



## Case Report

# Calvarial angiomatous meningioma developed in the diploe

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## ABSTRACT

**Background:** Angiomatous meningioma is a rare subtype of meningiomas. To the best of our knowledge, there have been no reports of intradiploic angiomatous meningioma.

**Case Description:** A 53-year-old previously healthy woman was diagnosed with a calvarial lesion during a brain checkup. Cerebral magnetic resonance imaging showed an intradiploic tumor, 11 × 14 × 12 mm, in the right parietal bone. It was an enhancing, lobular tumor presenting as isointensity on T1- and hyperintensity on T2-weighted sequences, with an intense enhancement of the adjacent dura mater. Computed tomography revealed bone erosion at the tumor site, extending predominantly into the inner side, and sclerotic changes in the surrounding bone. Total resection was performed. Microscopically, the tumor tissue comprised cells with low-grade meningioma and intervening prominent vasculatures, consistent with angiomatous meningioma.

**Conclusion:** Angiomatous meningioma should be considered as a differential diagnosis when an intradiploic tumor shows a lobular structure, intense enhancement of the adjacent dura mater, and sclerotic changes in the surrounding skull. These findings can support prompt tumor resection.

**Keywords:** Angiomatous meningioma, Diploe, Lobular structure, Osteosclerosis

## INTRODUCTION

Meningiomas are the most common primary brain tumors in adulthood. They commonly grow in subdural sites, whereas approximately 1% of them are estimated to arise in extradural sites involving the calvarium or diploe.<sup>[8,13]</sup> The diploe can be affected by various pathologies, such as bone cysts, encephaloceles, arachnoid cysts, lipomas, teratomas, dermoid, epidermoid, cavernous hemangiomas, metastatic tumors, neurofibromas, and meningiomas.<sup>[1-7,11-24,26]</sup> Intradiploic meningioma is an infrequent but distinct entity that commonly develops as a benign tumor.<sup>[1,2,4-7,10,12,16-18,20,21,26]</sup> Angiomatous meningioma is a rare subtype of the World Health Organization Grade I meningioma, characterized by tumor cells consistent with low-grade meningioma and prominent microvessels of varying sizes.<sup>[9,25]</sup> To the best of our knowledge, there have been no reports documenting intradiploic angiomatous meningiomas.

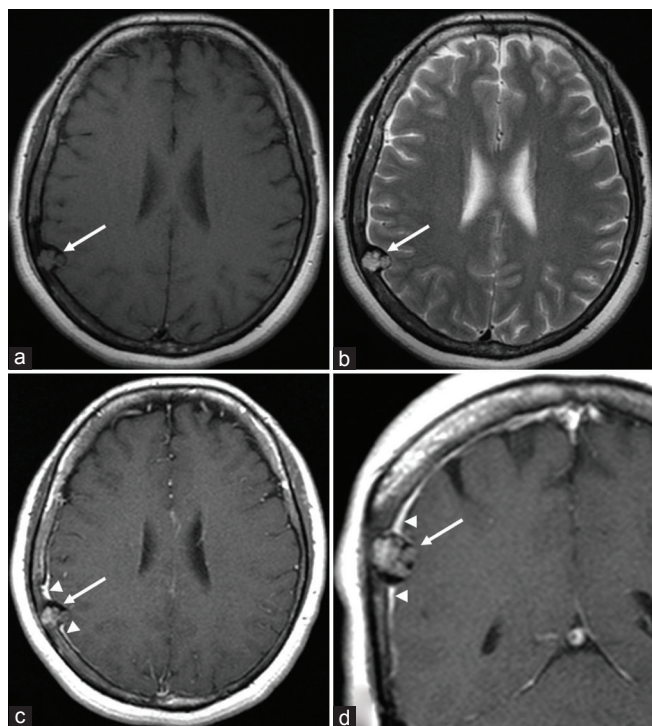
Here, we present a unique case of intradiploic angiomatous meningioma with distinctive radiological and intraoperative findings.

