

# A Case of Intraosseous Microcystic Meningioma Without a Mass Lesion

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## Abstract

Both intraosseous and microcystic meningiomas are rare tumor types. We report the case of a 66-year-old woman with intraosseous microcystic meningioma without a mass lesion. She presented with a rare intraosseous microcystic meningioma manifesting as pain. Radiological examination revealed an osteolytic lesion in the right parietal bone. Magnetic resonance (MR) images showed iso- to hypointensity on T<sub>1</sub>-weighted images and hyperintensity on T<sub>2</sub>-weighted images corresponding to the lesion. T<sub>1</sub>-weighted MR imaging with gadolinium enhancement better defined the marginal area. The inner table of the skull was disrupted prominently, and both sides of the outer table were eroded. There was fluid leakage during surgery but no obvious tumor mass. Histological examination revealed microcystic meningioma in the inner part of the defective bone. A macroscopic lesion was not found, because most of the tumor comprised microcysts, and their contents leaked out during the surgical procedure. Intraosseous microcystic meningioma may be considered as one of the differential diagnoses when the intraosseous tumor in the skull has fluid leakage and does not have a mass lesion during the surgery.

Key words: fluid leakage, intraosseous meningioma, microcystic meningioma, without a mass lesion

## Introduction

Primary extradural meningiomas are rare lesions, accounting for less than 2% of all meningiomas.<sup>13)</sup> Intraosseous meningiomas are increasingly rarer, occurring in 14% of patients with ectopic or primary extradural meningiomas.<sup>6)</sup> Microcystic meningioma is also rare, accounting for only 1.6% of intracranial meningiomas.<sup>11,12)</sup> It was originally described as a humid tumor because of its gross appearance.<sup>15)</sup> Here, we report a very rare case of a patient with intraosseous microcystic meningioma without a mass lesion.

## Case Report

A 66-year-old woman was admitted to our hospital because of pain in the right parieto-occipital region for 2 years. She had no previous history of trauma to that region. Neurological examination and laboratory data were normal. Skull radiography and bone window computed tomography (CT) scans revealed an osteolytic lesion on the right parietal bone (Figs. 1, 2).

Magnetic resonance (MR) images revealed that the intraosseous lesion was iso- to hypointense in T<sub>1</sub>-weighted imaging (Fig. 3A) and hyperintense in T<sub>2</sub>-weighted imaging (Fig. 3B). T<sub>1</sub>-weighted MR imaging with gadolinium enhancement showed prominent enhancement of the margin of the lesion (Fig. 3C).

After reflecting the scalp, a slight erosion was noticed in the outer tables of the right parietal bone (Fig. 4A), and fluid leakage was expelled. A right parietal craniotomy was performed. Neither an association of the lesion with cranial sutures nor any obvious tumor mass in the skull lesion was observed. The inner table of the skull was disrupted prominently (Fig. 4B). A small defect was found in the underlying dura mater, whereas the arachnoid mater was preserved (Fig. 4C). After the dura surrounding the lesion was removed, the dural defect was patched with periosteum. The bone defect was repaired by cranioplasty using titanium mesh. Histological examination of the specimen revealed microcystic meningioma in the inner part of the bone lesion and defective dura mater (Fig. 5A). The tumor had a myxoid appearance, was loosely textured and hypocellular, with widespread formation of microcysts (Fig. 5B). The MIB-1 staining index of the meningioma was 2.5%, suggesting low proliferative

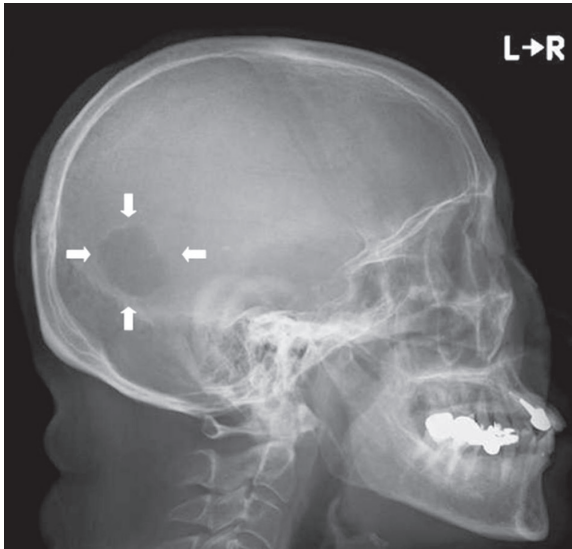


Fig. 1 Lateral skull radiograph revealing an osteolytic lesion in the parietal bone (arrows). L: left, R: right.

