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# Intraosseous Meningioma with Extra and Intracranial Invasion: Literature Review and Case Report

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**Abstract:** Primary intracranial meningiomas are a rare subgroup of tumors, accounting for less than 1% of all tumors. In addition, it is included among the tumors that penetrate into the dura mater and subcutaneous tissues and damage the tissues that are close to it. Unlike intracranial meningiomas, the diagnosis and treatment of PIOM is difficult due to their misleading clinical signs and the lack of clear radiological diagnostic criteria.

**Keywords:** intraosseous meningiomas, skull, osteogenesis, surgical techniques, PIOM, dural invasion, extracranial spread.

## Introduction

Most reviews of bone lesions in the head and neck region include the base of the skull, face, and spine [1, 2, 3], while, to our knowledge, there are fewer practical illustrated reviews that specifically address calvarium lesions. However, the calvarium deserves attention as it has its own limited range of diseases with some unique formations and others ubiquitous that may have specific features in this localization. Anatomically, the cranial vault or cranial vault is formed by the convex part of the cerebral skull, consisting of flat bones: both frontal bones, both parietal bones, the squamous part of both temporal bones and the interparietal part of the occipital bone. They are embryologically formed by intramembranous ossification, in contrast to the base of the skull (chondrocranium), which is formed by endochondral ossification [4]. Cranial lesions are often found incidentally on imaging of the skull. Thus, lack of clinical information or non-specific manifestations can further complicate characterization. In this context, mixed pseudo-lesions (eg, arachnoid granulations, vascular canals) are often found at this site. Moreover, specific benign lesions (meningioma, hemangioma) may have apparently aggressive bone involvement when analyzed using the classic imaging features used for other extracranial bones.

The purpose of this review is to serve as a practical, illustrative guide to a range of lesions that a radiologist may encounter. Thus, the anomalies were broadly classified according to their main components: lytic, sclerotic, or transdiploic (with soft tissue components exceeding the cranial vault). Additional features such as periosteal reaction, cortical bone expansion, transition zone, bone sequestration, bone penetration, soft tissue features and enhancement, as well as relevant clinical and demographic information, were taken into account. Finally, scenarios were included in which functional imaging techniques such as MR spectroscopy, MR diffusion, MR perfusion curve analysis, and PET can provide relevant clues. Recognition of benign and malignant imaging features is important for radiological diagnosis [1, 2, 3, 4, 5, 6]. In general, benign tumors have clear boundaries with a narrow transition zone; sclerotic margins are often present. On the other hand, malignant tumors have poorly defined margins, a wide transition zone, an aggressive periosteal reaction, and often have a soft tissue component; these lesions cause abrupt bone destruction with



intracranial or extracranial spread [1, 4].]. Skull lesions may be lytic or sclerotic, single or multiple, of varied composition; they may arise from osteogenic, chondrogenic, fibrogenic, vascular, and/or other bone elements.

## **Case presentation**

1. A 76-year-old man was admitted to the neurosurgery department because of a headache and a 6month-old tumor in the right frontal area of the skull. There was no previous trauma in this area. Neurological examination and laboratory data do not show significant abnormal changes. Computed tomography (CT) scan of the skull shows a  $61 \times 55 \times 56$  mm osteogenic calvarial lesion in the forehead area. The damage covered the entire layer of the frontal bone, forming gall-like holes in the outer layer of the skull (Fig. 1A). Considering the possibility of osteosarcoma as judged by the radiologist, we performed a skull resection at the most prominent site of the lesion. The result confirmed intraosseous ectopic meningioma. Before the operation, the approximate size of the resection was determined according to neuroimaging examinations. Information obtained during CT 3D reconstruction was used to ensure a good cosmetic effect. After the scalp was separated during surgery, the damage to the skull was not strongly attached to the scalp, but partially infiltrated into the subcutaneous tissue. Loss of normal bone tissue is observed in the periosteum and diploic spaces of the outer plate. The dura mater was thickened in the center and a layer of easily separated gelatinous tissue appeared on the frontal lobe surface without brain tissue invasion. Then, the affected tissues related to the scalp, skull and dura were completely resected. The dura mater defect was closed with normal autologous fascia and dural sutures were placed to prevent epidural fluid collection. After that, the cranioplasty was completed.

Postoperative CT scan showed complete resection of the skull tumor and complete symmetric restoration of the bone (Fig. 1B). Microscopic examination of stained sections of the histological preparation revealed that the intertrabecular spaces were infiltrated by tumor cells with eosinophilic cytoplasm and round nuclei, which had invaded the subcutaneous space and dura. According to the general characteristics, the lesion was evaluated as a meningioma of the meningothelial type (WHO grade I). Recovery went smoothly. After 3 months of follow-up, no recurrence or other complaints were noted.

