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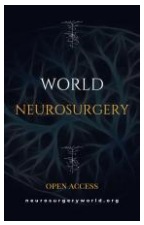

Case Report and Review of the Literature

Hidden in the Bone: Intraosseous Meningioma, a Rare Variant of a Well-Known Tumor: Case Report and Review of the Literature

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ARTICLE INFO

Keywords:
Intraosseous meningioma
parietal bone
surgical resection
arachnoid cells

ABSTRACT

Introduction: Int primary intraosseous meningiomas are rare extradural tumors. They are typically slow-growing, painless, and asymptomatic until they cause a mass effect. We report the case of a 71-year-old woman presenting with an intraosseous meningioma located in the right parietal bone, a rare entity that often poses a diagnostic challenge.

Case Report: We report the case of a 71-year-old woman who presented with a right parietal swelling that progressively increased in size over 2-3 years without associated clinical signs. The patient benefited from a full radiological workup returning in favor of a right parietal intraosseous lytic meningioma. The lesion had a preoperative diagnosis of osteoid osteoma of the right parietal bone and the tumor was completely excised. Histopathological examination confirmed the diagnosis of an intraosseous meningothelial meningioma. Primary intraosseous meningiomas should be considered, especially when they can exhibit radiological features that may be indistinguishable from cranial osteoid osteomas. Key radiological diagnostic features are presented.

Conclusion: Intraosseous meningiomas are rare lesions that originate in the skull and represent the most common type of extradural meningioma; the lesions are often asymptomatic but can cause proptosis and neurologic symptoms depending on their size and location. Intraosseous meningiomas likely originate from entrapment of arachnoid cells within the bone. The treatment of choice is surgical resection, which is potentially curative.

1. Introduction

Meningiomas are extra-axial tumors and represent the most common form of tumor of the meninges, those originate from arachnoid cap cells of the meninges. Les méningiomes extracrâniens sont rares; l'incidence rapportée est de 1 à 2 % de tous les méningiomes [1, 2]. Intra-osseous meningioma, also referred to as primary intra-osseous meningioma, is a rare subtype of meningioma that accounts for less than 1% of all ossesous tumors. They are the most common type of primary extradural meningiomas [3]. Here, we present a case of primary intraosseous meningioma.

2. Case Report

A 71-year-old woman presented with a painless swelling over the right parietal region that had progressively increased in size over a period of 2-3 years. There was no associated history of trauma or other clinical signs. Physical examination revealed a firm, non-tender swelling over the right parietal area. The overlying skin was normal, and there were no signs of neurological deficit. Laboratory tests were within normal limits. Noncontrast CT of the head was performed (Figures 1a & 1b). The lesion was further characterized by using pre- and postcontrast MR imaging (Figures 2a-2f).

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https://dx.doi.org/10.31487/j.WNEURO.2025.01.07
Received 25 June, 2025; Accepted 16 July, 2025
Available online 22 July, 2025

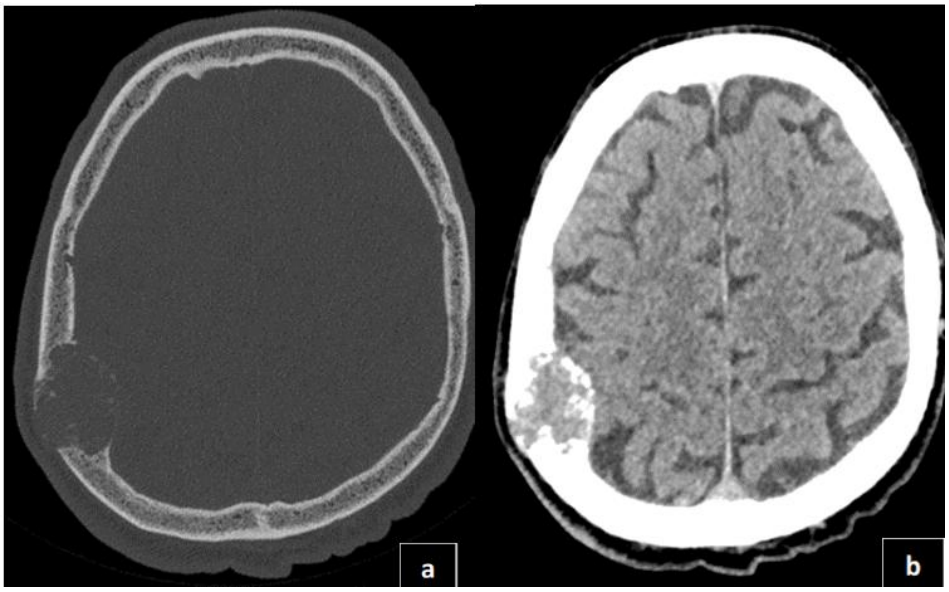

Fig. 1. Contrast-free CT scan of the brain, axial section, **a)** ossicular window and **b)** parenchymal window showingan osteolytic lesion with anterior hyperostosis of the right parietal bone. Intraosseous right parietal lesion, 34 × 24 mm, blowing, multiloculated, partially calcified with peripheral cortical rupture without soft tissue invasion.

