

## Case Report

# A Case of Calvarial Hemangioma in Cranioplasty Site

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It is not uncommon for hemangiomas to occur in the calvarium, accounting for about 10% of the benign skull tumors. A 46-year-old man was presented with a palpable scalp mass on the left parietal region. Past medical history indicated that he had undergone cranioplasty 25 years prior due to a depressed skull fracture suffered from a traffic accident. Magnetic resonance imaging revealed mixed signal intensity mass on T1 -and T2- weighted images pushing a linear signal void lesion outward in the left parietal region. After total surgical removal along with cranioplasty, he was discharged without neurological deficits. Histological examination of the surgical specimen revealed a cavernous hemangioma. A skull hemangioma occurring at the site of a cranioplasty has not yet been reported. Therefore, authors report this case in combination with a pertinent literature review.

**KEY WORDS :** Cavernous hemangioma · Calvarium · Cranioplasty.

## INTRODUCTION

Primary hemangioma in the calvarium is not uncommon benign tumors that accounts for 10% of all benign skull tumors<sup>6,12</sup>. It involves mostly the parietal or frontal bone<sup>10,20</sup> and less commonly, arises from the craniofacial bone, including the zygoma, maxilla, vomer, and mandible. The majority of hemangiomas are asymptomatic, but patients may complain of headache, scalp pain, and a palpable mass. Because of their infrequent occurrence, vague symptoms, and absence of prototypical radiologic findings, hemangiomas may be misdiagnosed as multiple myeloma or osteosarcoma in many cases. We present a case of a calvarial hemangioma, which occurred at the site of a previous cranioplasty site, together with a brief review of the literature.

## CASE REPORT

A 46-year-old, right-handed male presented with a round mass on the left parietal area for several months. The size was about 6 cm in diameter and he had no specific past

medical history except a motorcycle accident 25 years ago, for which he underwent cranioplasty for the treatment of a depressed skull fracture (Fig. 1). On admission, he was alert

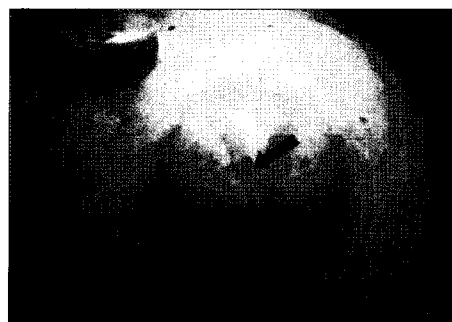


Fig 1. Preoperative photographs show a round mass on the left parietal region and a previous skin incision (arrow).

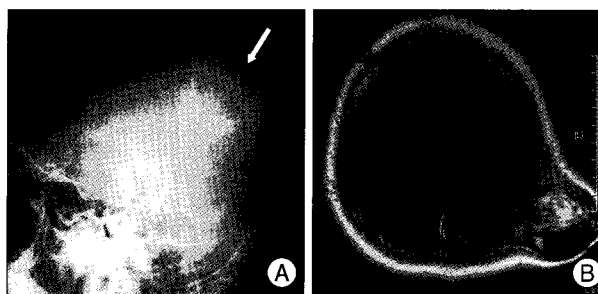
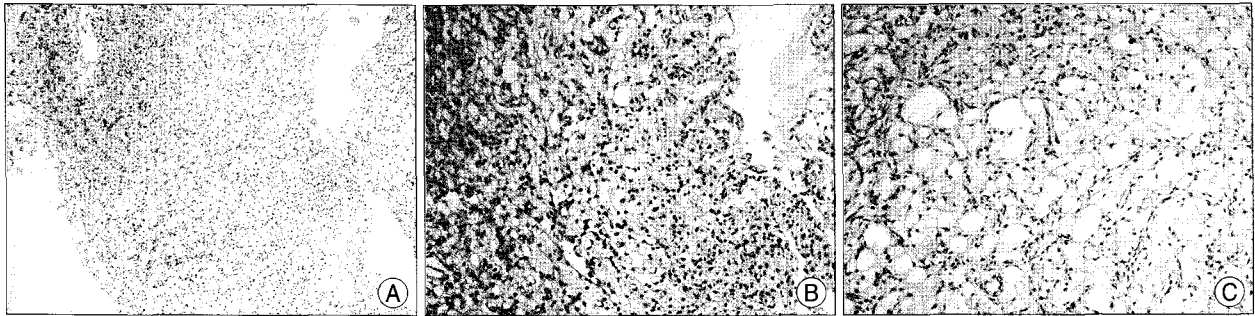


Fig 2. Preoperative radiological findings. A : Plain skull radiograph shows a radiolucent lesion in the left parietal bone. B : Preoperative magnetic resonance image shows a left parietal intradiploic round mass with heterogeneously enhancing signal intensity.

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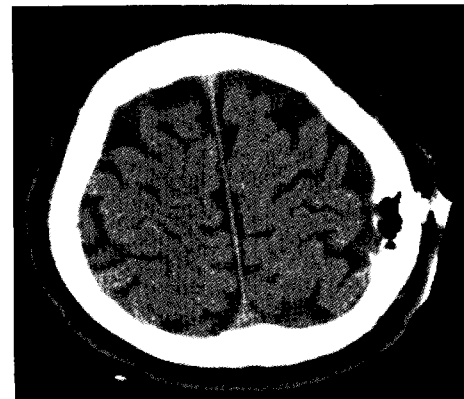


**Fig. 3.** A : Intraoperative photograph after removal of the mass and the resin plate. B : The removed resin plate. The size was  $3.8 \times 3$  cm. C : The mass including hematoma.