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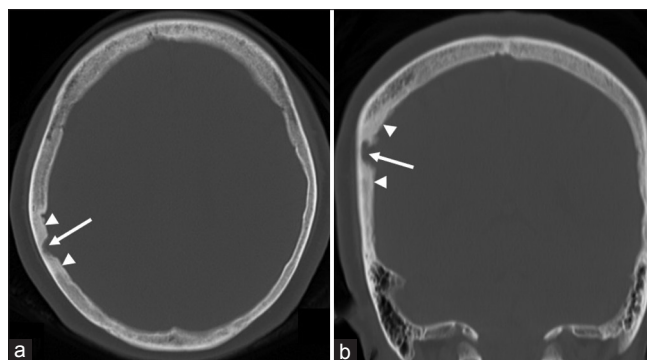
## CASE PRESENTATION

A 53-year-old previously healthy woman was diagnosed with a calvarial lesion on a brain checkup and was referred to the hospital. She had not undergone cranial radiotherapy in her life. Cerebral magnetic resonance imaging (MRI) revealed an intradiploic tumor in the right parietal bone protruding into the cranial cavity. It was a lobular mass  $11 \times 14 \times 12$  mm, presented isointensity on T1- and hyperintensity on T2-weighted sequences, and was inhomogeneously enhanced with intense enhancement of the surrounding dura mater. The inner table and dura underlying the tumor appeared intact. There was no identifiable peritumoral brain edema [Figure 1]. On computed tomography (CT), the tumor was accompanied by a well-demarcated calvarial erosion at the site of the tumor, extending more predominantly into the inner side compared to the outer side. In addition, sclerotic changes were observed in the surrounding bone [Figure 2]. The patient was requested to undergo a tumor resection. Intraoperatively, the tumor was reflected with the surrounding bone that presented sclerotic changes and

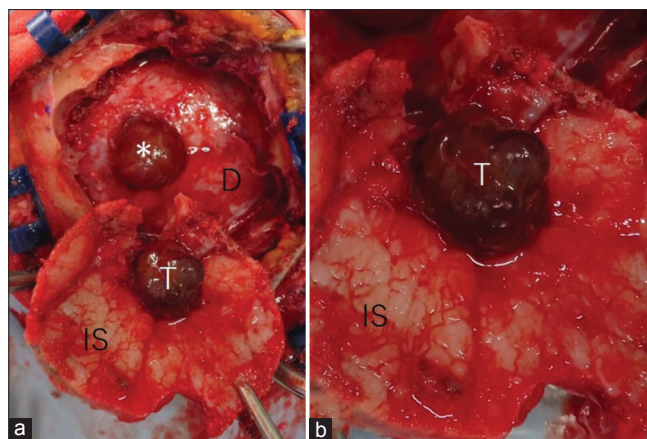


**Figure 1:** Axial T1- (a), T2- (b), and postcontrast axial (c) and coronal (d) T1-weighted magnetic resonance images show the intradiploic mass in the right parietal bone, protruding into the cranial cavity (arrow). It involves a lobular mass  $11 \times 14 \times 12$  mm in diameter, presenting isointensity on T1- and hyperintensity on T2-weighted sequences, respectively, and is inhomogeneously enhanced with an intense enhancement of the surrounding dura mater (c and d, arrowheads). The inner table and dura underlying the tumor appear intact. There is no identifiable peritumoral brain edema (b).