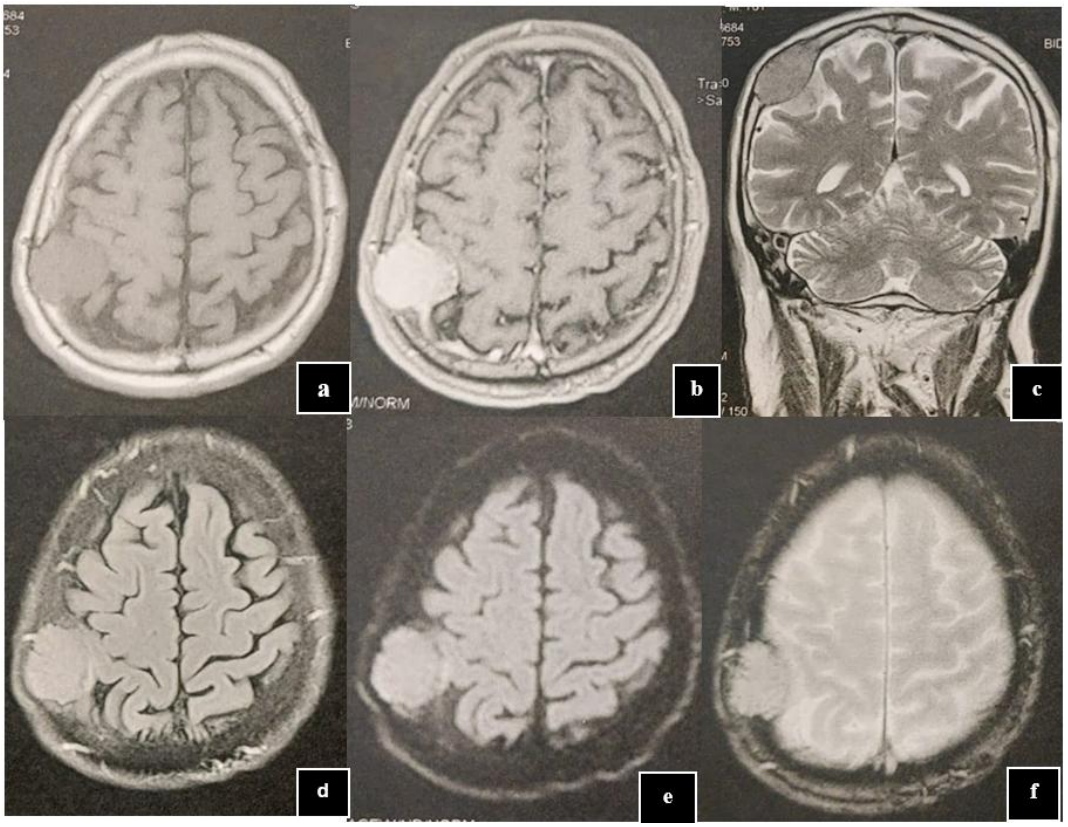

Fig. 2. Cerebral MRI showing a well-circumscribed intraosseous lesion in the right parietal calvaria, with no extension to subcutaneous soft tissue or underlying brain parenchyma. On **a)** T1: The lesion appears hypointense, with no obvious fatty or haemorrhagic component. **c)** T2: The lesion is also hypointense. On **d)** FLAIR and gradient echo sequences **f)** (T2)*: The lesion is isointense. **e)** Diffusion (DWI/ADC): No diffusion restriction. After injection of **b)** Gadolinium: Homogeneous intralesional enhancement, without meningeal extension or cerebral mass effect, pointing to a benign, slow-growing intraosseous lesion. Radiological differential diagnoses primarily include intraosseous meningioma, but also osteoma, intraosseous hemangioma and, more rarely, metastatic lesions or plasmacytoma.

The patient was operated on under general anaesthetic. A bone flap adherent to the dura was removed. This flap, adherent to the dura, was carefully detached using metzenbaum scissors. Local hemostasis was achieved by applying bone wax to the bone and bipolar coagulation to the dura, which had been invaded by the tumor. The invaded dura was then carefully incised with hook, scalpel and metzenbaum scissors to fully expose the lesion. Ecompete the adherent intradural tumor was progressively dissected from the underlying arachnoid and feeder vessels, using bipolar coagulation and careful sectioning with micro-scissors. The tumour material was sent for anatomopathological study. Careful parenchymal and dural hemostasis was achieved by bipolar coagulation. A dural plasty was performed using galea, followed by a cranioplasty performed with bone cement fixed in place. Surgical closure was performed plane by plane using standard techniques.

