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Primary Intraosseous Meningioma of the Spheno-Orbital Region: Value of CT and MRI in Preoperative Diagnosis

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Abstract **Case Report**

Primary intraosseous meningiomas are rare tumors belonging to the group of extradural meningiomas. Their clinical presentation is often nonspecific, dominated by progressive cranial swelling and, in the spheno-orbital region, by exophthalmos. We report the case of a 60-year-old patient with a right fronto-temporo-spheno-orbital mass and ipsilateral exophthalmos that had progressed slowly over five years. Computed tomography (CT) showed an expansile sclerotic bone lesion centered on the right sphenoid, associated with enhancing meningeal thickening extending to the orbit, the optic canal and the cavernous sinus; magnetic resonance imaging (MRI) completed the extension work-up. The patient underwent surgical resection followed by cranioplasty, and histopathological examination confirmed a meningioma. The outcome was favorable, with no recurrence after 6 months of follow-up. Intraosseous meningiomas can be mistaken for other bone lesions of the skull, notably fibrous dysplasia, sclerotic metastases and en-plaque meningioma with hyperostosis. Combined analysis of CT and MRI is essential for preoperative diagnosis and surgical planning, whereas histopathological examination remains the reference for diagnostic confirmation.

Keywords: Primary intraosseous meningioma, extradural meningioma, spheno-orbital region, exophthalmos, hyperostosis, computed tomography, magnetic resonance imaging.

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INTRODUCTION

Primary intraosseous meningiomas are rare tumors belonging to the group of primary extradural meningiomas, which account for 1–2% of all meningiomas, intraosseous forms making up approximately two-thirds of them [1]. They develop within the bone of the cranial vault or skull base, without an obvious dural origin in most cases. Their pathophysiology remains incompletely understood.

The clinical presentation is often insidious and nonspecific. Patients generally consult for a progressively enlarging cranial swelling, sometimes associated with headaches, local pain or, more rarely, neurological deficits depending on the location and extension of the lesion. Because of their rarity and variable radiological appearance, intraosseous meningiomas can be mistaken for other bone lesions of the skull, such as metastases, osteomas, fibrous dysplasia or hemangiomas, which sometimes makes preoperative diagnosis difficult.

Imaging plays an essential role in the evaluation of these tumors. Computed tomography (CT) allows analysis of bone changes, whether sclerotic, osteolytic or mixed, whereas magnetic resonance imaging (MRI) defines extension to the soft tissues, the dura mater and the intracranial structures. However, the definitive diagnosis rests on histopathological examination after surgical resection.

Treatment relies mainly on complete surgical resection when feasible, combined, if necessary, with cranial reconstruction. The prognosis is generally favorable for benign forms after complete resection, although recurrences may occur in cases of incomplete resection or aggressive histological variants.

Through this case, we wish to highlight the clinical, radiological and histopathological features of intraosseous meningiomas, as well as the importance of multimodal imaging in their early diagnosis and therapeutic management.

Case Presentation

A 60-year-old patient with no particular medical history was admitted for investigation of a right fronto-temporo-spheno-orbital mass associated with ipsilateral exophthalmos that had developed progressively over five years, without any neurological deficit. Clinical examination revealed non-pulsatile right exophthalmos and an ipsilateral fronto-temporal swelling.

A brain and orbital CT without and then with contrast injection revealed a relatively homogeneous, expansile, sclerotic lesional process centered on the greater wing and body of the right sphenoid, extending upward to the frontal squama and downward to the squamous part of the ipsilateral temporal bone. The lesion, with irregular contours in places, did not enhance after contrast injection and measured approximately 24 × 58 × 50 mm (anteroposterior × transverse × craniocaudal diameters) (Figure 1).



FIGURE 1: CT, bone window: expansile sclerotic lesion of the right sphenoid, extending to the frontal squama and the ipsilateral temporal bone

It also showed adjacent fronto-temporal meningeal thickening, spontaneously isodense, with intense and homogeneous enhancement after injection, with a maximal thickness of approximately 10 mm at the right temporal pole (Figure 2). This thickening extended into the orbit, infiltrating the intra- and extraconal fat as well as the lateral rectus and superior oblique muscles. It came into contact with the ipsilateral optic nerve, which it displaced, while insinuating itself into the optic canal (Figure 3). This invasion was responsible for grade III exophthalmos (Figure 4).

