transitional, psammomatous, microcytic and even atypical, malignant meningiomas. WHO grade I meningiomas such as meningothelial, fibroblastic, transitional, psammomatous and microcytic meningiomas are classified as a benign tumors [12, 13]. The histologic subtype in our case was a transitional meningioma.

On simple skull radiographs, primary intraosseous meningioma can show either osteoblastic or osteolytic features. Hyperostosis is another imaging finding of this tumor. Magnetic resonance imaging (MRI) findings of primary intraosseous meningioma are relatively nonspecific.

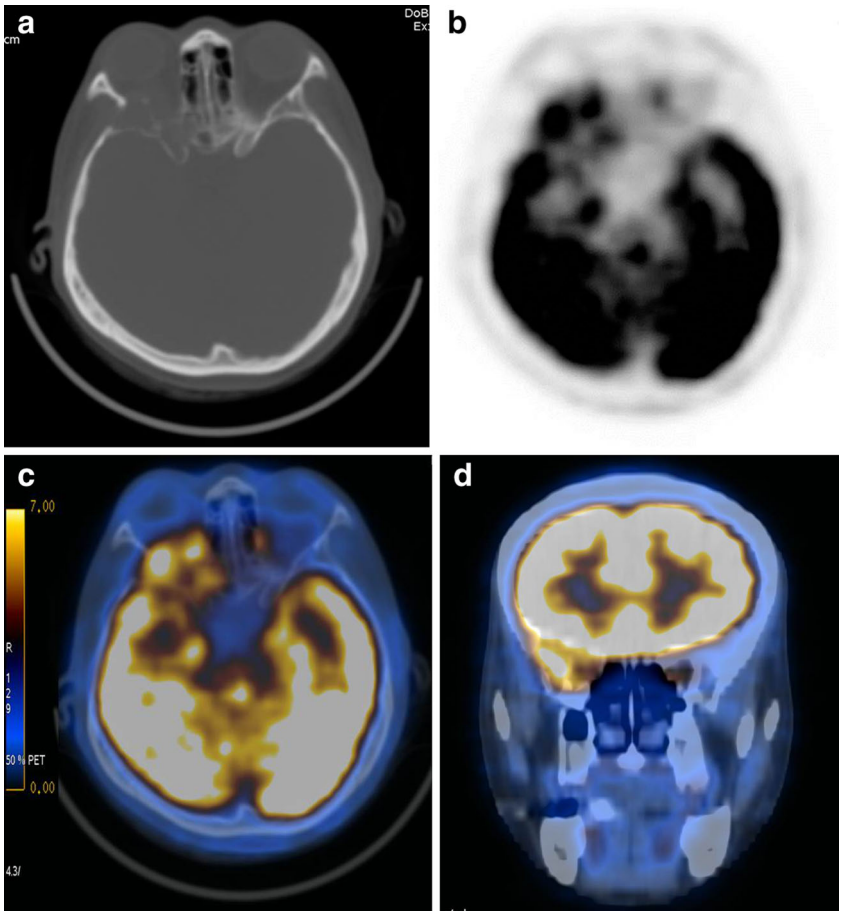

Fig. 2 CT (**a**), transaxial PET (**b**) and fused transverse (**c**) and coronal (**d**) PET/CT images show a hypermetabolic lesion involving the right sphenoid bone and right retrobular area (SUV_{max}=9.1)

T1-weighted images show hypo- or isodense features with prominent enhancement by gadolinium. T2-weighted images usually show hyperintense features [14]. Several previous studies dealt with nuclear medicine imaging in primary osseous meningioma. This tumor had high ^{99m}Tc-diphosphate, ^{99m}Tc-medronate and ^{111}In-pentetreotide uptake. On the contrary, the ^{99m}Tc-pertechnetate scan was negative [15, 16].