Pathological examination was in favor of a meningothelial meningioma infiltrating the dura mater and the bony cap, without invasion of the cerebral parenchyma. Tumor proliferation with very low expression of anti-RP antibodies. Proliferation index estimated with Ki67 is 1%. The immediate post-operative course was favourable, with no complications and a satisfactory follow-up brain scan (Figure 3) showing complete removal of the lesion with satisfactory bone reconstruction. At the end of a six-month follow-up, the patient maintained a good clinical course, with no neurological deficits or complications. Brain MRI (Figure 4) at this stage revealed no tumour recurrence and adequate tissue healing.

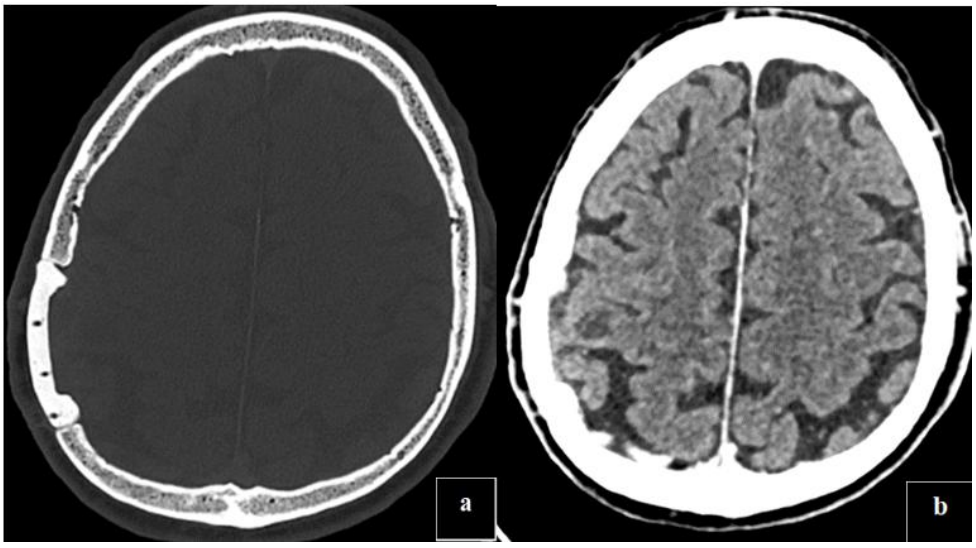

Fig. 3. Post-operative brain scan **a)** bone window and **b)** parenchymal window.

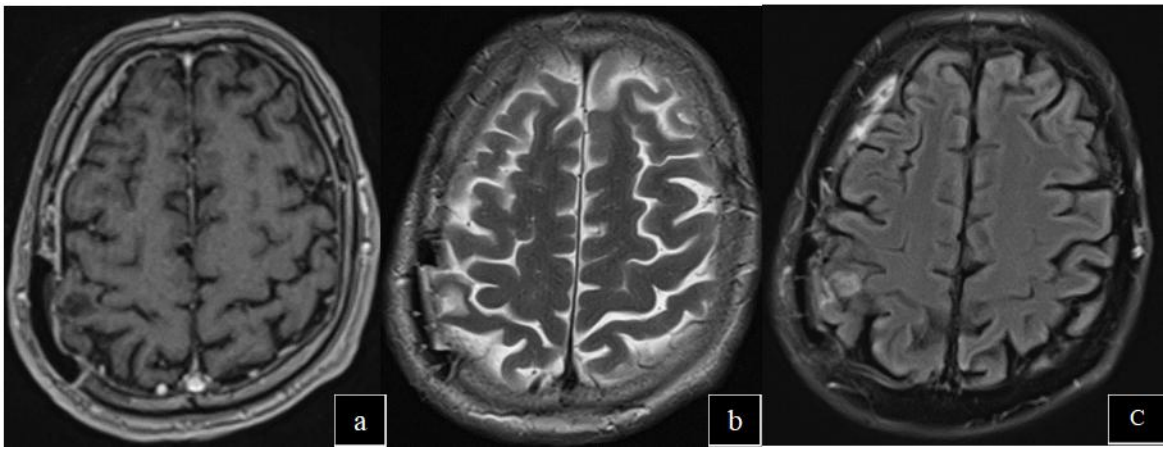

Fig. 4. Six-month post-operative follow-up brain MRI **a)** injected axial sequence, **b)** axial T2, and **c)** axial FLAIR, shows no evidence of recurrence or complication.