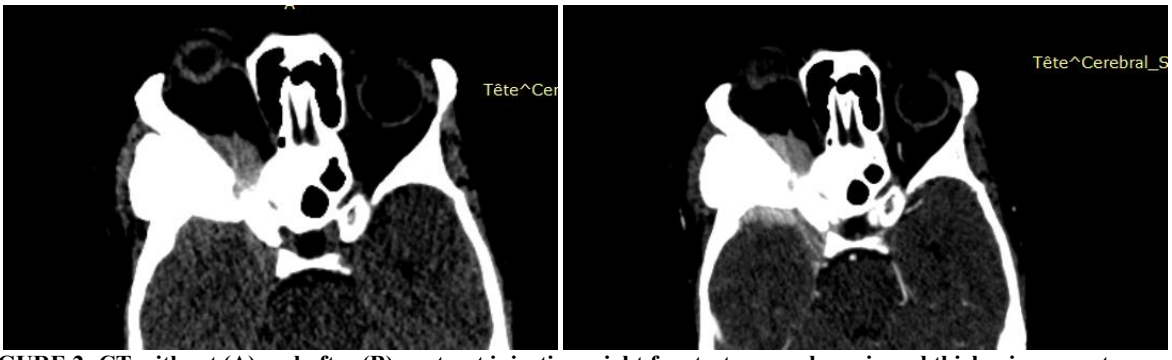


FIGURE 2: CT without (A) and after (B) contrast injection: right fronto-temporal meningeal thickening, spontaneously isodense, with intense and homogeneous enhancement

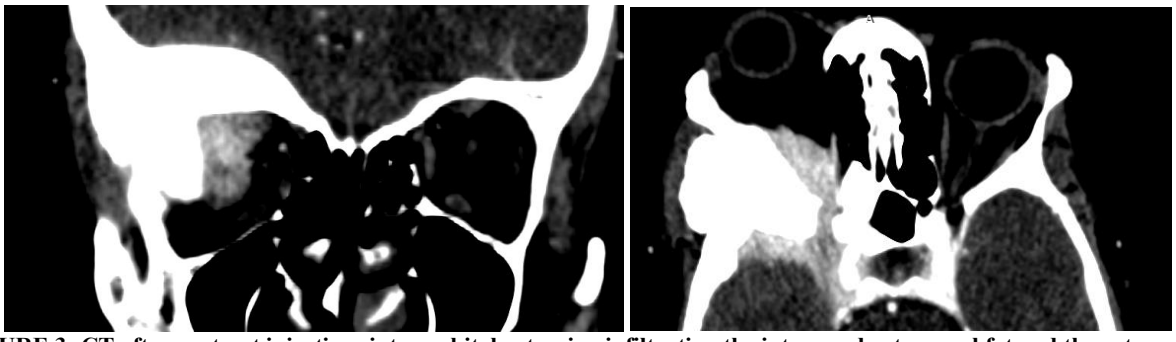


FIGURE 3: CT after contrast injection: intra-orbital extension infiltrating the intra- and extraconal fat and the extraocular muscles (lateral rectus and superior oblique), with displacement of the ipsilateral optic nerve and extension into the optic canal

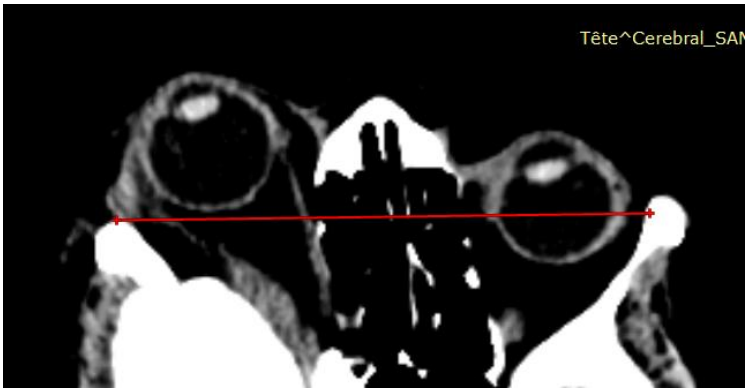


FIGURE 4: CT, axial section: grade III right exophthalmos, the globe lying entirely anterior to the external bicanthal line (line joining the lateral orbital rims)

Medial infiltration of the right cavernous sinus was also noted, which remained well opacified (Figure 5). The lesional process filled the inferior orbital fissure and encased the right superior orbital fissure, pterygopalatine fossa and foramen rotundum (Figure 6).