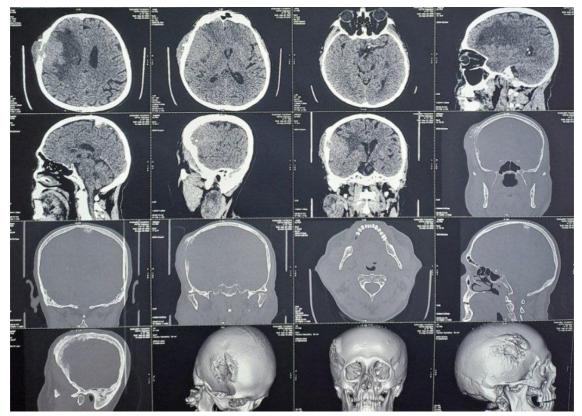


Fig 1. A Preoperative MSCT scan.



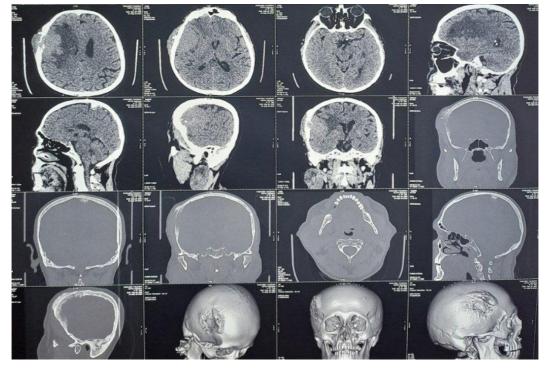


Fig 1. B Postoperative MSCT scan.

2. A 40-year-old male patient presented with complaints of lesion of skull and seizures. The patient has been ill for 5 years. Neurological examination and laboratory findings were within normal limits. Neuroimaging studies revealed a 105x77x98mm with a mass-effect and destruction of skull (Fig. 2A). Subsequent magnetic resonance imaging (MRT) revealed a hypointense intraosseous lesion in T2 mode (Fig. 2B). According to MSCT data, malignant osteosarcoma was suspected, and total removal of the neoplasm without simultaneous defect plasty was planned. The patient underwent the operation "Cranioectomy of the right fronto-parietal-temporal region with the removal of an extracerebral extra- and intracarnial neoplasm". Postoperative images showed total resection of the neoplasm with elimination of the mass effect (Fig. 2C). Histological analysis showed a benign G1 - transient meningioma.

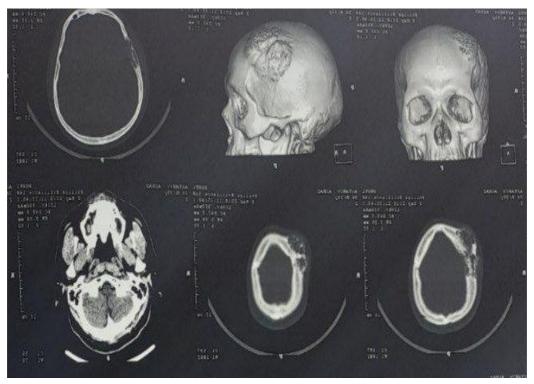


Fig 2. A Preoperative MSCT scan.

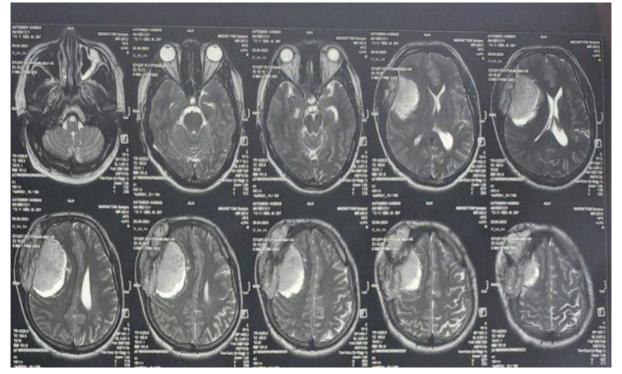


Fig 2. B Preoperative MRI.

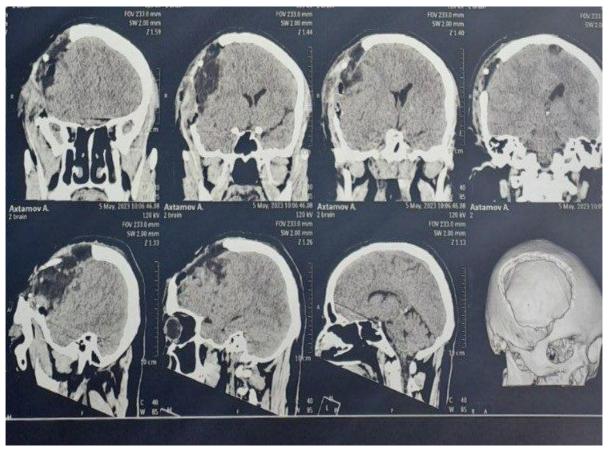


Fig 2. C Postoperative MSCT scan.