3. Discussion

Intraosseous meningiomas (IM) are rare lesions that originate in the skull and represent the most common type of extradural meningioma; the lesions are often asymptomatic but can cause proptosis and neurologic symptoms depending on their size and location [4]. The etiology of IM has not been cleared yet. However, proposed theories include some presumed ectopic arachnoid cap cells in an extradural location, entrapment/detachment of displaced pacchionian bodies during embryonic development, displacement of arachnoid islets by a traumatic

event or cerebral hypertension or a separate origin from a differentiated or multipotential mesenchymal cell [5]. Wrinkler, in 1904, first described a meningioma originating in an extradural location [6]. The true incidence of primary meningiomas is unknown as they have largely been reported in the literature as case reports [7]. True primary IM is defined as a lesion that does not involve the underlying dura [8, 9]. However, IM scan be classified as primary and secondary, and the dura may become involved later in the course of the tumor growth even in primary IM [10]. Secondary IM is due to the extension of an intracranial meningioma into the calvarium [10]. However, the underlying dura may be affected by primary or secondary IMs [11].

The frontoparietal and orbital regions are the most common sites of primary intraosseous meningiomas [12]. Patients usually exhibit no neurological symptoms but only a local, slow-growing bulge. Ophthalmoplegia or visual field defects may be present when the tumor is located in the skull base [13]. The radiological differential diagnosis of cranial bone lesions mainly includes fibrous dysplasia, osteoma, osteosarcoma and Paget's disease. Fibrous dysplasia usually stops growing after puberty, while primary intraosseous meningiomas (PIM) occur at a later age and progress slowly [14]. The typical MRI appearance of MIPs is that of a T1 and T2 hyposignal lesion, with homogeneous contrast after gadolinium injection, which distinguishes them from osteoma (dense, well-limited lesion with no enhancement), osteosarcoma (aggressive lesion with irregular contours, heterogeneous signal and enhancement), and Paget's disease (heterogeneous signal with variable enhancement) [15, 16]. In our case, the lesion, well circumscribed on CT, was located in the right parietal bone. On T1 and T2 MRI sequences, the lesion was low-positive, with homogeneous contrast and enhancement of the adjacent dura mater, with no invasion of the soft tissues. These features suggested an intraosseous meningioma.

The optimal treatment is surgical excision, with a reported recurrence rate of 12.6% to 22% postsurgery [17]. The skull base meningiomas have a higher recurrence rate due to the challenges in achieving complete resection in these areas [17]. Cependant, Ueno *et al.* ont signalé un cas de méningiome transitionnel (WHO grade I) convexity PIOM qui est réapparu sept ans après l'intervention chirurgicale [18]. Chen *et al.* ont rapporté que même les PIOM histologiquement bénins mais non résécables peuvent présenter des changements malins au fil du temps [19]. Adjuvant therapy is recommended for unresectable or partially resectable symptomatic tumors or those with malignant or atypical histology. Treatment options may include radiation therapy such as external beam or gamma knife surgery, as well as chemotherapy and bisphosphonate therapy [20, 21].

Limited long-term follow-up information is available for PIM due to their uncommon nature. Therefore, regular imaging surveillance and close monitoring of patients are crucial to evaluate the extent of resection, early detection of recurrence or malignant progression, and timely intervention [22]. In cases of suspected meningiomas or those classified as WHO grade 1, annual MRI scans are recommended for 5 years. Following this period, the frequency of the scans may be increased to every 2 years [23] and continued monitoring for any neurological changes or tumor regrowth is recommended.

The diagnosis of primary intraosseous meningiomas (PIOM) can be difficult, due to their often insidious evolution and rarity. Medical imaging is essential to detect them, but only anatomopathological examination can confirm their nature. This rarity means that PIOMs should be systematically included in the differential diagnosis of any suspected bony lesion of the skull. Prompt management depends on accurate diagnosis, which calls for close collaboration between the various specialists involved, notably radiologists, neurosurgeons and pathologists. Finally, long-term clinical and radiological follow-up is essential, whatever the histological results or degree of resection obtained, to detect any recurrences [24].

4. Conclusion

Intraosseous meningiomas, although rare, represent a distinct clinical entity that requires special attention in the diagnosis of bone tumors, and must be included in the differential diagnosis of bone lesions of the skull. Medical imaging is crucial, but only anatomopathology can confirm the diagnosis. Prompt management with long-term clinical and radiological follow-up is essential to detect recurrence.

Funding

None.

Ethical Approval

None.

Consent

The patient's consent has been granted. We carried out this case report with respect for patient anonymity and confidentiality of information.

Author Contributions

Kajeou Meriem conceived the study, supervised data collection, analyzed the results, and drafted the manuscript. Lyna Benhaddou participated in data collection and data analysis. Yasser Arkha contributed to data interpretation and critical revision of the manuscript.

Registration of Research Studies

This study is a case report and, as such, is not required to be registered in a publicly accessible database according to current guidelines.

Guarantor

The guarantor of this study is Kajeou Meriem, who takes full responsibility for the work, had full access to the data, and holds final authority over the decision to publish.

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