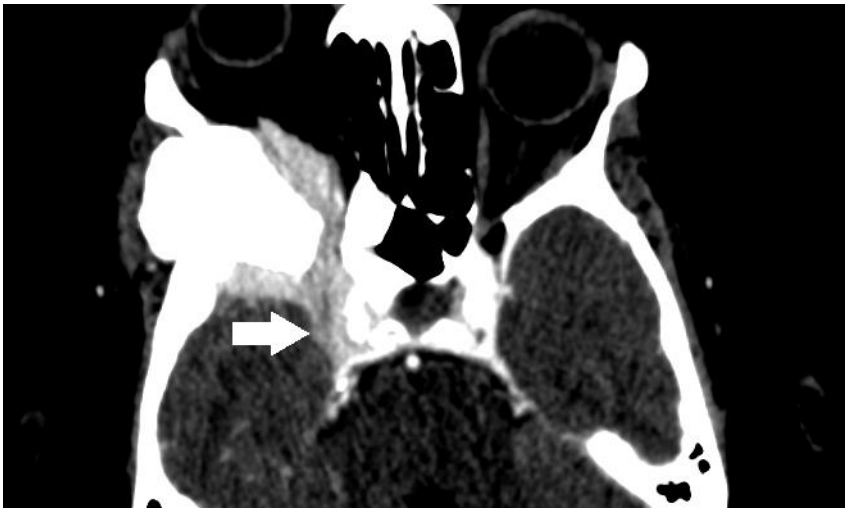


FIGURE 5: CT after contrast injection: medial infiltration of the right cavernous sinus, which remains well opacified

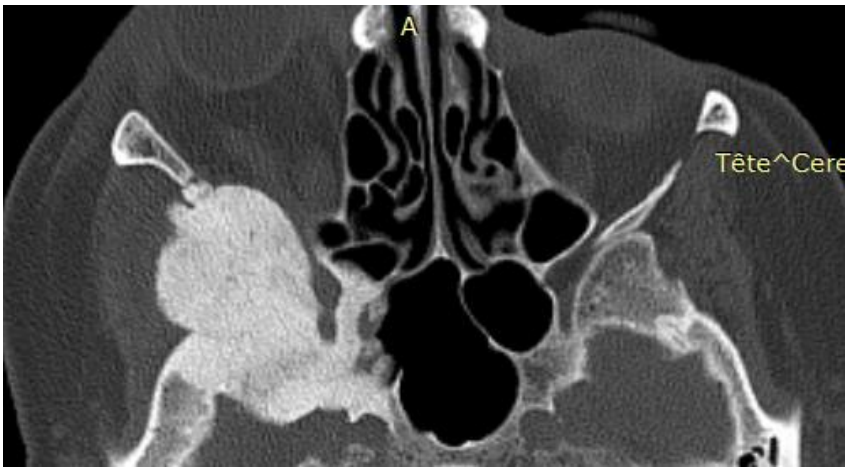


FIGURE 6: CT, bone window: filling of the inferior orbital fissure, with extension to the right superior orbital fissure, pterygopalatine fossa and foramen rotundum

Brain and orbital MRI was then performed. The bone component appeared hypointense on T1 and of heterogeneous intermediate signal on FLAIR, without enhancement on fat-suppressed T1 (T1 FS) after gadolinium injection, and without restricted diffusion (no hyperintensity on diffusion-weighted imaging and no decrease in ADC) (Figures 7, 8). No associated intracranial parenchymal lesion was noted.

