

Multifocal cavernous hemangiomas of the skull presenting as lytic skull lesions

Abstract

Multiple osteolytic lesions of the skull raise a clinical suspicion of multiple myeloma or metastasis in an elderly patient and require further investigation when detected. Multifocal cavernous hemangiomas of the skull presenting as multiple osteolytic lesions are a rare cause. Here we discuss a case of multiple cavernous hemangiomas of the skull in a patient with coexisting trigeminal neuralgia. This article highlights the fact that multiple osteolytic lesions of skull due to a purely benign cause like a cavernous hemangioma, is a rare but distinct possibility.

Keywords: Multiple cavernous hemangioma, Skull, Multiple osteolytic lesions

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Introduction

Multifocal cavernous haemangioma of the skull presenting as lytic lesions are a rare entity with only a few case reports till date. Haemangioma are benign lesions, well known to occur in the spine and are of the capillary type whereas the haemangioma that occur in the skull are of the cavernous type and are usually solitary. Here we present a case of multifocal cavernous haemangioma of the skull in a patient with coexisting trigeminal neuralgia.

Case presentation

History and examination

A 64 year old female patient came to the neurosurgery OPD with complaints of a sharp stabbing pain over the right side of the face on her cheek for 3 months duration. Pain was moderate to severe in intensity and was triggered when she washed her face, brushed her teeth and while chewing. Pain lasted for 10 to 20 seconds, occurring several times in a day. Pain was along the V2 distribution. Neurological examination was normal. Fifth cranial nerve testing showed no abnormalities.

Radiographic examination

A MRI brain was done for the evaluation of facial pain. The MRI showed a loop of Superior cerebellar artery in conflict with the right trigeminal nerve and incidentally picked up multiple T1 hypo, T2 hyper. and brightly enhancing lesions in the frontal parietal and occipital skull. CT scan (Figure 1) showed multiple lytic lesions. Spine screening did not pick up any similar lesions.

Treatment

The right frontal lesion was biopsied under local anaesthesia. The biopsy was required to rule out other more serious causes of lytic skull lesions at this age. After getting a marker CT, a small incision was made over the marked site. There was a brownish red discolouration over the overlying bone which was thinned out. The overlying bone was broken using a curette. A brownish red, soft, vascular lesion was noted which was curetted out. The inner table was not breached. Haemostasis was achieved using bone wax. Patient was started on

medication to control her trigeminal neuralgia. The neuralgic pain was under control with medications and patient opted for the medical line of management. As the hemangiomas were asymptomatic in this patient, follow up with evaluation was recommended.

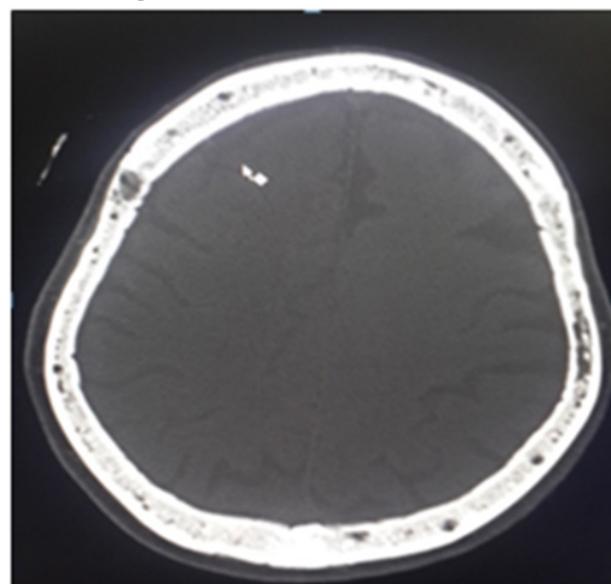


Figure 1 CT scan showing lytic lesions in right frontal and left parietal bones.

Histopathology

Histopathological examination showed multiple, large vascular channels lined by flattened endothelium interspersed with bony trabeculae and areas of haemorrhage. The vascular spaces are separated by fibrous stroma (Figure 2). The vascular spaces appear collapsed and blood is no longer present in some because of the processing. This morphology is commonly associated with cavernous lesions which have been curetted.

Discussion

The first reported case of cavernous hemangioma of the skull was in 1845 by Toynbee.¹ Cavernous hemangiomas of the skull are usually

solitary lesions and multiple lesions are very rare with a few case reports in clinical literature.² Hemangiomas can occur in any bone but they frequently occur in the spine (30-50%) and skull (20%).³

