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Case Report

Isolated intracranial osteoblastic meningioma

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Abstract: Osteoblastic meningioma is a subtypes of meningioma. Intracranial isolated osteoblastic meningioma is extremely unusual. We reported a case of osteoblastic meningioma in the right frontal region in a 15-year-old girl who presented with repeated unconsciousness for 20 days. Computed tomography (CT) revealed a well-defined calcified mass with CT value of 802.5 HU in the right frontal region next to the midline. Magnetic Resonance Imaging (MRI) showed a 4×4×3 cm mass, appearing hypointense on both T1 and T2-weighted imaging and heterogeneous enhancement on contrast-enhanced imaging. A right frontal craniotomy via coronal approach was performed and a bony mass was found in the inferior frontal lobe without attachment of cerebral falx. Pathological examination of the neoplasm was osteoblastic meningioma. Due to its rarity, osteoblastic meningioma was usually misdiagnosed preoperatively. After reviewing related literature, we made differential diagnosis between central nervous system tumors with ossification or calcification and announced that intracranial well-defined calcified mass, even without attachment of convexities or Sylvian fissure, should also be considered as osteoblastic meningioma before operation.

Keywords: Osteoblastic meningioma, intracranial, diagnosis

Introduction

Osteoblastic meningiomas is a subtype of meningioma consisting of mesenchymal differentiation, accounting for approximately 1% of all meningiomas [1, 2]. These lesions are usually located over the cerebral convexities or Sylvian fissure [1], and occasionally involved in spine [3, 4], ventricles and skull [5-7]. We reported a case of isolated osteoblastic meningioma in the right frontal region. To our knowledge, isolated intracranial osteoblastic meningioma in frontal region has not been reported previously.

Case presentation

A 15-year-old girl was admitted to our hospital because of repeated unconsciousness for 20 days. Physical examination did not find any abnormality. CT revealed a well-defined calcified mass with CT value of 802.5 HU in the right frontal region next to the midline (**Figure 1A** and **1B**). MRI showed a 4×4×3 cm mass, appearing hypointense on both T1 and T2-weighted imaging and heterogeneous enhancement on contrast-enhanced imaging, anterior inferior of which was a cystic lesion (**Figure 1C-F**). The differential diagnosis at that time included intracerebral chondroma, glioma and atypical meningioma.

A right frontal craniotomy via coronal approach was performed and a bony mass was found, supplied by surrounding brain parenchyma vascular, but without attachment of cerebral falx. The mass was totally removed and the excited tumor was subjected to pathologic examination. Histological examination revealed a bony structure, with local proliferation of fibrous tissue, between which foci epithelium aggregated. Immunohistochemical staining demonstrated that tumor cell was positive for Vimentin, EMA and Ki-67 (<2%), suspicious for S-100, but negative for GFAP and CD163 (**Figure 2A-D**). The histological diagnosis was osteoblastic meningioma.

The postoperative course was uneventful, and the patient was discharged on the 10th postoperative day. At her 1-year follow up visit, the patient was completely free of symptoms and

