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Calvarial hemangiomas: Series of 6 cases and review of literature

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ABSTRACT

Calvarial haemangiomas are benign, vascular tumours of the skull involving parietal and frontal bones. Mostly these lesions remain asymptomatic, and present with cosmetic deformity, headache, uncommon neurological symptoms and reported as case reports and case series. The radiological appearance can range from sessile growing intradiploically to globular and the lesions may extend outwards or inwards after eroding the outer and inner tables of the skull. "Sunburst appearance" and "Wagon-wheel sign" are classical radiological findings but the lesions may present simply as a lytic expansile or even sclerotic calvarial mass. Because of varied clinical presentation and atypical radiological characteristics, the final diagnosis can be clinched by histology only. In selected cases where these lesions are not cosmetically acceptable, en bloc resection with tumour free margins followed by cranioplasty is the treatment of choice. Most reports of calvarial haemangiomas in literature are in the form of case reports.

Key message

Haemangiomas must be kept in mind while operating on calvarial lesions even if the radiological appearance is not classical. En bloc resections with tumour free margins results in both less operative haemorrhage and surgical cure.

1. Introduction

Calvarial haemangiomas (CHs) are rare, benign, slow growing tumours that account for 2%–10% of calvarial tumours and 0.2% of all bony neoplasms.[1–5] They are commoner in women,[1,3–9] in the 2nd to 4th decades[1,5–7,9] and in the parietal and frontal bones.[1,3,6–10] Osseous haemangiomas have been variously described as being tumours of blood vessels in bones[6] or as vascular hamartomas of the skeletal system[1] and after vertebrae the skull is the commonest location of these tumours.[4] The majority of these lesions are small and incidentally diagnosed on radiological imaging [7,10] but some may reach extremely large sizes at presentation.[5,7–9] Though there is no consensus on what constitutes a giant CH, Jha et al[11] have proposed that tumours above 6 cms in size be classified as giant CHs. We present here a series of 6 operated cases of calvarial haemangiomas and review the existing literature on these uncommon tumours.

1.1. Illustrative cases

We had operated 6 cases of CHs in our centre in the last 8 years [Table 1, Figs. 1–6]. An equal proportion of these (3 each) were in males and females. Three patients were in the paediatric age group (<18 years of age) while the rest were in adults. History of trauma was present in 2 cases and the commonest presenting complaints were local site pain and cosmetic disfigurement. Not all patients of our series had a full complement of preoperative neuroimaging, and a preoperative suspicion of a CH was present in only 2 of these cases. All patients underwent en bloc resection of the lesion wide of the tumour margins with cranioplasty in the same sitting (polymethyl methacrylate cement in 4 and titanium

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Table 1
Details of patients in present series.

Age/ Sex	Presenting complaints	Imaging findings	Provisional preoperative diagnosis	Surgery
76/F	Memory loss, vertigo of 1 year duration	Expansion of the inner table of the skull with classical, well demarcated, spiculated, T1 hyperintense mass	Calvarial hemangioma	Craniotomy and excision with cranioplasty using PMMA bone cement
17/ M	Pain over right parietal region for 4 months. Progressively increasing swelling on the right parietal region for 1 year.	Erosion of outer and inner tables of the skull by a T1 and T2 mixed intensity mass that showed non homogenous enhancement	Unsure	Craniotomy and excision with cranioplasty using PMMA bone cement
45/F	Diffuse dull aching pain over the left side of the head for 3 years with history of trauma to frontal bone several years ago	T1 and T2 isointense mass expanding the diploic space and not causing any cortical breech involving the left parietal bone with extension into the left temporal bone as well	Fibrous dysplasia	Craniotomy and excision with cranioplasty using titanium mesh
12/F	Swelling and pain over right parietal region for 6 months	CT scan shows an expansile lytic mass with erosion of inner cortex	Eosinophilic granuloma	Craniotomy and excision with cranioplasty using PMMA bone cement
14/ M	Progressive swelling on the vertex for last 1 year and itching and ulcerations following topical treatment (scarification) by an unqualified village practitioner	Sclerotic thickened calvarium involving both parietal bones (right more than left)	Fibrous dysplasia	Craniotomy and excision with cranioplasty using titanium mesh
61/ M	Swelling and headache on the right parietal region for 5 years	Lytic expansile lesion on the right parietal bone with intact inner and outer cortices with specs of calcification inside	Calvarial hemangioma	Craniotomy and excision with cranioplasty using PMMA bone cement

PMMA –polymethyl methacrylate.

mesh in 2 cases). A globular mass was found in 3 cases (one of which was necrotic and suckable) while in remaining 3 the bone was grossly normal and sectioning it with a craniotome revealed a mass with clear margins from the surrounding bone. In all cases the histopathology confirmed the diagnosis.