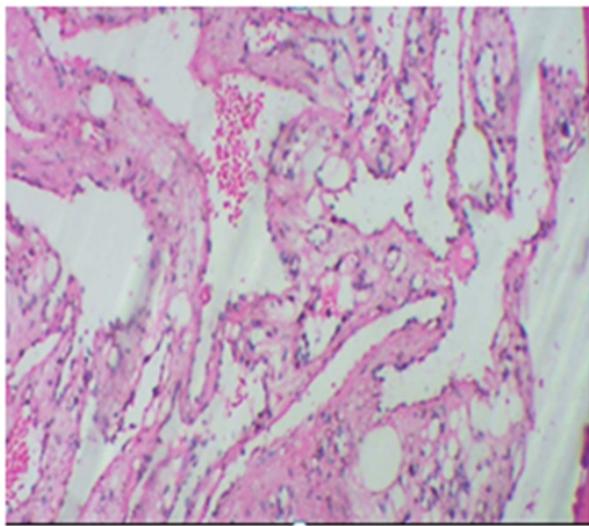


Figure 2 Section showing blood vessels with flattened endothelium separated by fibrous stroma. Few vascular channels show blood in them (10x).

Cavernous hemangiomas of the skull present in the fourth to fifth decade of life. M:F ratio ranges from 3:1 to 2:1.⁴ Out of a total of 93 cases of cavernous hemangiomas reviewed in literature from 1845-2015, frontal bone location was seen in 44.1%, 12.9% in the temporal bone, 11.8% in the occipital bone, 12.9% in the parietal bone and 5.4% in the skull base. Cavernous hemangiomas have also been reported to occur in sphenoid, clivus, orbital rim, ethmoid and zygoma.⁵

These lesions present as a dull aching headache with throbbing pain or as immobile lumps that usually grow extra cranially without eroding the inner table.⁶ But occasionally cavernous hemangiomas are known to erode the inner table and cause extradural hematomas and erode the dura to cause subdural hematomas as well. Cavernous hemangiomas of the skull were first characterized by Rowbotham.¹ Cavernous hemangiomas are benign tumours of the blood vessels. They grow very slowly.⁸ These lesions grow between the outer and inner tables and are supplied by the external carotid artery.⁷

On gross examination cavernous hemangiomas appear as soft vascular tissue with a rubbery consistency, separated by thin bony trabeculae.⁹ These trabeculations or bony interstices are the consequence of reactive osteoclastic and osteoblastic bone remodeling.¹⁰

Microscopically, cavernous hemangiomas possess thin walled vascular channels. These are lined by a single layer of flattened endothelial cells separated by fibrous tissue. These vascular channels are seen to be interspersed among the bony trabeculae.¹¹ They differ from their counterpart the capillary hemangiomas, which have smaller vascular lumens and do not have fibrous septa. The endothelial lining cells are small.¹¹

Skull X-ray, CT and MRI are modes of radiological investigations. CT is considered to be an exceptional investigation as it gives meticulous depiction of cortical as well as trabecular bone. It is usually seen as an expansile lesion with thin borders and intact external and internal skull plates.¹² Sunburst trabeculations and honeycomb appearance is seen in larger lesions and are seldom appreciated in smaller lesions. The sunburst appearance is due to multiple trabeculae arranged radially from a central bony core.¹³

On MRI these lesions have a heterogeneous signal which can be either hypointense or hyperintense on T1 and T2 sequences (Figure 3 & 4). The signal depends upon the quantity of slow moving blood and amount of red marrow which is converted to fat.¹⁴ They enhance on gadolinium contrast with increased peripheral vascularity. Angiography shows a hypervascular lesion with delayed blush with feeding artery and no draining veins.¹⁵

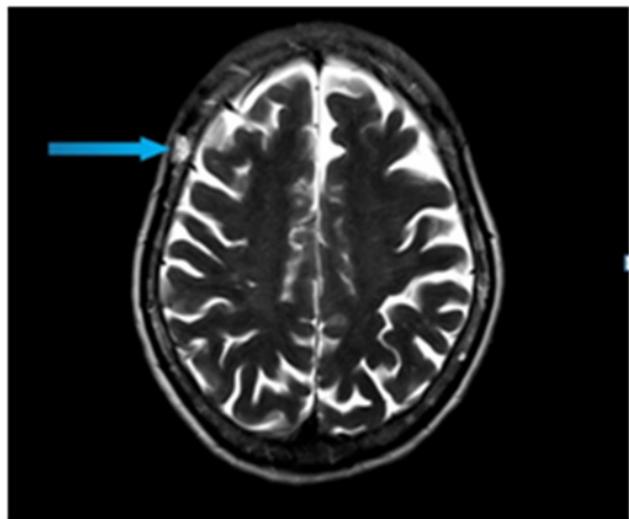


Figure 3 MRI T1 weighted image showing lesion is hyperintense.