**Fig. 4.** Histopathologic pictures illustrating the proliferation of slit-like blood vessels lined by endothelial cells and filled with red blood cells (hematoxylin and eosin, A :  $\times 40$ , B :  $\times 100$ , C :  $\times 200$ ).

and well oriented. Physical examination revealed unremarkable findings except for the scalp mass. In addition, there were no abnormalities in the laboratory findings. Plain skull X-rays (Fig. 2A) revealed a radiolucent, circumscribed lesion in the left parietal region, and a brain computed tomography (CT) scan showed a large, partially enhancing mass with mixed osteolytic and sclerotic patterns with the contrast media. Brain magnetic resonance imaging (MRI) demonstrated a pushing linear signal void plate-like lesion, suggestive of previous cranioplasty material. This region was heterogeneously enhanced with contrast administration and results showed no dural involvement (Fig. 2B). Based on radiographic studies, differential diagnosis included interosseous meningioma, metastasis, multiple myeloma, hemangioma, and other rare skull tumors such as aneurysmal bone cyst, epidermoid, fibrous dysplasia, chondrosarcoma, osteosarcoma. During surgery, when the scalp was opened, we found the resin bone flap which had been elevated by the tumor. After removing the resin bone flap, a hemorrhagic mass was exposed and dissected along with the surrounding abnormal bone margin. The inner table of the bone and the dura appeared to be intact (Fig. 3A and B). We removed the mass successfully and performed cranioplasty on the bony defect site with bone cement. On gross pathologic examination, the soft mass including hemorrhagic contents measured  $4.8 \times 3.5 \times 2.2$  cm (Fig. 3C). Based on the microscopic findings of large blood vessels, the



**Fig. 5.** Postoperative computed tomography scan shows complete resection of the mass.

pathology was diagnosed as a cavernous hemangioma (Fig. 4). After surgery, the patient was alert without neurological deficits. CT scans obtained immediately after surgery confirmed complete resection of the mass (Fig. 5).

## DISCUSSION

Cavernous hemangiomas of the cranium are benign tumors accounting for 10% of benign skull tumors. Hemangiomas most commonly affect the vertebral column, followed by the skull<sup>6,13</sup>, especially in the parietal and frontal bones<sup>13,17</sup>. These tumors were found to occur more frequently in female patients, and are most commonly

diagnosed in the middle decades of life, with a peak age around the fourth decade<sup>10,17,22</sup>. Hemangiomas are histopathologically classified as cavernous and capillary<sup>4,10,13</sup>. Cavernous hemangiomas are composed of a group of large, dilated blood vessels separated by fibrous tissue, whereas capillary hemangiomas lack fibrous septa and have smaller vascular lumens<sup>10</sup>. Most calvarial hemangiomas are of a cavernous type, supplied by branches of the external carotid artery. The middle meningeal, superficial temporal and posterior occipital arteries are the main sources of blood supply<sup>9,10</sup>.

The clinical features of hemangiomas include pain and a palpable mass; however, patients often present with neurological deficits such as facial nerve paralysis and hearing loss due to a temporal lesion<sup>2</sup>. Hemangiomas erode the surrounding bone as they grow; however, the inner table usually remains intact<sup>16</sup>. This bone erosion may be identified as a radiating lattice-like or web-like trabecular pattern of the skull. Plain skull X-rays may show an oval or round lesion with a honeycomb-like appearance. The typical sunburst pattern on a tangential view is associated with the osteoclastic activity of the tumor and secondary reactive osteoblastic remodeling with the trabecular bone<sup>7,13</sup>. A CT scan is the diagnostic choice since it confirms the plain skull findings showing detailed intracranial extension<sup>23</sup>. Hemangiomas often exhibit an expanding pattern with a high-density amorphous mass and multiple interposed trabeculae that may be suggestive of fibrous dysplasia. In most cases, this tumor lacks a vascular blush or an identifiable flow-void on MRI<sup>11</sup>. Conventional angiography can play an important role in preoperative planning for small calvarial tumors, and in embolization for large calvarial tumors<sup>1</sup>. The angiography shows increased vascularity in the area of the lesion associated with feeder vessels, and may not always show the true nature of the lesion because the dye may be excreted through the abundant gaps in the endothelium of the cavernous hemangioma before the late pool filling has a chance to occur<sup>19,21</sup>.

Physical examination of a hemangioma may reveal a reddish blue, firm lesion that may or may not be tender<sup>10</sup>. Differential diagnosis includes any firm, slow-growing mass of the skull, such as fibrous dysplasia, meningiomas, osteomas, osteogenic sarcomas, Paget's disease, multiple myelomas, dermoid cysts and hyperparathyroidism<sup>20</sup>.

The etiology of hemangiomas is still unknown. A previous history of head trauma is not considered to be a predisposing factor in the development or genesis of hemangiomas<sup>24</sup>. However, some authors believe that hemangiomas originate from undifferentiated mesenchymal tissue that subsequently differentiates into vascular elements or head

trauma, probably in response to various stimuli that remain unknown<sup>3,5</sup>. The mechanism is not clear, but we would presume that cavernous hemangiomas of the skull develop from trauma or resin material which stimulates certain growth factors.

The best treatment for hemangiomas appears to be a surgical resection of the entire tumor<sup>8,11,14</sup>. Embolization before surgery is helpful in preventing excessive bleeding in large tumors<sup>10</sup>. Radiation has been attempted to treat hemangiomas, and it has been shown to stop tumor growth; however, it has not been shown to reduce the size of the tumor<sup>15,18</sup>.

## CONCLUSION

Although primary calvarial hemangiomas are not rare, a skull hemangioma occurring at a previous cranioplasty site has not yet been reported in the literature. The mechanism is not clear, but we would presume that cavernous hemangiomas of the skull develop from trauma or resin material which stimulates certain growth factors and the best treatment for hemangiomas appears to be a surgical resection of the entire tumor.

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