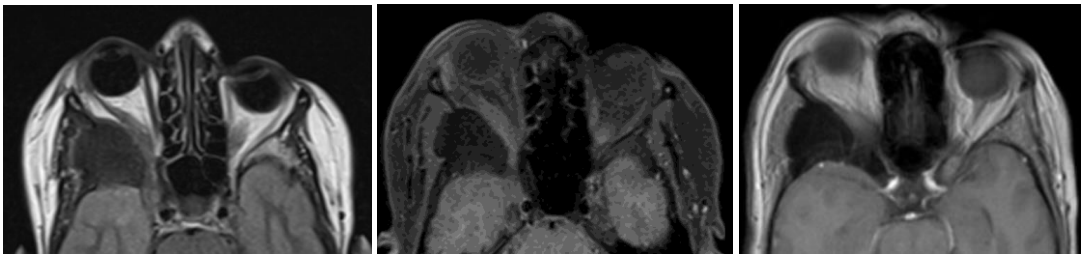


FIGURE 7: Brain and orbital MRI: (A) T1: diffuse hypointensity of the bone component; (B) T1 FS after gadolinium injection: no definite enhancement of the bone component; (C) FLAIR: heterogeneous intermediate signal

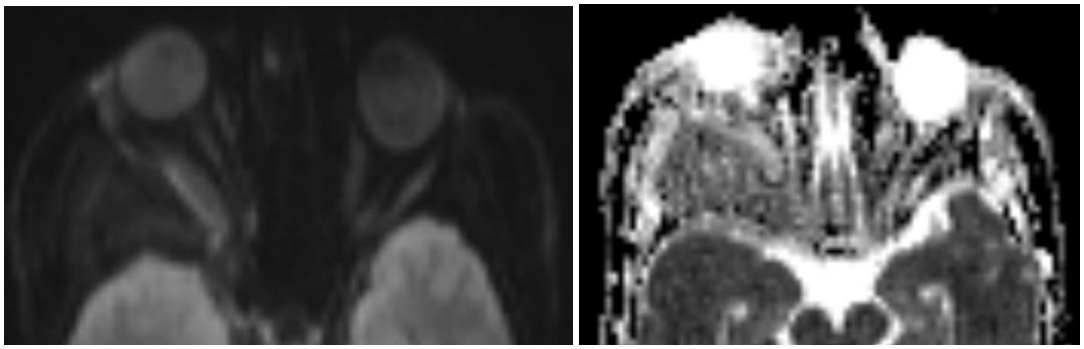


FIGURE 8: Diffusion-weighted imaging (A) and ADC map (B): no restricted diffusion within the lesion

Given the radioclinical suspicion of a primary intraosseous spheno-orbital meningioma, surgical resection was performed with cranioplasty during the same operation.

Histopathological examination revealed a meningothelial tumor proliferation made up of spindle and polygonal cells arranged in sheets and whorls within a fibrous stroma. Calcified psammoma bodies were present in places, with no marked cytonuclear atypia or significant mitotic activity. Immunohistochemical study showed strong positivity of the tumor cells for EMA (epithelial membrane antigen) and vimentin, with focal positivity for the progesterone receptor (PR). S100 protein staining was weak or focal, whereas GFAP expression was negative. The Ki-67 proliferation index was low (< 5%). Together, these findings led to the diagnosis of a grade 1 spheno-orbital intraosseous meningioma according to the 2021 WHO classification [7].

DISCUSSION

Primary intraosseous meningioma is a rare form of extradural meningioma, a group that accounts for 1–2% of all meningiomas [1]. It develops preferentially in the cranial vault and the spheno-orbital region. Its course is generally slow, and its clinical presentation depends on its location and its extension to adjacent structures. The most frequent manifestations include progressive craniofacial deformity, exophthalmos and visual disturbances related in particular to optic nerve compression [1-3].

Lang *et al.,* proposed a classification of primary extradural meningiomas according to their location: type I, purely extracalvarial; type II, purely calvarial (intraosseous); type III, calvarial with extracalvarial extension. Types II and III are subdivided according to their location at the convexity (C) or the skull base (B) [1].

