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CASE REPORT Open Access

Surgical treatment of elderly patients with primary osteolytic atypical meningioma: a case report and review of the literature

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Abstract

Background: Elderly patients with primary intracranial osteolytic and externally growing atypical meningiomas are rare and easy to be misdiagnosed. Recently, a patient with an atypical meningioma was treated in our department and analyzed the case by reviewing the historical literature.

Case presentation: We describe a 63-year-old female with primary intracranial osteolytic atypical meningioma at our neurosurgery department, and retrospectively reviewed previous literatures about its diagnosis, surgical treatment, pathological results, and clinical outcome. Simpson grade I resection was accomplished through a pterional approach. First-stage skull reconstruction was performed, and the patient underwent an uneventful recovery.

Conclusions: The final diagnosis of the primary osteolytic atypical meningioma is dependent on a pathological examination. First-stage skull reconstruction could avoid a secondary lesion and improve the patient's quality of life.

Keywords: The elderly, Primary, Atypical meningioma, Osteolytic

Background

Elderly patients with primary intracranial osteolytic and externally growing atypical meningiomas are rarely encountered in clinical practice [1]. Such tumors not only develop intracranially, but also invade the dura mater to grow subdurally. Atypical meningiomas differ from primary ectopic meningioma tumors within the skull and common subdural meningiomas. Recently, our department treated a patient with an atypical meningioma and analyzed the case by reviewing the historical literature.

Case presentation

A 63-year-old female patient with an atypical meningioma was treated in our department with a complaint of dizziness for 1 month. She had fallen down accidentally while riding and sustained a chest contusion without head injury 1 month earlier. The dizziness appeared thereafter. Cranial computed tomography (CT) showed a slightly hypertensive lesion in the lower inner panel of the left temporal bone and an expansive local skull plate, including soft tissue density, leading to the consideration of an intracranial mass (Fig. 1). The patient reported no headache, nausea, hemiplegia, aphasia, or limb convulsions since the onset of dizziness. Additional relevant examinations were conducted after admission, and a physical examination showed no positive pathological signs. A cranial dual-source and thin-layer CT scan revealed local expansive lesions in the left temporal bone with invasion into the skull and osteolytic lesions. Based on 3-dimensional skull imaging, a postoperative skull defect was designed and a 3-dimensional artificial skull was created for first-stage reconstruction. Magnetic resonance imaging (MRI) revealed an area of frontotemporal, subdural, and intracranial homogeneous enhancement on the left side with an approximate diameter of 4 cm, with no obvious dural tail sign (Fig. 2). The tumor was divided into inner and outer parts by the inner skull plate.

The tumor was removed via a modified pterional approach under general anesthesia. The skin flap was turned to the base of the skull, and the local skull bulge, measuring 2 cm × 2 cm, was visible. Next, a 4-cm × 4-cm

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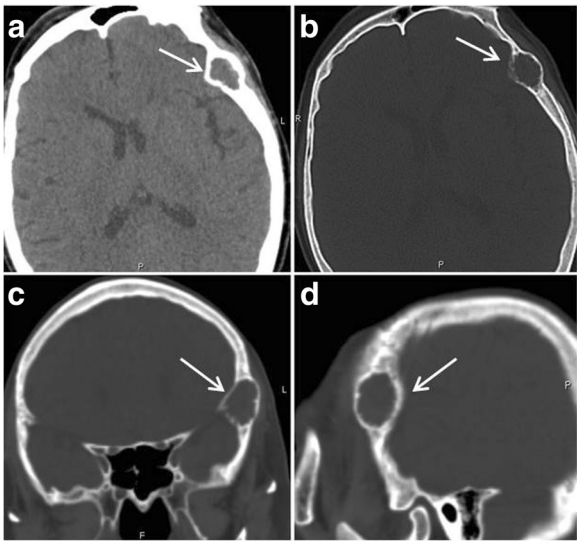

Fig. 1 **a** and **b** shows presurgical cranial computed tomography revealed a slightly hypertensive lesion in the frontotemporal region and an inconsistent inner skull plate. **c** and **d** represent the local skull plate was expansive and included soft tissue density (*white arrow*)

bone flap, centered on the skull bulge, was resected to reveal tumor protrusion to the inner side of the skull. Although the tumor had not adhered to the dura mater, local tumor invasion into the dura mater was visible. Having incised the dural mater, the subdural part of the tumor measured approximately 2 cm × 2 cm and had compressed the brain lobe in a convex manner. The tumor boundary was clear without obvious adhesions to brain tissues, and the en block resection of the tumor along with the dural pedicle was performed to achieve a Simpson grade I resection (Fig. 3). Following artificial dura repair, the skull was repaired using the preoperatively designed artificial titanium skull.