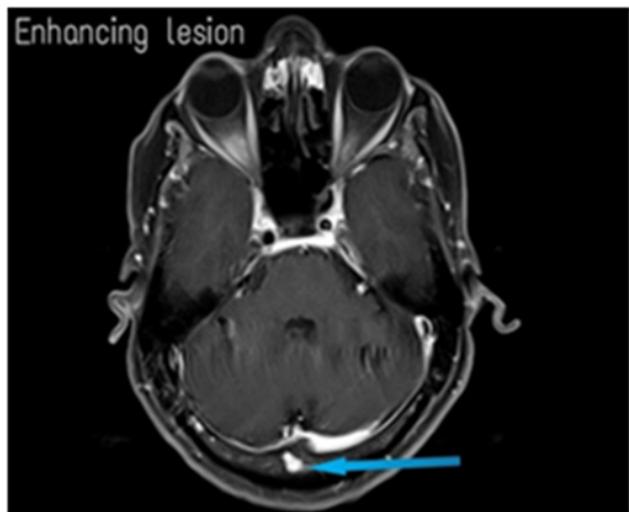


Figure 4 MRI contrast showing enhancing lesion in occipital bone.

Han Hsiao et al.¹⁶ have described a case of a 29 year female patient who presented with a painful skull defect over the right parietal area (Figure 5). CT/MRI showed two individual osteolytic lesions with contrast enhancement over right parietal and frontal areas. MRI showed T1 hypo and T2 hyper with heterogeneous enhancement. A large craniotomy was performed for evacuation of the two osteolytic lesions. Cranioplasty with polymethylmethacrylate was carried out for reconstruction. Histopathology confirmed the diagnosis of a cavernous haemangioma. The patient recovered and is doing well with no recurrence after 4 years.¹⁶ Peterson DL et al.⁶ described a case of a 64 year old male with complaints of frontal and occipital headache. He was found to have two radiolucent lesions in the left frontal and left occipital regions (Figure 6) of the skull on X-ray.⁶ These lesions were proved to be cavernous¹⁷ hemangiomas as well (Figure 7).

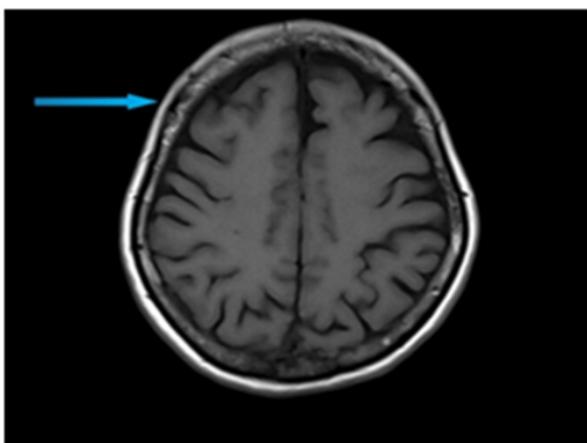


Figure 5 MRI T1 weighted image showing lesion is iso to hypo intense.

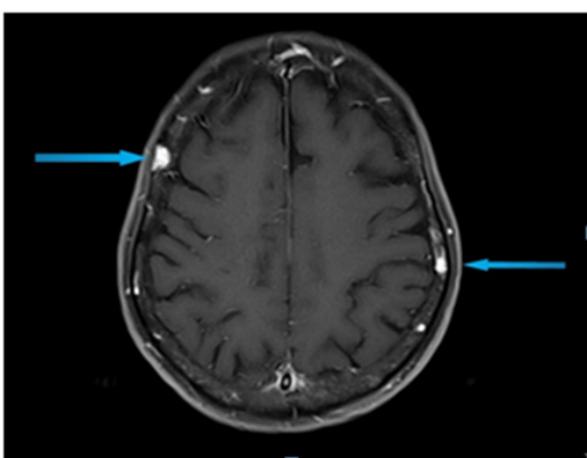


Figure 6 Axial contrast MRI showing enhancing lesions in right frontal and left parietal bones.

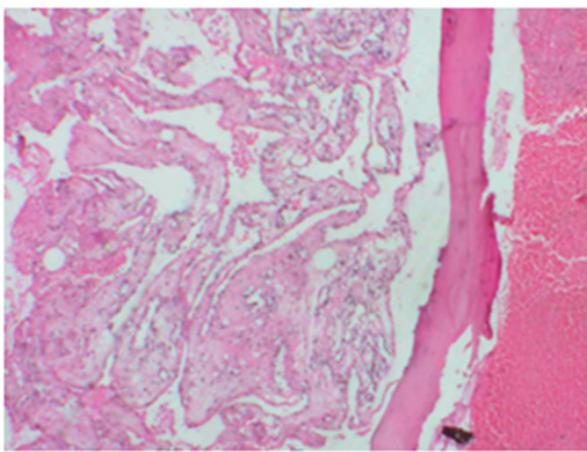


Figure 7 Scanner view of the cavernous vascular spaces.

Treatment of choice of these lesions is en bloc resection with a normal bone margin and cranioplasty. 4 Small asymptomatic and incidentally detected lesions can be followed up conservatively.¹⁸ Curettage alone can cause bleeding and recurrence. Radiotherapy may slow the growth but will not eradicate these lesions and is hence not recommended. However, it may have some role in unresectable skull base lesions.¹

Acknowledgments

None.

Conflicts of interest

None

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