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Case Report

Discrepancy between radiological and histological findings in ganglioglioma of the optic chasm: Case report

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Abstract

Background: Gangliogliomas involving the optic nerve or chiasm are extremely rare tumors, which can be confused radiologically with other neoplasms. α-[*N*-methyl-^{11}C]-methylaminoisobutyric acid (^{11}C-MeAIB) is a new artificial amino acid positron emission tomography (PET) tracer, which is metabolically more stable *in vivo* and may be more specific for tumors than ^{11}C-methionine. However, the utility of ^{11}C-MeAIB PET in the diagnosis of brain tumors has not yet been reported.

Case Description: A 26-year-old man presented with visual field defects and headache, and magnetic resonance imaging demonstrated a suprasellar mass involving the optic chiasm. A biopsy and partial tumor resection were performed via an interhemispheric approach. We diagnosed the tumor as ganglioglioma (WHO grade I) involving the optic chiasm. Although this lesion was histologically benign, ^{11}C-MeAIB PET, 2-deoxy-2-[^{18}F] fluoro-D-glucose (^{18}F-FDG) PET and proton magnetic resonance spectroscopy indicated malignant features.

Conclusion: The discrepancy between radiological and histological findings implies that this new amino acid tracer PET may have a limitation in the diagnosis of gangliogliomas. Although further study is necessary, gangliogliomas should be included in the differential diagnosis of suprasellar tumors, even if PET findings show malignant features.

Key Words: Ganglioglioma, MeAIB PET, optic chiasm

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INTRODUCTION

Gangliogliomas, which are composed of both neuronal and glial components, are rare and account for only 0.3–1.3% of all brain tumors.[11] Gangliogliomas usually occur in the temporal lobe but can arise anywhere in the central nerve system, including the brain stem and spinal cord. However, gangliogliomas involving the optic nerve, chiasm, or tract are extremely rare, and it is difficult to diagnose these lesions radiologically.[6,11,13] To date, gangliogliomas of the optic chiasm, which are evaluated by positron emission tomography (PET) or proton magnetic resonance spectroscopy (MRS), have not been reported. α-[*N*-methyl-^{11}C]-methylaminoisobutyric acid (^{11}C-MeAIB) is a new artificial amino acid PET tracer, which is

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metabolically more stable *in vivo* and may be more specific for tumors than ^{11}C-methionine (^{11}C-MET). However, the utility of ^{11}C-MeAIB PET in the diagnosis of brain tumors has not yet been reported. Here, we report the first case of ganglioglioma of the optic chiasm, which was evaluated by ^{11}C-MeAIB PET, 2-deoxy-2-[^{18}F] fluoro-D-glucose (^{18}F-FDG) PET and MRS. We also show discrepancies between radiological and histological findings in this lesion.

CASE DESCRIPTION

A 26-year-old man presented with a 1-month history of progressive visual field defects and headache. Neuro-ophthalmological examination revealed temporal hemianopsia and bilateral papilledema. As part of the diagnostic work-up at our institution, computed tomography (CT), magnetic resonance imaging (MRI), MRS (Ingenia 3.0T, Philips Medical System, Amsterdam, Nederland), and PET (GE Advance, GE Healthcare, Waukesha WI, USA) were performed. CT revealed a mixed-dense suprasellar mass lesion without signs of calcification, as well as hydrocephalus caused by obstruction of the foramina of Monro. MRI demonstrated a suprasellar mass involving the optic chiasm, which had cystic components. The lesion was hypointense on T1-weighted images (WI), hyperintense on T2-WI, and showed marked enhancement after the administration of contrast material [Figure 1].

^{11}C-MeAIB PET revealed remarkably high uptake of MeAIB]standardized uptake value (SUV) max = 9.8] consistent with enhanced lesion on MRI, and lesion to gray matter ratio (LGR) was 56.9 [Figure 2]. ^{18}F-FDG PE*t al*so showed high uptake of FDG (SUVmax = 9.97 and LGR = 1.6) [Figure 2]. MRS demonstrated that choline/creatine (Cho/Cr) peak-height and Cho/Cr peak-area values were 2.85 and 2.33, respectively (TR/TE = 2000/32) [Figure 3]. Cho/N-acetylaspartate (NAA) peak-height and Cho/NAA peak-area values were 1.32 and 0.93, respectively. ^{11}C-MeAIB PET, ^{18}F-FDG PET, and MRS data suggested a malignant behavior.

