Giant primary calvarial hemangioma over torcula: Radiological features and operative nuances

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Abstract
Background: Although rare, primary calvarial hemangioma is a known entity, surgical excision of which usually results in massive blood loss. Successful total excision of such a lesion remains a challenge, especially when these are in close vicinity of major venous sinuses.

Case Description: Authors describe a rare case of intra-osseous occipital cavernoma along with radiological findings in a 50-year-old male. En bloc resection of tumor was performed using a high-speed pneumatic drill and no recurrence was noted at 6 months of follow-up.

Conclusion: Intra-osseous cavernous hemangiomas have classical radiological features and can be excised completely, even when lying above major venous drainage channels like torcula.

Key Words: Intra-osseous cavernous hemangioma, radiology, total excision

INTRODUCTION

Primary intra-osseous cavernous hemangiomas (PICHs) account for only 0.2% of all bone neoplasms.[12] Calvarial involvement by giant PICHs[2,4,6,9,11] [Table 1] and PICH involving squama of occipital bone[3,4,7] are rarely reported. These lesions occur in fourth decade and women are affected 2–4 times more often than men.[4] Authors describe a case harboring PICH of occipital squama, exactly above the torcula for which en bloc excision was successfully performed using a high-speed drill.

CASE REPORT

A 50-year-old male presented with history of huge swelling at the midline of the back of the head, progressively increasing in size over the past decade. The patient had no other complaints except for the cosmetic deformity due to swelling. On examination, the lesion was 10 cm in diameter, centering at external occipital protuberance. The swelling was firm to hard in consistency, non tender and immobile. Overlying skin was shiny with visible dilated vessels and was freely mobile. There was no bruit heard over the swelling. Neurological examination was unremarkable. Fundoscopy revealed no papilledema. Computed tomography (CT) scan showed an intra-diploic tumor expanding both inner and outer tables, centered over torcula, with a characteristic sunburst pattern with striations radiating from the center. The underlying brain was normal. Magnetic resonance imaging (MRI) scans showed an extra-axial lesion which was iso-intense and hypointense on T1 weighted sequences and hyperintense on T2 weighted sequences with enhancement. The lesion was centered over the torcular herophili and extended both inwards and outwards with characteristic radiating striations. The underlying brain parenchyma showed no features of raised intracranial pressure.

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Table 1: Reported cases of giant primary hemangioma of calvaria

| Author/year          | Age/sex | Location | Presentation                  | Radiology                                                                 | Management                              | Outcome       |
|----------------------|---------|----------|--------------------------------|---------------------------------------------------------------------------|-----------------------------------------|---------------|
| Toyne/1845[5]        | Parietal| Local swelling | -                             | Total excision                                                           | -                                       | No recurrence |
| Muzumdar et al./2002[2] | 26/female | Torcula  | Local swelling                | 8 cm × 6 cm calvarial lesion at torcula with sunburst appearance          | Preoperative embolization, partial excision, radiotherapy | No recurrence |
| Muzumdar et al./2002[2] | 30/female | Torcula  | Local swelling                | 7 cm × 3 cm intradiploic lesion expanding inner and outer tables           | Preoperative embolization, partial excision, radiotherapy | No recurrence |
| Nasrallah et al./2009[5] | 17/female | Frontal | Local swelling                | 11 cm × 9 cm calvarial mass with bony erosion hyperintense on T1, T2, heterogeneous contrast enhancement | Preoperative embolization, surgical debulking | No recurrence |
| Martinez-Lage et al./2010[2] | 1/male | Parietal | Local swelling                | Ossified lesion with bleed                                               | Complete excision                       | No recurrence |
| Nair et al./2011[4]  | 20/female | Occipital | Headache, gait disturbance local swelling | Extra-axial lesion compressing cerebellum, isointense on T1, hyperintense on T2, brilliantly enhancing | Total excision                          | No recurrence |
| Tyagi et al./2011[10] | 28/female | Parietal | Local swelling                | Hyperdense lesion, erosion of inner and outer tables                    | Total excision                          | No recurrence |
| Patnaik et al./2012[6] | 27/male | Frontal | Local swelling                | Large expansile calvarial lesion, calcification, intracranial extension, dural tail sign | Total excision                          | No recurrence |
| Verma et al./2015[11] | 9/female | Parietal | Local swelling                | Bony lesion with erosion of inner and outer tables                     | Total excision                          | No recurrence |
| Present case          | 50/male | Occipital | Local swelling                | Bony lesion, iso-intense on T1, hyper-intense on T2, brilliantly enhancing | Total excision                          | No recurrence |

on T1-weighted imaging (T1WI) and hyper-intense on T2-weighted imaging (T2WI), enhancing brilliantly on contrast administration [Figures 1a-f and 2a, b]. Magnetic resonance venography (MRV) was not performed.