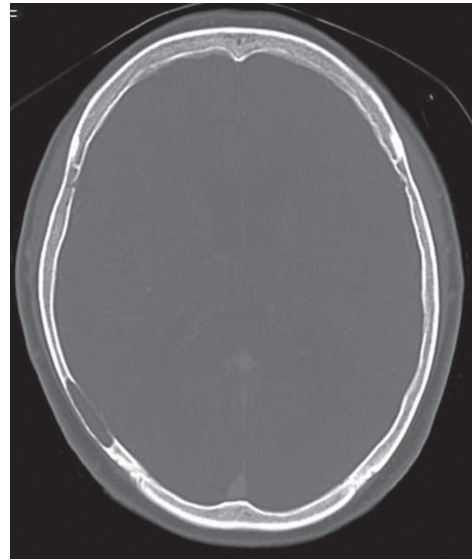


Fig. 2 Bone window computed tomography (CT) scan depicting erosion on the inner table of the skull.

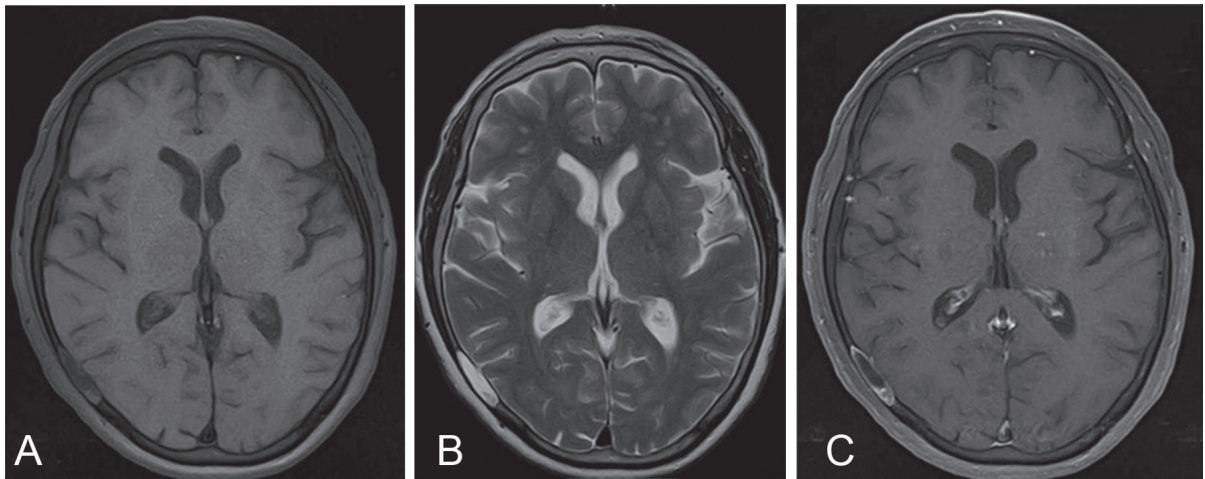


Fig. 3 A: T<sub>1</sub>-weighted magnetic resonance (MR) images showing iso- to hypointense lesion. B: T<sub>2</sub>-weighted MR images showing a hyperintense lesion. C: T<sub>1</sub>-weighted MR images with gadolinium showing enhancement of the margin of the lesion.