Radiologically, CT is the reference examination for the analysis of bone lesions. It allows characterization of hyperostosis, assessment of bone expansion and delineation of extension to skull-base structures, notably the foramina. Intraosseous meningiomas usually present as a sclerotic, sometimes expansile lesion, reflecting tumor infiltration of the bone. In spheno-orbital locations, this bone involvement may predominate and constitute the main radiological sign, or even the mode of presentation of the disease [3,4].

MRI completes the work-up by defining extension to the intracranial and orbital structures. It notably allows study of the associated dural thickening, meningeal enhancement after gadolinium injection, intra- and extraconal extension, and the relationships of the lesion with the extraocular muscles, the optic nerve and its canal. This complementarity between CT, which is particularly effective for bone analysis, and MRI, which better characterizes soft-tissue extension, is essential for establishing the lesional work-up and planning therapeutic management [3,5].

In our case, the combination of an expansile sclerotic bone lesion involving the greater wing and body of the sphenoid, intensely enhancing adjacent meningeal thickening and orbital extension with involvement of the optic canal was highly suggestive of a spheno-orbital intraosseous meningioma. The absence of enhancement of the bone component is not a sufficient argument to rule out this diagnosis. Indeed, enhancement may

predominate in the adjacent dura mater and soft tissues, whereas the bone component appears mainly as hyperostosis. This dissociation of radiological appearances underlines the importance of a combined analysis of CT and MRI data [2,4].

The main differential diagnosis is en-plaque meningioma of dural origin, with reactive hyperostosis or bone invasion, which is frequent in the spheno-orbital region. The distinction rests on the predominance of the bone component over the dural component and, above all, on operative and histological findings, which establish the intraosseous origin of the tumor and the secondary nature of any dural involvement [1,3]. Fibrous dysplasia is usually characterized by a ground-glass bone matrix and a gradual transition with the adjacent bone, without tumoral meningeal thickening. Other lesions should also be considered, notably sclerotic metastases, osteoma, Paget's disease, intraosseous hemangioma and certain primary bone tumors [3,4].

The origin of primary intraosseous meningiomas remains incompletely understood. Several pathophysiological hypotheses have been proposed, notably the inclusion of arachnoid cells within the cranial sutures during embryonic development, their implantation into bone following trauma, and the differentiation of pluripotent mesenchymal cells into meningothelial cells. These mechanisms could explain the occurrence of meningiomas outside their usual intradural location [1,2,6].

Therapeutic management is essentially surgical, aiming for resection that is as complete as possible while preserving neurovascular structures and visual function. However, the spheno-orbital location sometimes makes complete resection difficult, particularly in cases of extension to the optic canal, the orbital apex or the cavernous sinus. The operative strategy must therefore take into account the extent of the lesion, its anatomical relationships and the functional risk. Radiotherapy may be discussed in certain situations, notably in cases of progressive residual tumor, incomplete resection or aggressive histological features, depending on the grade and the clinical context [5,6].

The prognosis depends mainly on the histological grade, the extent of bone and adjacent tissue involvement, and the possibility of achieving durable tumor control. Although intraosseous meningiomas are often low-grade, their spheno-orbital location can cause significant functional morbidity, particularly because of involvement of the optic nerve and orbital structures. Prolonged clinical and radiological follow-up is therefore necessary to detect any tumor progression or recurrence.

CONCLUSION

Primary spheno-orbital intraosseous meningioma is a rare, slowly progressive entity whose clinical presentation is most often dominated by progressive exophthalmos, sometimes associated with visual impairment, reflecting a frequently late diagnosis. Imaging plays a fundamental role in diagnostic orientation: CT allows precise analysis of hyperostosis and bone extension, whereas contrast-enhanced MRI defines dural, orbital and canalicular involvement as well as the relationships with the cavernous sinus. The combination of an expansile spheno-orbital lesion with a hyperostotic component and enhancing meningeal thickening is highly suggestive of the diagnosis, which should be considered in any spheno-orbital hyperostosis associated with signs of meningeal or orbital involvement. Management is essentially surgical, guided by extension to the orbital apex, the optic canal and the skull-base foramina, with the aim of the most complete resection possible while preserving visual function. The definitive diagnosis rests on histopathological examination.

ADDITIONAL INFORMATION

Conflict of interest: The authors declare that they have no conflicts of interest.

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