#### Discussion

Most PEMs are rare tumors involving bone and subcutaneous tissue, approximately two-thirds of which are PIOMs. The prevalence of these meningiomas is highest at the age of 60. It is more common in women than in men (85%) (5). The frontal and orbital areas are the most common sites of PIOMs(6). These tumors grow slowly without causing neurological symptoms in patients. The presence of some tumors that invade the meninges during surgery raises the question of whether they



should be classified as PEMs. Bassiouni et al (3) demonstrated dural invasion in 14 (88%) of 16 PEM patients who underwent surgery. In another report, intraoperative macroscopic examination showed no evidence of dural invasion, but subsequent pathology confirmed tumor infiltration of both layers of the dura (8). Recently, Ahmed et al. (9) proposed a new classification of PIOMs, including type IV (mixed variety) as tumors that grow outside the skull from the dura. Therefore, dural resection and duraplasty were performed in our case. Localization of the primary area of the tumor within the skull can be done to determine its origin and determine whether it is PIOM or PEM, regardless of the presence of dural invasion. Several studies have found (4, 10, 11) that PIOMs have a wider base in the skull than in the dura, and tumors of meningeal origin, such as meningiomas, have a wider base in the dura than in the skull. Therefore, we considered our case to be a PIOM involving dura and soft tissue rather than an intracranial meningioma involving bone and soft tissue.

To date, several hypotheses have been proposed for the origin of PIOMs. They may result from abnormal differentiation and migration of mesenchymal stem cells (12,16). In addition, blood vessels and nerves entering the skull can transport arachnoid cells to various locations where they can then proliferate (7). Other theories suggest that arachnoid cover cells may be trapped at post-traumatic fracture lines or cranial sutures during cranial growth (13,15). In one study, imaging features of PIOMs included hyperostosis in 56% of meningiomas, osteolysis in 33%, and mixed features in 11% (14). These features may be due to neoplastic changes occurring within the skull that stimulate osteogenic or osteoclast activity to produce different stages of bone reaction and bone resorption (5). These features also make the radiological diagnosis of PIOMs challenging. Osteomas, primary cranial lymphoma, fibrous dysplasia, eosinophilic granulomas, and metastatic tumors are often part of the differential diagnosis (4, 14). In our case, CT bone mode showed osteoblastic hyperplasia. MRI is valuable for detecting extraosseous involvement. We observed dural enhancement distinct from the dural tail sign of intracranial meningiomas in our patient. Preoperative diagnosis has been mistaken for cranial osteosarcoma because its radiographic appearance is similar to PIOM, but the treatment of the former mainly includes radiotherapy and chemotherapy (11). The final diagnosis of PIOM was confirmed by pathology, indicating the importance of histopathological biopsy. A biopsy is appropriate if the diagnosis is difficult to make when imaging a skull injury. Intraosseous meningiomas are mostly of good quality. However, they often present as malignant tumors with both intraosseous and extraosseous spread (14,15). In general, the osteoblastic type has less atypical or malignant features than the osteolytic type (2). Even when the histology is benign, PIOMs can appear very aggressive. In this case, osteoblasts of the skull penetrate into the subcutaneous tissue and dura and spread to the surface of the forehead as a layer of gelatinous tissue without breaking through the pia mater, which is extremely rare. According to the latter criteria, an atypical meningioma (WHO grade II) is considered if the tumor cells invade the pia mater and spread to the brain (13,15). Therefore, in this case, the damage tends to get worse.

In the literature, good-grade PIOMs have a recurrence rate of 22%, and lesions located at the base of the skull have a higher recurrence rate than those located on the surface of the brain, which may be related to incomplete resection (1, 14,16). Therefore, total resection should be attempted, including removal of any surrounding tissue that may contain tumor cells. In our case, the patient may be at risk of relapse. We did not use a neuronavigation system to determine the resection line before surgery, and it was not possible to accurately and maximally remove the damaged skull. The extent of resection at a distance of at least 10 mm from the margin of damage corresponded to the safe distance reported in the literature (12). Damaged dura and subcutaneous tissues were also resected. In addition, computer-aided reconstruction technology was used to individually adjust the titanium mesh for the planned resection area to complete the cranioplasty in one step to achieve a better aesthetic effect. For our cases, titanium mesh was chosen because of its low cost, aesthetic effect and low infection rate, poor thermostability and resistance to deformation under trauma. The dural defect was repaired by watertight suturing of the temporalis muscle fascia rather than artificial meningeal application for the following reasons. First, the former can reduce the likelihood of chronic inflammatory reactions and has a good histocomposition. Second, the use of artificial dura mater is more prone to epidural fluid accumulation. In addition, the high cost of artificial dura mater was also considered.



# Conclusion

We encountered a rare case of osteoblastic intraosseous meningioma arising from the cranial diploic layer and invading the dura mater and subcutaneous tissue. Even when the histology is benign, PIOMs can appear very aggressive. Surgery is required to achieve complete resection of the lesion to prevent recurrence and worsening. The clinical features and surgical techniques presented in this work may be a good reference for the diagnosis and treatment of similar diseases in the future.

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