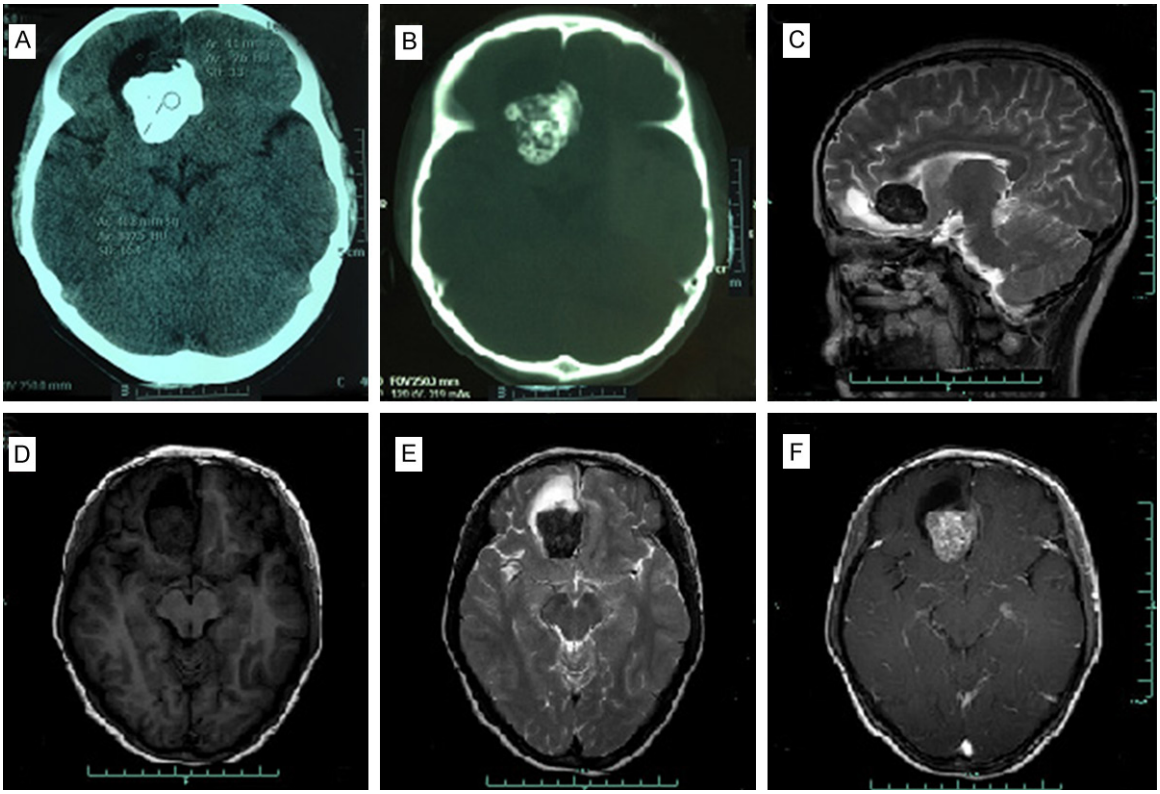

Figure 1. Cranial CT and MRI. Axial CT (A) demonstrates a homogeneous high-density lesion in the frontal lobe when set to bone window (B). Sagittal T2 (C), axial T1 (D), T2 (E) and postcontrast T1 (F) MRI demonstrate a heterogeneous lesion in the frontal lobe, anterior inferior of which was a cystic lesion.

the postoperative imaging revealed no evidence of residual tumor or recurrence.

Discussion

Osteoblastic meningioma is a rare subtype of meningioma, accounting for approximately 1% of all meningiomas [1, 7]. But fewer of them were reported, because hard or calcified lesion was usually not examined. According to Alafaci C's reviewing, only 19 cases have been reported in the literature, including intracranial and spinal meningioma before 1999 [3]. These reports were so early, that the detail could not be found. To the best of our knowledge, there were only 8 cases of intracranial osteoblastic meningioma reported in English, since 1960 [3, 7-9]. After reviewing the published cases, including the present one, the mean age at diagnosis of intracranial osteoblastic meningioma was 42 years old, ranging from 15 to 77 years old, with a female-to-male ratio of 8:1. Three cases were located over the cerebral convexities, three cases involved in lateral ventricles, one in the fourth ventricle and one in the temporo-basal region. We reported an isolated osteoblastic meningioma in the right frontal region. Meningiomas originate from arachno cytes, so they are usually located over the cerebral convexities, Sylvian fissure, or ventricular system. However, some ectopic meningiomas originated from the residual arachnoid tissue in embryonic period, so they were found in scalp, skull, and orbit, including some isolated intracranial meningiomas, like our case. But the mechanism of extraskeletal ossification and cartilage formation is still unknown. Barresi et al assessed the histological findings and immune-expression of bone matrix proteins osteocalcin and osteopontin in seven osteoblastic meningiomas. They found that osseous component occurred in association with psammoma bodies and dystrophic calcification in 5/7 cases, osteocalcin, a marker of terminal osteoblastic differentiation, was positive within the bone spicules in all meningiomas, but not in the chondroid mineralized matrix, and osteopontin, an early osteogenic marker, was extensively expressed by the cell of cases without