2. Discussion

A PubMed search using the terms "Calvarial", and "Haemangiomas" yielded 96 results. Excluding the papers that deal with radiological approach to diagnosis of calvarial lesions, skull base haemangiomas, other lesions mimicking these like haemangioendothelioma, angiosarcomas, epithelial hyperplasia's, subgaleal haemangiomas, intraosseous venous malformations, aneurysmal bone cysts and those not in the English language we collected a total of 49 reports of 61 cases that are enumerated in Table 2.[1–3,5–50] The classical radiological appearance of CHs consists of an expansile lytic lesion on computed tomography (CT) scans.[3,5,13–15,17] However, there are reports of finding sclerotic lesions mimicking an osteoma as well and also lesions that expand the diploic space with a central sclerosis resembling osteoblastoma.[21] We found expansile lytic lesions in two of our 3 patients who had preoperative CT imaging done. Large lesions have radiating spicules of bony trabeculae giving it the classical sunray (or sunburst) appearance on CT scans and plain radiography of skull.[3–5,51] Sunburst appearance of CHs have been reported by[2–4,17–19,27,28,33,36,39,41,42,44] authors on imaging either in x rays or CT scans. While several authors state that the inner table is usually not violated and that growth occurs by erosion of the outer table,[3,5,9] there are reports describing not only breech of the inner table of the skull[5] but also the dura[10,15,16,27,40,51] In 2 of our cases we encountered inner table violation on preoperative CT imaging and in one another on magnetic resonance imaging (MRI) but there was no intradural extension of the tumour in any of our cases. Calcification is found in CT scans within CHs[11,35] and 3 types of the same have been described[35] – a nonspecific amorphous variant, phleboliths (which are more common) and rarely metaplastic ossification. The last was found in one of our patients on CT scan where we had not considered a CH preoperatively.

On MRI scans the lesions are sharply demarcated, hyperintense on T1 and T2 weighted sequences and enhance on contrast.[5] Hyperintensity on T1 sequences is related to the presence of fat in the lesion and is considered to be characteristic of CHs.[4] It may also be because of haemorrhage in the lesion as has been described by Zhong et al.[49] Nair et al[7] state that loss of T1 hyperintensity is associated with more aggressive tumor behaviour and have also described a CH with dural enhancement akin to a meningioma. We encountered only 1 patient of CH with the characteristic findings on MRI while in another there was intradiploic spread along the parietal bone and in a third there was a mixed density mass eroding both tables of the skull and in the last 2 cases no preoperative provisional diagnosis was reached.

The differential diagnosis of calvarial lesions includes metastatic deposits, eosinophilic granulomas, fibrous dysplasia, osteomas, Paget disease, intraosseous meningiomas, aneurysmal bone cysts and dermoid and epidermoid cysts.[6,8] While radiology can often differentiate between these lesions, atypical and small CH may be difficult diagnose on imaging alone[6] and only histological diagnosis should be considered to be conclusive.[7] In 2 of our cases, we had suspected CHs preoperatively. In the rest we suspected fibrous dysplasia (in 2 cases), eosinophilic granuloma (in 1 case) while in 1 case we could not even reach a presumptive diagnosis.

The commonest bones involved are the frontal and parietal respectively. However, calvarial haemangiomas crossing sutural lines and involving more than one bone have also been described.[11,14,25,33,42] In 2 of our cases the lesion was involving more than one bone.

In all our 6 cases the lesion was a solitary one, but discrete multifocal haemangiomas have been described. [3, 6, 9]Several authors hold that these tumours are congenital in origin[1,8] but due to slow indolent growth[9] and lack of symptoms may present late in life.[6] A number of authors have described paediatric calvarial haemangiomas[5,9,18,20,21,23,29,31,36,40,41,45–49] some as young as 4 months of age[47,48] who had a skull swelling since birth. 3 of our 6 patients were adults and 3 were less than 18 years of age.