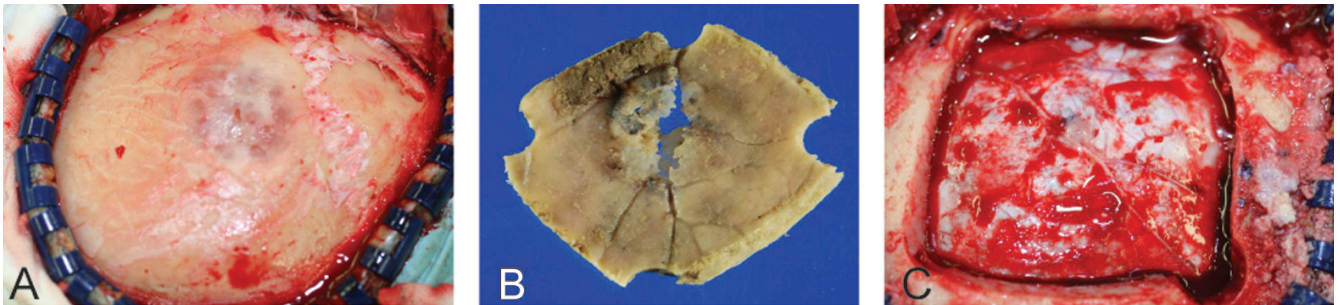
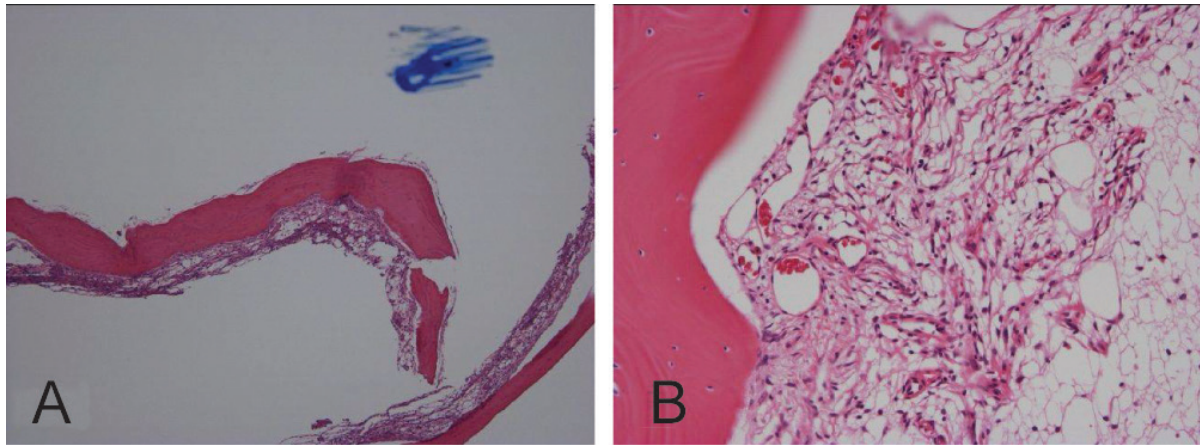


Fig. 4 Intraoperative findings. A: Erosion of the outer tables is observed and watery fluid was expelled. B: The inner table of the skull is disrupted, and both sides of the outer table are eroded. Tumor mass is not observed. C: Dura mater has a visible defect, whereas the arachnoid mater is preserved.



**Fig. 5** A: Photomicrograph showing meningeoma cells in the inner part of the bone defect. Hematoxylin and eosin stain (original magnification, 40 $\times$ ). B: Variably sized cystic spaces and vacuolated cytoplasm. Hematoxylin and eosin stain (original magnification, 200 $\times$ ).

potential. Immunohistochemical staining revealed a positive reaction to vimentin and epithelial membrane antigen. The postoperative course was uneventful. We concluded that the tumor had mainly occupied the parietal bone and extended to the dura mater. The fluid leakage had been edematous fluid from the microcysts and not the cerebrospinal fluid.

### Discussion

According to the literature, 68% of primary extradural meningiomas involve the calvaria.<sup>13)</sup>

The frontoparietal and orbital regions are the most common locations for intraosseous meningiomas. Other sites reported in the literature include the subcutaneous tissue of the skin, the paranasal sinuses, nasal cavities, oral cavities, parapharyngeal space, neck, salivary glands, and areas along the perineural sheath of the cranial nerves. Rare meningiomas occurring in the lung, mediastinum, adrenal gland, paraspinal region, and even in the finger, have also been reported.<sup>13,20)</sup> Primary extradural meningiomas are classified as purely extracalvarial (type I), purely calvarial (type II), or calvarial with extracalvarial extension (type III). Considering the site of the tumor, Lang et al. further subdivided type II and type III lesions into convexity (C) or skull-base (B) forms.<sup>13,20)</sup> The case presented here is a type IIC meningioma. Extradural meningiomas, including intraosseous meningiomas, are reported to occur with the same frequency in both male and female individuals, or with a slight female predominance, unlike the intradural meningiomas that occur twice as frequently in women as in men.<sup>20)</sup> In general, the meninx are derived from mesenchymal cells. Extradural meningiomas could arise in numerous unusual locations