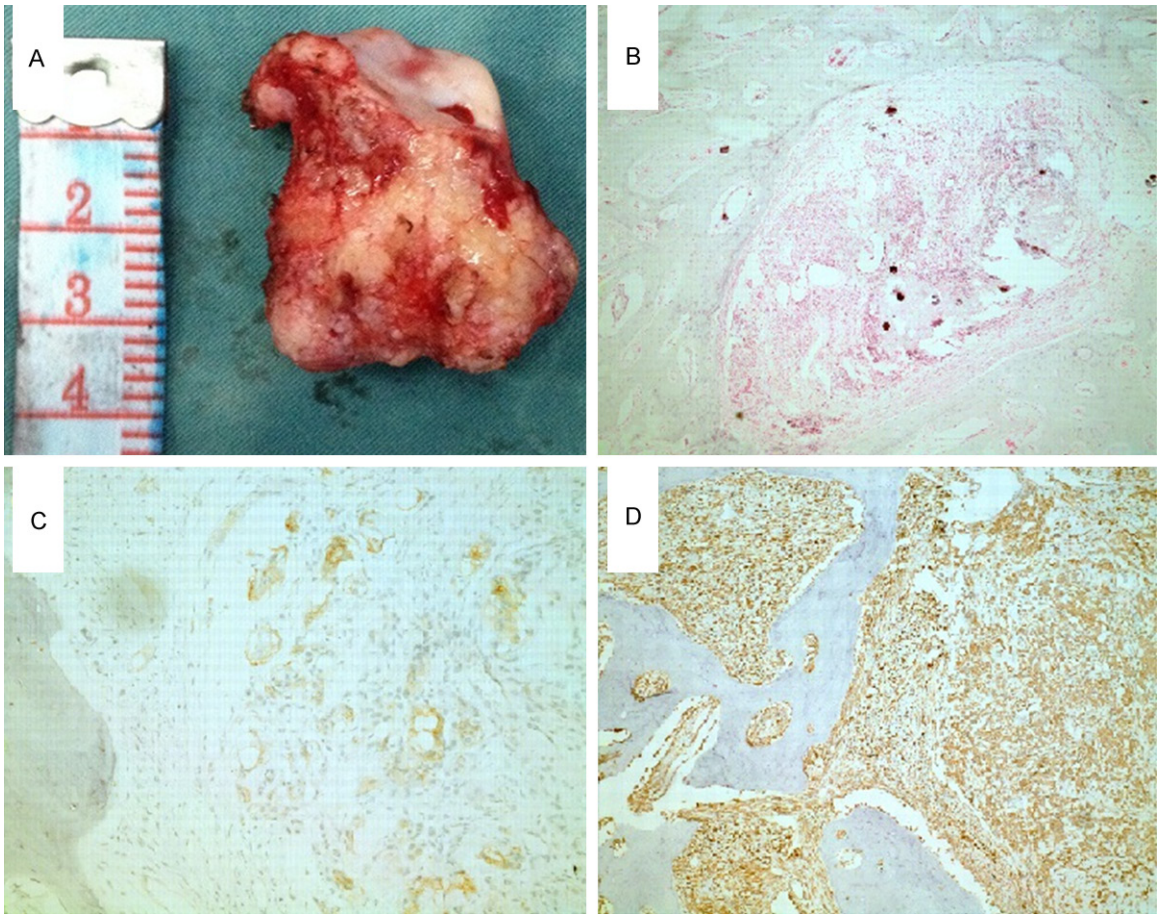

Figure 2. Pathologic photographs. Resected surgical specimen (A) shows a bony mass. Histological section reveals the bone tissue structure, with fibroplasias and small focus of epithelial cells (H&E, ×50) (B). Immunohistochemical staining demonstrates that tumor cells are positive to EMA (×200) (C) and Vimentin (×200) (D).

calcification of psammoma bodies. So they suggested the bone formation may occur through two different pathways, as the final step of calcification or through enchondral ossification [8].

Due to its rarity and no dural tail sign, isolated intracranial osteoblastic meningioma was usually misdiagnosed preoperatively. Clinically, most patients of intracerebral osteoblastic meningiomas presented with headache. Neoplasm locating different eloquent domains may cause different symptoms, such as diplopia, hyposthenia and seizure. In our case, seizure was the initial symptom, because the osteoblastic meningioma was located in the frontal region. Well-defined calcification on CT is the characteristic of osteoblastic meningioma. On MR imaging, osteoblastic meningioma appears hypointense on both T1 and T2-weighted imaging, but with heterogeneous enhancement on T1-weighted imaging with contrast medium and inner core of hypointensity consistent with ossification. Isolated intracranial osteoblastic meningiomas do not display the dural tail sign, as do classic convexity meningiomas.