Trauma has also been postulated as a cause[3,8] with the injury leading to the release of growth factors [4] that may result in proliferation of primitive mesenchymal cells within bone.[8] Two of our patients had history of head injury in the past but neither was at the location where the lesion was found. There is a case report in literature of growth of the tumour at the site of a cranioplasty[24] while other authors to document

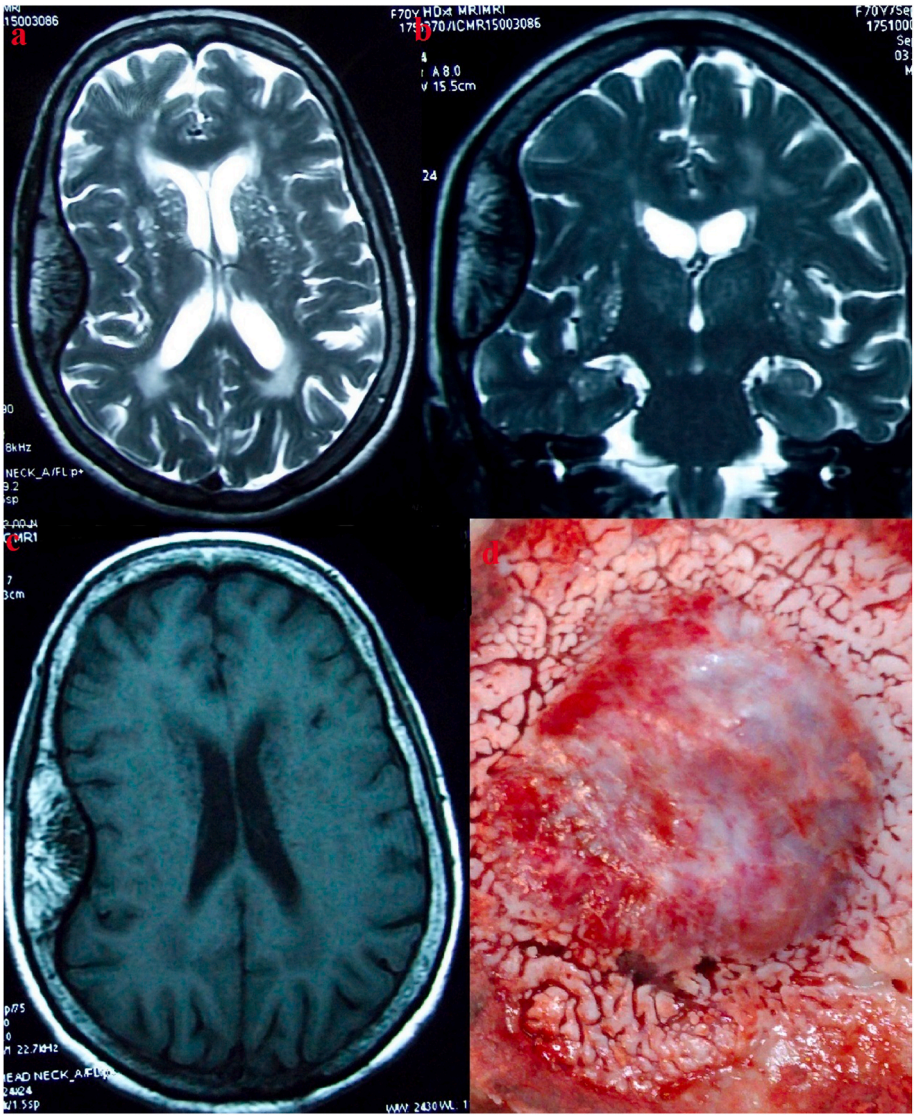

Fig. 1. T2 weighted axial (a) and coronal (b) sections showing expansion of the inner table of the skull by a right sided calvarial lesion with spiculated maxed intensity appearance. The T1 axial image (d) shows hyperintensity in the lesion suggestive of fat. Operative image showing the mass attached to the inner table of the skull with impression of feeding vessels on the latter.