The involved skull, subdural tumor, and dural pedicle were sent for a pathological examination (Fig. 4). The pathological results including immunohistochemical analysis indicated a left frontotemporal tumor with invasion into the dura mater and bone tissue, leading to a diagnosis of atypical meningioma (WHO Grade II). Paraffin-embedded sections revealed that the skull had been invaded, with a tumor cell cluster appearing inside the skull. High-density areas of tumor cells were seen in the endocranium and tumor pedicle. The tumor cells were small with large nuclei, a high nucleus:cytoplasm ratio, and prominent nucleoli. Immunohistochemical analysis revealed SYN (–), CD56 (+), KI67(N) (with a minority of positive cells), AE1/3 (–), GFAP (–),EMA (+), VIM (+), OLIG2 (–), CD34 (–), S100 (–), DES (–), SMA (–). The patient underwent an uneventful recovery, and a follow-up cranial MRI revealed total tumor resection.

Discussion and conclusions

Meningioma is the second-most common type of central nervous system tumor, accounting for 15–20% of all such tumors. Most meningiomas are benign, with only 10% of meningiomas and 4.7–7.2% of atypical meningiomas presenting with malignant characteristics [2]. Atypical meningioma is a type of low-grade malignant meningioma and is classified as Grade II according to the World Health Organization Classification of Tumors

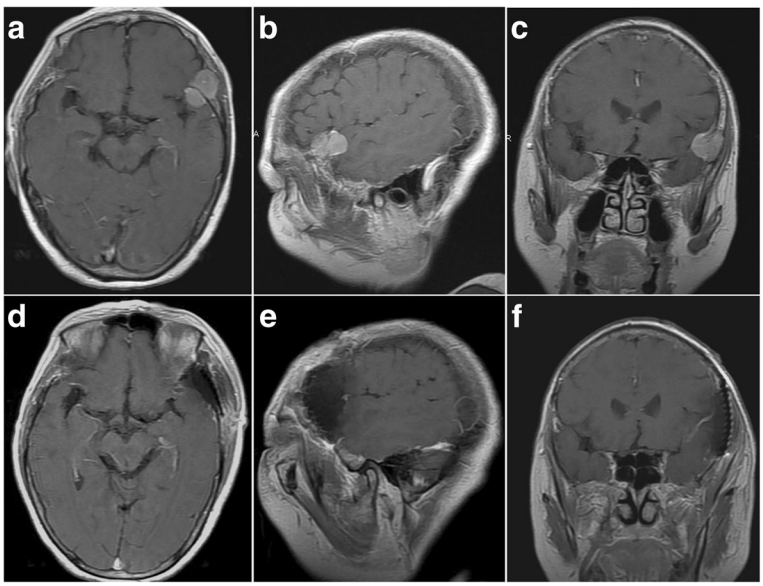

Fig. 2 Presurgical and postsurgical cranial magnetic resonance imaging revealed *left* frontotemporal, subdural, and intracranial homogeneous enhancement with no obvious dural tail sign (**a**, **b** and **c**). First-stage repair was performed after en bloc tumor resection (**d**, **e** and **f**)

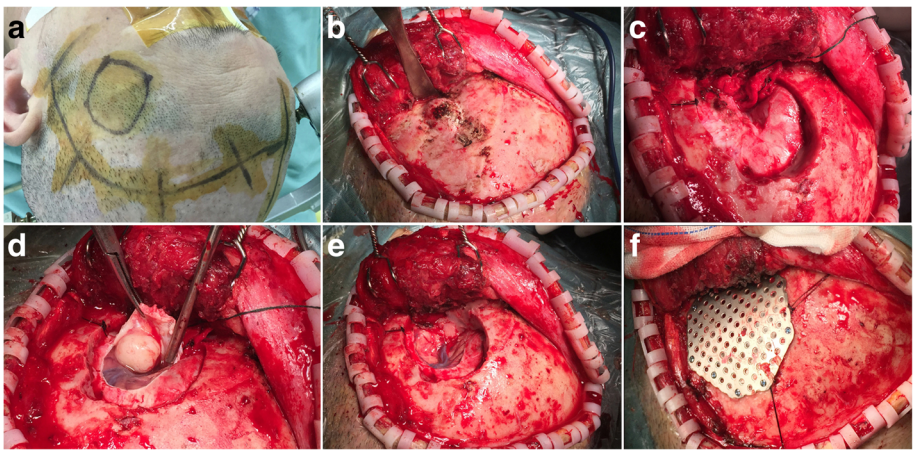

Fig. 3 The surgical procedure was as follows: the skin flap was opened to reveal the local skull bulge through a *left* frontotemporal incision (**a** and **b**). **c** After removing the invaded skull, the dura mater and skull were not found to adhere to each other. **d** The dura mater was incised to reveal the tumor, and the base of the tumor was found to adhere to the dura mater. Therefore, the tumor and dura mater were resected (**e**), and first-stage repair was performed (**f**)

of the Central Nervous System. The radiological manifestations of atypical meningiomas vary, although the main manifestation is a strongly invasive, non-uniformly dense mass with surrounding edema.