A biopsy and partial tumor resection were performed via an interhemispheric approach. The intraoperative findings were that the tumor originated from the optic chiasm without a clear plane of dissection. Therefore, total resection was abandoned to avoid damage to the optic nerve. Histopathological evaluation of the tumor revealed that the tumor comprised both neoplastic glial and ganglion cells. Immunohistochemical staining demonstrated that dysmorphic ganglion cells were positive for synaptophysin, and pleomorphic glial cells were positive for glial fibrillary acid protein (GFAP). No high-grade neoplastic feature was detected, and Ki-67 immunoreactivity was less than 3%. We diagnosed the tumor as ganglioglioma (WHO grade I) involving the

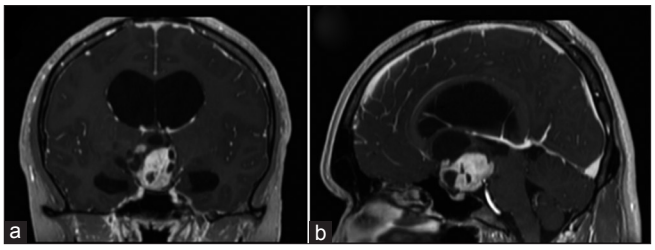

Figure 1: Contrast-enhanced T1-weighted MR images, coronal (a) and sagittal (b) views, show a suprasellar mass lesion with cystic components

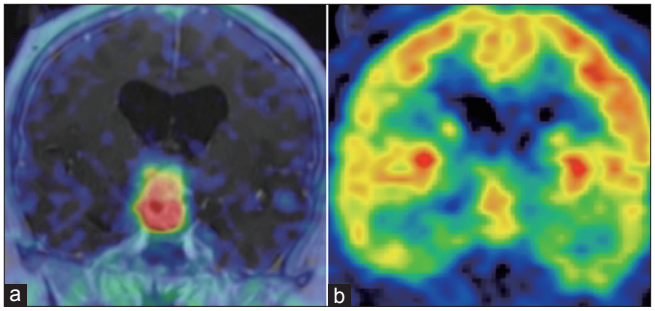

Figure 2: Both ^{11}C-MeAIB PET (a) and ^{18}F-FDG PET (b) show high accumulation in the suprasellar lesion

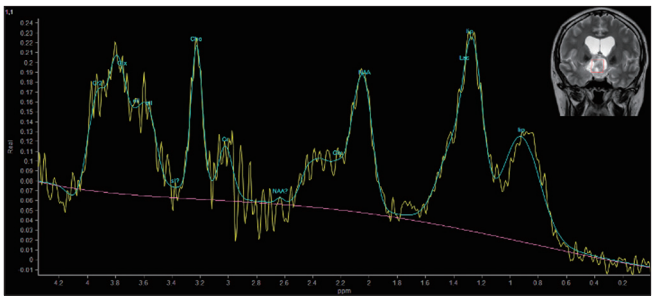

Figure 3: MRS demonstrates elevated Cho peak in the tumor (TR/TE = 2000/32)

optic chiasm. Although ^{11}C-MeAIB PET, ^{18}F-FDG PET, and MRS indicated malignant features, the lesion was histologically benign. Because ventricular dilatation did not improve, a ventriculoperitoneal shunt was performed. Postoperative course was uneventful. Although the role of postoperative radiotherapy is controversial, most cases of optic gangliogliomas reported in the literature have received adjuvant radiation when only biopsy or subtotal resection could be performed.[11,13] We performed fractionated stereotactic radiotherapy (54 Gy in 30 fractions).