In pathological examination, psammoma bodied and calcification, as well as evidence of bone spicules and osteon formation can been found in majority of osteoblastic meningiomas. Immunohistochemical staining of the tumor cell were positive for Vimentin and EMA, Ki-67 was usually ≤2% [8].

Extraskeletal ossification and cartilage formation is relatively uncommon among central nervous system tumors. It has been also described in intracranial teratomas, craniopharyngiomas, occasionally in pituitary adenomas [10], gliomas [11, 12], chondroma [13], chondrosarcomas [14, 15] and osteochondroma [16, 17],

besides meningiomas. For differential diagnosis with osteoblastic meningioma, there are some radiological characteristics for other tumors. Intracranial teratomas usually arise from axis, such as suprasellar region, pineal gland, quadrigeminal plate, wall of the third ventricle and cerebellar vermis. Intratumoral cysts admixed with calcified regions and low signal-attenuation supports the diagnosis of intracranial teratomas [18]. Craniopharyngiomas are sellar and/or suprasellar region tumors, with mixed solid and cystic components. Shell-like calcification on the cyst wall is the characteristic of craniopharyngiomas on CT scans. Pituitary adenomas are sellar tumors, and calcification is detected in 0.2% to 14% of cases radiographically. Linear, capsular and granular calcification are the main patterns, and large intratumoral ossification is also reported on rare occasions [10]. Gliomas containing bone and cartilage have been also reported, usually seen in ependymomas, choroid plexus papillomas and oligodendrogliomas [11, 12]. Ependymomas with bone or cartilage arise from the fourth ventricle and occurrence in early childhood with ages of onset from three to five years [12]. Ossified choroid plexus papillomas were also reported, most of which were located in the fourth ventricle. They can appear as well-defined calcification on CT scan and heterogeneous enhancement with hypointensity consistent with ossification [11]. Expect special location, ossified choroid plexus papillomas look like isolated osteoblastic meningiomas on radiological imaging. Intracranial chondromas usually arise from the base skull, such as sphenopetrosal, sphenooccipital and petrooccipital regions, but less frequently from the dura of convexity, the falx cerebri or the choroid plexus. Lobulated, extra-axial lesions with calcification, appearing isodense on plain CT and minimal to moderate levels enhancement are the characteristics for intracranial chondromas on CT scans. On MRI, they appear as homogenous isointense on T1-weighted imaging, mixed hyperintense/hypointense on T2-weighted imaging, and delayed or slight ring-like enhancement on contrast-enhanced imaging [13]. Intracranial chondrosarcomas are similar to chondromas, usually, arising from skull base, but sometimes there is no demonstrable connection with skull or meninges. Mesenchymal chondrosarcomas appear as well defined, firm, and calcified, contrast-enhancing masses on CT scan and heterogeneous intensely enhancing masses on MRI. Invasive growth and widespread metastases are usually found in chondrosarcomas [14]. Osteochondromas are another skull base neoplasms. It is reported that half of them arose in the skull base, and the others were found in the convexity and the falx. They appear as high-density masses, with or without enhancement after contrast infusion on CT. The MR appearance did not change between T1 and T2-weighted imaging, consistent with the calcification with the tumor [16].

Operation is an effective treatment for intracerebral osteoblastic meningioma, and the prognosis is good. Recurrence did not occur in all of the patients, including our case, in the one-year fellow-up after the operation.

Conclusion

In conclusion, we provided a rare case of isolated intracranial osteoblastic meningioma in the right frontal region. Osteoblastic meningioma typically appears as well-defined calcification on CT and hypointense on both T1 and T2-weighted imaging, but with predominant enhancement. Due to the rarity and no dural tail sign, isolated osteoblastic meningioma was usually misdiagnosed preoperatively. Operation is an effective treatment and the prognosis is good for the patient receiving total resection of the neoplasm.

Disclosure of conflict of interest

None.

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