However, atypical meningiomas not only cause osteolytic destruction but also can invade toward both the inner and outer surfaces of the skull; accordingly, an intact dura mater is quite rare. In the present case, an elderly patient presented with a primary osteolytic atypical meningioma and concurrent invasion into the dural mater. Primary osteolytic atypical meningioma is a term used to describe a subset of extradural atypical meningiomas that

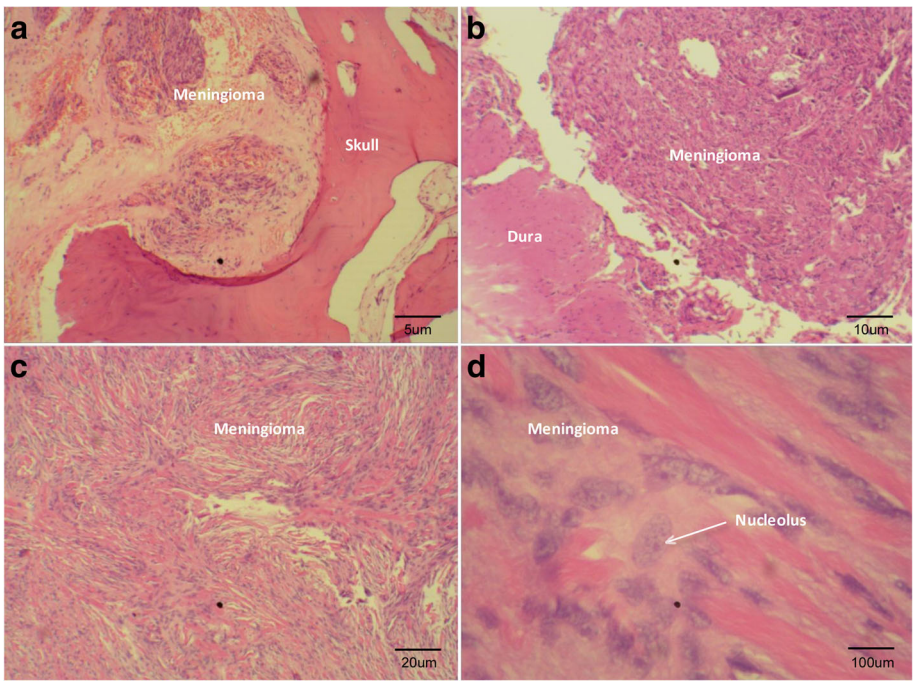

Fig. 4 a Postoperative paraffin pathology demonstrated tumor invasion into the intraoperative resected skull, with a tumor cell cluster inside the skull (magnification, × 40). High-density areas of tumor cells were observed in the endocranium and tumor pedicle (**b** and **c**). **d** The tumor cells were small with large nuclei, a high nucleus:cytoplasm ratio, and prominent nucleoli

Table 1 Case reports of primary atypical meningiomas with osteolytic invasion as confirmed via pathological examination

No	Year	Author	Sex	Age	Location	Invasion of endocranium
1	1995	Partington [4]	Female	84	Left frontal	Full layer
2	2000	Lang [3]	Male	59	Right sphenoid greater wing	Full layer
3	2005	Tokgoz	Male	44	Right frontotemporal	Outer layer
4	2006	Bassiouni	Female	62	Left frontal	Full layer
5	2011	Cheng HT	Female	68	Left frontal	Full layer
6	2012	Kim	Male	68	Left parietal	Outer layer
7	2014	Yun JH	Female	65	Right frontal	Full layer

arise in bone [3]. This type of meningioma approximately accounts for two-thirds of all extradural meningiomas [3].This type of case is rare in clinical practice, with only 7 cases reported to date [3, 4] (Table 1). Literatures about its diagnosis, these tumors can be divided into 3 categories [3]: (1) tumors that only violate the skull and grow in diploe; (2) tumors that invade the skull to grow outward, possibly into the skin; and (3) tumors that produce osteolytic destruction and grow into the dural mater, possibly causing subdural damage. In this case, the main part of the tumor was located in the skull, so we considered that this tumor was primarily an intraosseous meningioma. The present case belonged to the third category.