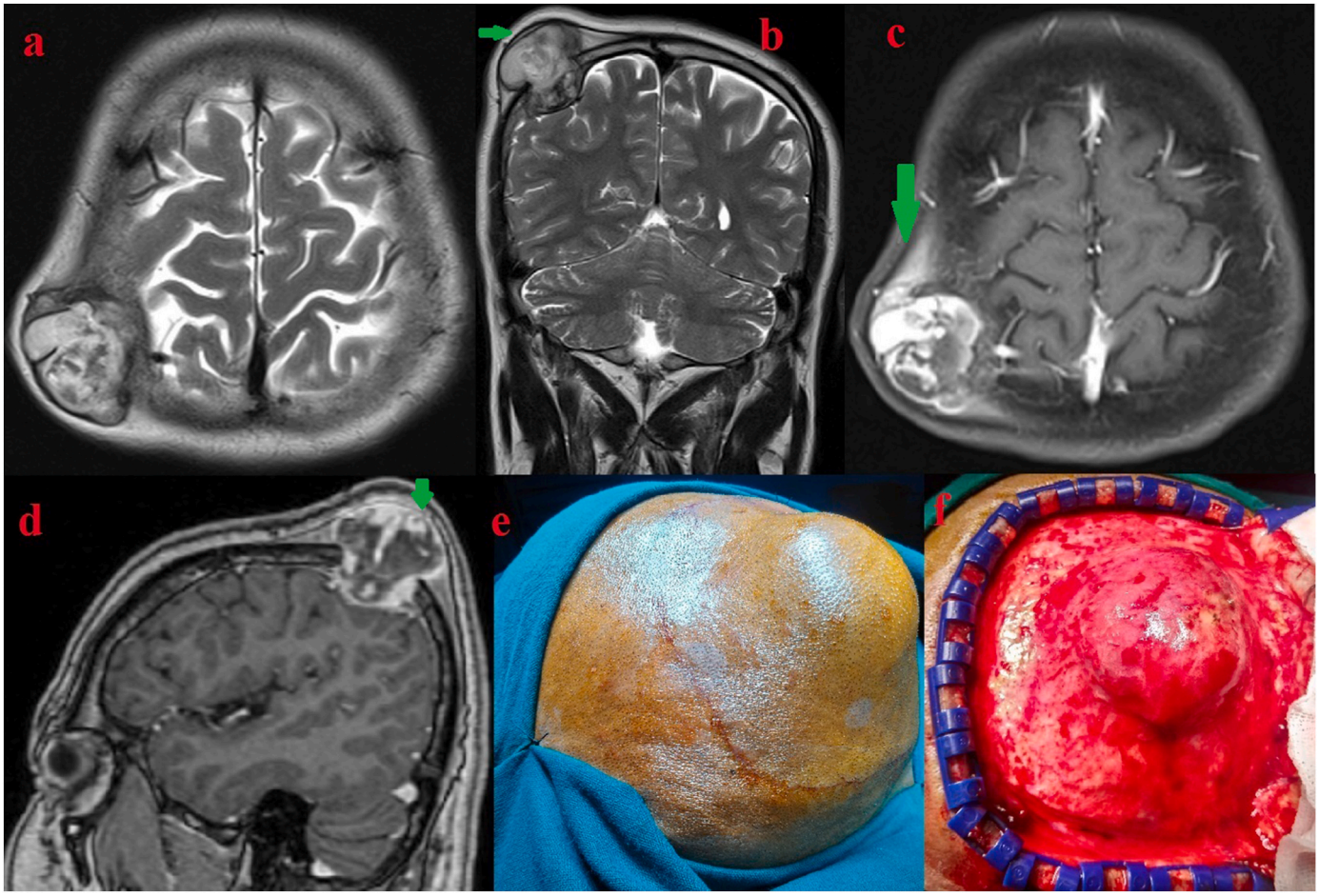

Fig. 2. T2 weighted axial (a) and coronal (b) sections showing a mixed intensity mass eroding through inner and outer tables of the calvarium that is enhancing non homogenously on T weighted axial (c) and sagittal (d) contrast images with no violation of the intact pericranium (green arrows). Clinical photographs showing the lesion causing a scalp swelling (e) preoperatively and (f) intraoperatively. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