With a presumptive diagnosis of intra-osseous cavernous hemangioma, meningioma or metastases, patient underwent surgery in prone position. Whole of the bony mass lesion was exposed by linear skin incision and both outer/inner tables were noted to be thin and papery. After delineating the normal calvarial bone all around the tumor, four burr holes were made over there, encircling the bony tumor using cutting burr of the high-speed pneumatic drill. Further, all four burr holes were connected using high-speed pneumatic craniotome and en bloc tumor was gradually lifted up after peeling of dura from the inner table with a penfield dissector. Multiple venous drainage communications were sealed using gelfoam and there was no injury to major venous sinuses. Estimated blood loss was 750 ml and all cut surfaces of vault were sealed with bone wax to avoid inadvertent air embolism. Tumor, with a rim of normal bone, was excised completely and sent for histopathology which showed large thin-walled vascular channels with small thin walled and angulated capillary sized blood filled vessels, suggestive of a cavernous hemangioma of occipital bone [Figure 2c and d]. Postoperatively, patient made an eventless recovery and follow-up radiology depicts total excision of hemangioma [Figure 2e and f]. We have planned cranioplasty at follow-up of 1 year.

**DISCUSSION**

PICHs are rare, benign, slow-growing skeletal neoplasms and more than half of cases occur in vertebrae or skull.[8] Radiography of the cranium is the most effective method to identify PICH as an intra-diploic, extensive, well-defined area of rarefaction with a honey-cob configuration on axial views and a classic sunray pattern of trabeculation on tangential views.[9]

Devoid of reactive sclerosis at the margins, these lesions may present as lytic or dense bone expanding masses.[3] These lesions usually grow by expanding the outer table, rarely encroach intracranially. Our case had characteristic calvarial radiographic sunray pattern of radiating trabeculae without a thin peripheral sclerotic rim. CT scan is more precise in describing tumor location, but MRI is mandatory to describe the relationship of great venous channels underneath. On CT scan, these lesions may have multiple bony spurs projecting into center of the lesion which lends them the appearance often referred to as “corduroy-cloth” or the “sunburst appearance.”[11] On MRI, PICHs appear well-circumscribed, hyper-intense on both T1WI and T2WI, with brilliant enhancement on contrast administration. Change of signal intensity from hyper- to hypo-on T1WI scans suggests loss of fat and an aggressive behavior of lesion as in the present case. Information about patency of venous sinuses can be gathered through MRV, which may be useful while drilling the peri-lesional calvarial vault. Similar information was...
obtained by invasive diagnostic procedure like digital subtraction angiography in two reported cases.\textsuperscript{3}

The literature describes only two cases where such a neoplasm was described just in proximity to torcular.\textsuperscript{11} Both cases underwent partial tumor excision, followed by radiation therapy. We decided for total tumor excision, as the radiology had described intact inner table and no invasion of underlying venous channels or dura. As these lesions have high propensity for recurrence, we favored en bloc resection rather than curettage or partial resection. Intraoperatively, delineating the normal bone circumscribing the bony tumor is very important in surgical strategy. There is no risk of significant bleeding as the sinusoids are intact.\textsuperscript{12} Hence, utilizing high-speed drills for cutting normal bone can prevent massive blood loss and total tumor excision a reality. We advocate the use of high-speed cutting burr for making numerous burr-holes and further joining them with a high-speed craniotome. Gradual lifting of offending intraosseous neoplasm from dura can be done by a dissector after ensuring intact inner table and dura on radiological scans.

**CONCLUSION**

Calvarial cavernous hemangiomas are benign neoplasms, which are occasionally located in close proximity to venous sinuses. These neoplasms have classical radiological features and if carefully noted, these imaging findings can guide for the bold surgical strategy of total cavernoma excision, even when these lesions are lying above great venous channels of dura.

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**Conflicts of interest**

There are no conflicts of interest.

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