Although it is not difficult to diagnose an atypical meningioma based on the clinical history and imaging characteristics, these tumors can be misdiagnosed as other tumors because of skull invasion. Intraosseous meningiomas most commonly present as slowly growing scalp masses, with possible relationship to a cranial suture. Intraosseous meningiomas are mostly characterized by osteoblastic or mixed osteoblastic osteolytic lesions. Accordingly, this disease primarily requires differentiation from the following diseases. (1) Secondary skull metastasis mainly occurs in elderly people. The appearance of a confirmed primary tumor concurrent with skull invasion indicates a secondary skull metastasis. This disease also presents with osteolytic damage, unclear borders, and obvious edema surrounding the lesion if the brain parenchyma is violated. (2) Eosinophilic granuloma often occurs in adolescents with a history of trauma. A local mass in the parietal, occipital, and temporal bones, slight pain, and slow proliferation are common. Eosinophilic granulomas originate from the skull diploe and simultaneously expand inward and outward; outward expansion with a clear border is the main presentation. The pathologic findings generally include a large number of proliferated and well-differentiated Langerhans cells. Large populations of eosinophils, lymphocytes, plasma cells, and neutrophils with occasional multinucleated giant cells are also present in these lesions. (3) Multiple myeloma mainly occurs in the middle-aged people and is much more prominent among men than women. Multiple myeloma can invade the bone marrow and reticuloendothelial systems and occurs preferentially in the red marrow. This disease usually involves the skull, vertebrae, ribs, pelvis, sternum, and long bones in sequence. In addition, the frontal, parietal, and occipital areas of the skull are often affected, and widespread metastasis can be observed in late-stage disease. So, we emphasize that atypical intraosseous meningioma must be considered in the differential diagnosis of osteolytic skull tumors.

Surgery is the main treatment for atypical meningioma, and totally tumor resection is the key predictive prognostic factor. In the present case, the tumor had invaded the skull and passed through the dura mater to reach the subdural area. However, in this elderly patient, the meningioma had invaded the skull, requiring maximum resection of the involved bone tissue, dura mater, and tumor. Based on the physiological characteristics of the elderly, we customized an artificial skull bone before surgery according to the possible bone defects, after which a first-stage skull reconstruction was performed. This procedure allowed us to avoid a second-stage surgery and facilitated the patient's uneventful recovery. According to our experience, the resection should extend to 10 mm distal from the involved lesion to reduce the possibility of tumor recurrence. Recurrence of atypical or primary intraosseous meningiomas has been reported to occur up to 2 years after resection, but histologically benign tumors have been reported to recurrent up to 10 years after surgery [3]. Given the high rate of recurrence of atypical meningiomas, many scholars preferred to administer immediately postsurgical adjuvant radiotherapy to achieve a lower recurrence rate [5]. However, we believe that for elderly patients in whom surgery has achieved a Simpson grade I resection, the indications for postoperative radiotherapy should be cautious and long term follow-up is required. For older patients with atypical intraosseous meningiomas, postoperative radiotherapy may damage the normal brain tissue and cause cranial nerve injuries, worsening the patients' quality of life.

The atypical meningioma with intracranial osteolytic and externally growing are rare in elderly patients. We emphasize the importance of considering primary intracranial osteolytic atypical meningioma in the differential diagnosis of osteolytic skull tumors.. A minimally invasive approach should be taken, based on the individual characteristics and tumor growth pattern. Meanwhile, Simpson grade I resection and skull reconstruction at the same time should be performed to reduce postoperative morbidity.

Additional files

Additional file 1: Ethics approval and consent to participate (CHINESE). (JPG 176 kb)

Additional file 2: Ethics approval and consent to participate (ENGLISH). (JPG 192 kb)

Additional file 3: List of Ethics committee. (JPG 219 kb)

Abbreviations
CT: Computed tomography; MRI: Magnetic resonance imaging

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Availability of data and materials
The authors declare that the data supporting the findings of this study are available within the article and its Additional files 1, 2, and 3.

Authors' contributions
All authors read and approved the final manuscript. Lei Du collected the patient data and wrote the primary manuscript. Yinda Tang helped to draft the manuscript. Hua Zhao and Xuhui Wang performed in the surgery of the case. Xuhui Wang participated in its design and coordination. All of authors read and approved the final manuscript.

Ethics approval and consent to participate
This study was undertaken at the Xinhua Hospital (Shanghai, China) with approval from the Xinhua Institutional Review Board(Approval No. XHEC-D-2017-033).

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests
The authors declare that they have no competing interests, and manuscript is approved by all authors for publication.

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