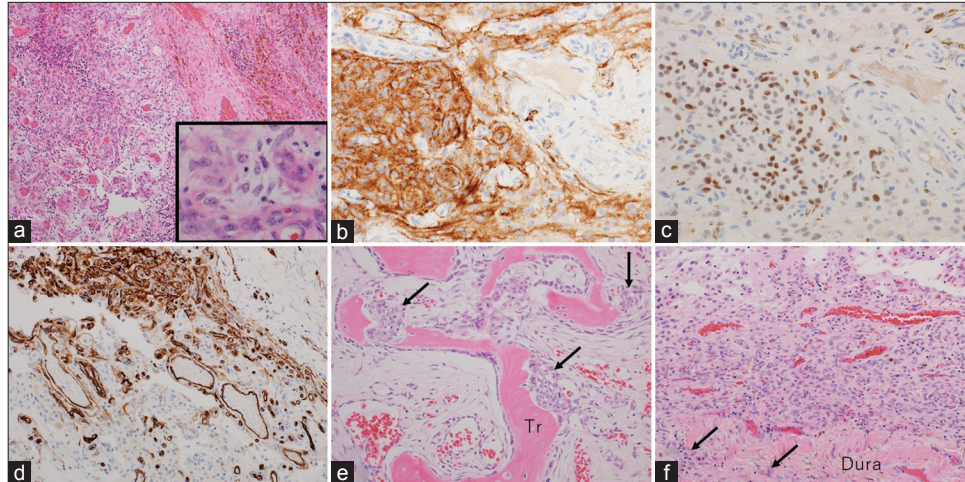
removed *en bloc*. The inner table compressed by the tumor was partially defective and the underlying dura mater was erosive, which was circumferentially resected and replaced by an artificial substitute [Figure 3]. Eventually, a Simpson Grade I resection was achieved. Adhesions between the tumor and surrounding dura mater were not found in the intact underlying cerebral cortex. Microscopically, the tumor comprised cells with oval-shaped nuclei and intervening vasculature of varying sizes. There were a few mitotic figures. Immunohistochemical examination showed positive staining for epithelial membrane antigen and progesterone receptor but negative staining for CD34. These findings are consistent with those of angiomatous meningiomas. In addition, tumor invasion into the adjacent bone and dura mater was observed [Figure 4]. At present, the patient is planning to undergo periodic MRI surveillance in every 6 months.



**Figure 2:** Axial (a) and coronal (b) bone target computed tomography scans showing well-demarcated calvarial erosion extending more predominantly into the inner side (arrow), compared to that in the outer side, with osteosclerotic changes in the surrounding bone (arrowheads).



**Figure 3:** Intraoperative photos at the reflection of the bone flap (a) and its magnified view (b) showing the tumor (T) protruding from the inner table and depressed dura mater by the tumor (asterisk). D: dura mater; IS: inner surface of the skull.



**Figure 4:** Photomicrographs of the resected specimens showing tumor tissue comprising cells with oval-shaped nuclei and intervening vasculatures with varying sizes (a and its inset, magnified view). There are few mitotic figures. Immunohistochemical examination shows positive staining for epithelial membrane antigen (b) and progesterone receptor (c), while negative for CD34 (d). Tumor invasions into the adjacent bone (e, arrows) and dura mater (f, arrows) are noted. (a) Hematoxylin and eosin stain,  $\times 40$ ; (b) epithelial membrane antigen,  $\times 200$ ; (c) progesterone receptor,  $\times 200$ ; (d) CD34,  $\times 100$ ; and (e and f) hematoxylin and eosin stain,  $\times 100$ . Tr: trabecula.

## DISCUSSION

Although the diploe can be affected by varying pathologies, intradiploic meningioma has been rarely reported.<sup>[1-7,11-24,26]</sup> Furthermore, cerebral angiomatous meningioma is known to be a rare subtype of low-grade meningioma.<sup>[9,25]</sup> In the present patient, the intradiploic tumor showed a lobular structure on MRI, with intense enhancement of the surrounding dura mater. In addition, the inner table and dura underlying the tumor appeared intact. Therefore, we assumed that the tumor arose from the diploe, instead of the dura mater.

Whereas CT revealed sclerotic changes in the skull surrounding the tumor. Furthermore, the histological appearance of the resected tumor was consistent with that of an angiomatous meningioma. Therefore, we assumed that the findings identified on the MRI and CT might represent the characteristic appearance of intradiploic angiomatous meningioma.

The present case involved a small intradiploic tumor incidentally detected during a brain checkup. However, intraoperative observation revealed a defective inner table due to compression by the intradiploic tumor. Furthermore, histological examination showed overt tumor invasion into the surrounding bone and underlying dura mater. Therefore, prompt tumor resection is recommended if an asymptomatic intradiploic tumor is assumed to be an angiomatous meningioma.

In addition, the present intradiploic tumor showed unevenly inward extension into the cranial cavity, with the outer table intact. This may be because the inner table of the patient was less resistant to compression by the tumor than the outer

table. Further case studies would clarify the pros and cons of these speculations.

## CONCLUSION

Angiomatous meningioma should be considered as a differential diagnosis when an intradiploic tumor shows a lobular structure, intense enhancement of the adjacent dura mater, and sclerotic changes in the surrounding skull. These findings can support prompt tumor resection.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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