as a result of aberrant differentiation or localization of multipotent mesenchymal stem cells.<sup>7,14,19,22)</sup> Alternatively, extradural tumors may originate from cells that are misplaced after differentiation into meningocytes or arachnoid cap cells. Arachnoidal cells from blood vessels or nerves traversing the skull may explain the origin of some intraosseous meningiomas.<sup>10)</sup> Cellular dedifferentiation within the skull could also possibly explain the formation of an intraosseous meningioma.<sup>4)</sup> The nomenclature of primary extradural meningiomas with dural invasion is still controversial, because these tumors are difficult to classify definitively, depending on the radiographic appearance and surgical findings.<sup>13)</sup> Some authors state that the localization of the mean tumor mass within the skull allows determination of the site of origin of the tumor and the designation of an intraosseous meningioma or a primary extradural meningioma, even in the presence of dural invasion.<sup>1)</sup> Other authors maintain that dural invasion precludes a diagnosis of intraosseous meningioma.<sup>2)</sup>

Yamazaki et al. reviewed 48 cases in the literature and found that the majority (30 cases, 62.5%) were of the meningothelial type; transitional (12 cases, 25%) cases were the next most common, followed by fibroblastic (4 cases, 8.3%), psammomatous (1 case, 2.1%), and malignant (1 case, 2.1%).<sup>21)</sup> Recent studies indicate that intraosseous meningiomas have a higher incidence of malignant features than intradural meningiomas do.<sup>9,18)</sup> Radiographic evidence of hyperostosis is noted in 59% of cases, whereas osteolytic changes in the surrounding bone are noted in 32% of cases. Six percent of the cases are of a mixed composition of both osteolysis and hyperostosis.<sup>5)</sup> The differential diagnosis for osteolytic, intraosseous meningioma includes metastatic cancer, plasmacytoma, giant cell tumor, hemangioma, epidermoid cyst, osteogenic

sarcoma, eosinophilic granuloma, aneurysmal bone cyst, and fibrous dysplasia.<sup>20)</sup>

Microcystic meningiomas have a similar location, clinical features, and prognosis to common benign meningiomas.<sup>8)</sup> The characteristic feature of microcystic meningiomas is cyst formation.<sup>8)</sup> The pathological mechanisms of microcyst formation include pia-arachnoid differentiation, secretory activity of the tumor cells, certain degenerative processes, and arachnoid trabecular cell origin.<sup>8)</sup> Characteristic MR images findings suggestive of microcystic meningiomas are hypointensity in T<sub>1</sub>-weighted imaging and hyperintensity in T<sub>2</sub>-weighted imaging.<sup>17)</sup> In particular, obvious hypointensity on T<sub>1</sub>-weighted images is the most valuable for the diagnosis of microcystic meningiomas.<sup>3)</sup> These features coincide with this case.

In this case, prominent enhancement of the marginal area of the lesion was revealed on MR images. We assume that hypointense region of the central area on T<sub>1</sub>-weighted imaging reflects the fluid leakage from the tumor, and the hyperintense enhancement of the marginal area with gadolinium represents the tumor body.

Although a case of intraosseous microcystic meningioma has been reported, this tumor had a well defined and soft tumor, did not have cystic change and was located within the skull without any evidence of extraosseous extension.<sup>16)</sup> Our case is the first report of intraosseous microcystic meningioma case without a mass lesion and with fluid leakage. A macroscopic lesion was inapparent in the preoperative period, because most of the tumor was composed of microcysts and the fluid leakage contained in the tumor was expelled during the surgery. The tumor component was observed on the inner portion of the bone defect.

In conclusion, the case presented here suggests that, however rare, intraosseous microcystic meningioma may be considered as one of the differential diagnoses when the intraosseous tumor in the skull has fluid leakage and does not have a mass lesion during the surgery.

### Conflicts of Interest Disclosure

None.

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