DISCUSSION

Gangliogliomas of the optic chiasm are extremely rare and radiologically difficult to distinguish from other suprasellar tumors.[6,11,13,14] ^{18}F-FDG PET has been used for glioma grading and the differentiation of recurrent lesions from radiation necrosis.[12] However,

to date, ^{18}F-FDG PET features of gangliogliomas of the optic chiasm have not been reported, while cerebral gangliogliomas scanned with ^{18}F-FDG PET usually show hypometabolism. In 8 of 9 cerebral low-grade gangliogliomas, ^{18}F-FDG PET showed hypometabolism, and the other showed isometabolism.[4] In 10 cerebral low-grade gangliogliomas scanned with ^{18}F-FDG PET, 8 cases showed hypometabolism, and one case showed isometabolism.[3] However, one case demonstrated hypermetabolic activity.[3] In 11 cerebral low-grade gangliogliomas, only one patient showed a moderately hypermetabolic lesion.[10] Löbel *et al.* have reported a case of cerebellar low-grade ganglioglioma which showed striking hypermetabolism on ^{18}F-FDG PET.[7] Mayer *et al.* have also reported a case of low-grade cerebral ganglioglioma, which showed hypermetabolism on ^{18}F-FDG PET.[8] In the present case, ^{18}F-FDG PET showed a markedly hypermetabolic suprasellar lesion, while the histopathological examination revealed a low-grade ganglioglioma (WHO grade I). Hypermetabolism shown by ^{18}F-FDG PET may reflect the larger neuronal cell population of gangliogliomas.

It is well known that ^{11}C-MET PET is more sensitive for brain tumors than ^{18}F-FDG-PET. Im *et al.* have reported a case of cerebral ganglioglioma, which showed hypometabolism on ^{11}C-MET PET.[3] Conversely, Phi *et al.* have reported that 3 of 6 cases of low-grade cerebral gangliogliomas showed markedly hypermetabolic lesions.[10] The mean LGR of ^{11}C-MET uptake was 2.114 ± 0.723. In contrast, ^{11}C-MeAIB is a new artificial amino acid PET tracer, which is metabolically more stable *in vivo* than ^{11}C-MET.[9,15] It has been shown that ^{11}C-MeAIB is a useful tracer for measurement of amino acid uptake in the diagnosis of lung malignancies, malignant lymphomas, and head and neck cancers.[9,15] However, the utility of ^{11}C-MeAIB PET in the diagnosis of brain tumors has not yet been reported. To our knowledge, the present report shows the first case of brain tumor, which was examined by ^{11}C-MeAIB PET. In the present case, ^{11}C-MeAIB PET showed high accumulation in the lesion, although it was a low-grade ganglioglioma (WHO grade I). The LGR of ^{11}C-MeAIB uptake was extremely high (39.01). These findings suggest that ^{11}C-MeAIB PET may reflect metabolic features other than proliferative activities of gangliogliomas. This discrepancy between radiological and histological findings implies that this new amino acid tracer PET may have a limitation in the diagnosis of gangliogliomas.

MRS provides useful information on tumor metabolism and has been proposed as a method for tumor grading.[1,2] However, to date, MRS findings of gangliogliomas of the optic chiasm have not been reported, whereas several cases of cerebral gangliogliomas evaluated by MRS have been reported. Im *et al.* have reported 9 cerebral or cerebellar low-grade gangliogliomas evaluated by MRS, which showed increased Cho/NAA ratios.[3] Löbel *et al.* have also reported a case of cerebellar low-grade gangliogliomas showing increased Cho/NAA ratio within the tumor.[7] Kumabe *et al.* have reported 2 cases of cerebral low-grade gangliogliomas evaluated by MRS, which showed increased Cho values.[5] These findings are suggestive of high-grade tumors, although all cases were histologically benign. In the present case, Cho/NAA ratio was increased, while it was a low-grade ganglioglioma (WHO grade I). This discrepancy is consistent with previous findings reported in cerebral low-grade gangliogliomas. This increased Cho/NAA ratio may indicate high glial and myelin content of gangliogliomas.

CONCLUSIONS

We have shown the first case of ganglioglioma of the optic chiasm, which was evaluated by ^{11}C-MeAIB PET, ^{18}F-FDG PET, and MRS. Although this lesion was histologically benign, ^{11}C-MeAIB PET, ^{18}F-FDG PET, and MRS indicated malignant features. Since this report shows only one case, further study is necessary to confirm the advantages and limitations of these diagnostic modalities. Gangliogliomas should be included in the differential diagnosis of suprasellar tumors, even if PET and MRS findings show malignant features.

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Disclosure
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Conflicts of interest
There are no conflicts of interest.

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