Recently ^{18}F-FDG PET/CT has been widely used for tumorous conditions for the differential diagnosis including intracranial meningiomas. In usual dural meniogiomas, WHO grade 2 or 3 meningioma shows higher ^{18}F-FDG uptake than WHO grade 1, which suggests that ^{18}F-FDG uptake in intracranial meningiomas reflects tumor aggressiveness [13, 17]. Until now, only one case of primary intraosseous

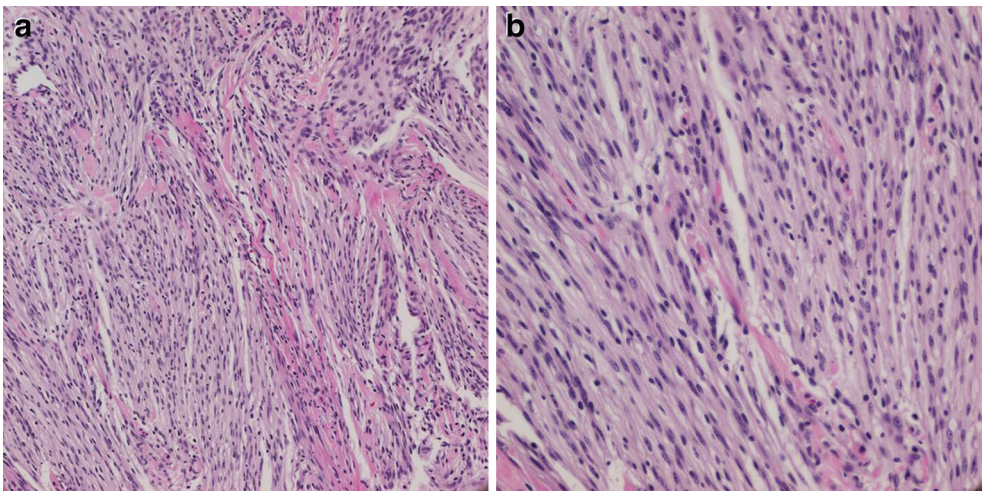

Fig. 3 Pathological specimen after surgery shows that the tumor is composed of a mixture of meningothlieal (**a**) and fibrous (**b**) types of meningioma, which suggests transitional meningioma. **a** Nested aggregate of epithelioid cells, compatible with meningothelial meningioma (H&E, × 200). **b** However, most of the tumor is composed of spindle cells and interspersed collagen bundles, consistent with fibrous meningioma. (H&E, × 400)

meningioma of the convex diploe detected by ^{18}F-FDG PET/CT has been reported [3]. This case showed a hypermetabolic lesion in the right parietal meningothelial meningioma that mimicked a metastasis in a patient with bladder cancer. However, the SUV_{max} of this tumor was not described. Our study was the second reported case of PET/CT findings of primary intraosseous meningioma and first case of primary intraosseous meningioma of the sphenoid bone and transitional type along with a description of the SUV_{max}. In our case, the hypermetabolic lesion involving the right sphenoid bone and right retrobulbar area was bone-destructive and showed high ^{18}F-FDG uptake with a SUV_{max} of 9.1. Therefore, it was considered a malignancy such as a metastatic carcinoma or lymphoma at the time of initial interpretation. However, the final pathological diagnosis was benign WHO grade I transitional meningioma. Thus, our case suggested that it is difficult to differentiate primary intraosseous meningioma from other malignant tumors by ^{18}F-FDG uptake. Although it was not clear why the primary benign intraosseous meningioma had high ^{18}F-FDG uptake, some benign intracranial meningiomas have been reported to have high ^{18}F-FDG uptake [18]. In addition, our case suggested that ^{18}F-FDG uptake might not be associated with tumor grade and aggressiveness in intraosseous meningioma, unlike intracranial meningioma.

In summary, our case demonstrates that primary benign intraosseous meningioma shows high ^{18}F-FDG uptake and bone destruction mimicking a malignancy. Although rare, primary intraosseous meningioma should be considered one of the false positives for the differential diagnosis of bone lesions detected by ^{18}F-FDG PET/CT.

Conflict of Interest Ho Seong Kim, Seok Hwi Kim, Hyung Jin Kim, Se Woong Kang, Soo Jeong Kim, Joo Hee Lee, Sun Pyo Hong, Young Seok Cho and Joon Young Choi declare no conflict of interest.

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