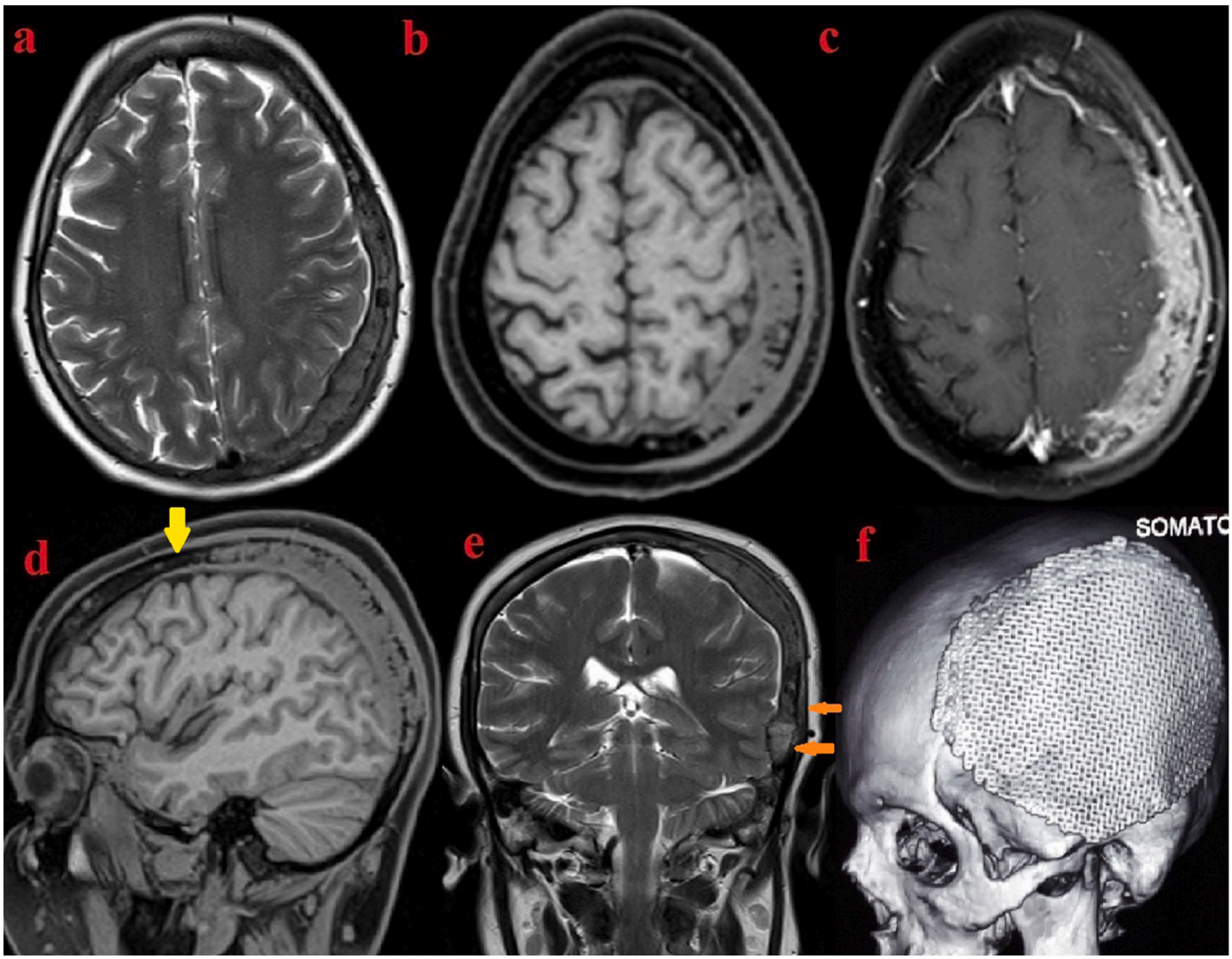

Fig. 3. T2 axial (a) and T1 axial (b) images showing isointense intradiploic mass involving the left parietal bone. T1 contrast axial (c) images show enhancement and T1 sagittal (d) showing lesion stopping short of the coronal suture (yellow arrow) and T2 coronal image (e) showing involvement of the temporal bone (orange arrow). Postoperative 3D reconstructed CT scan (f) showing cranioplasty mesh in situ. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

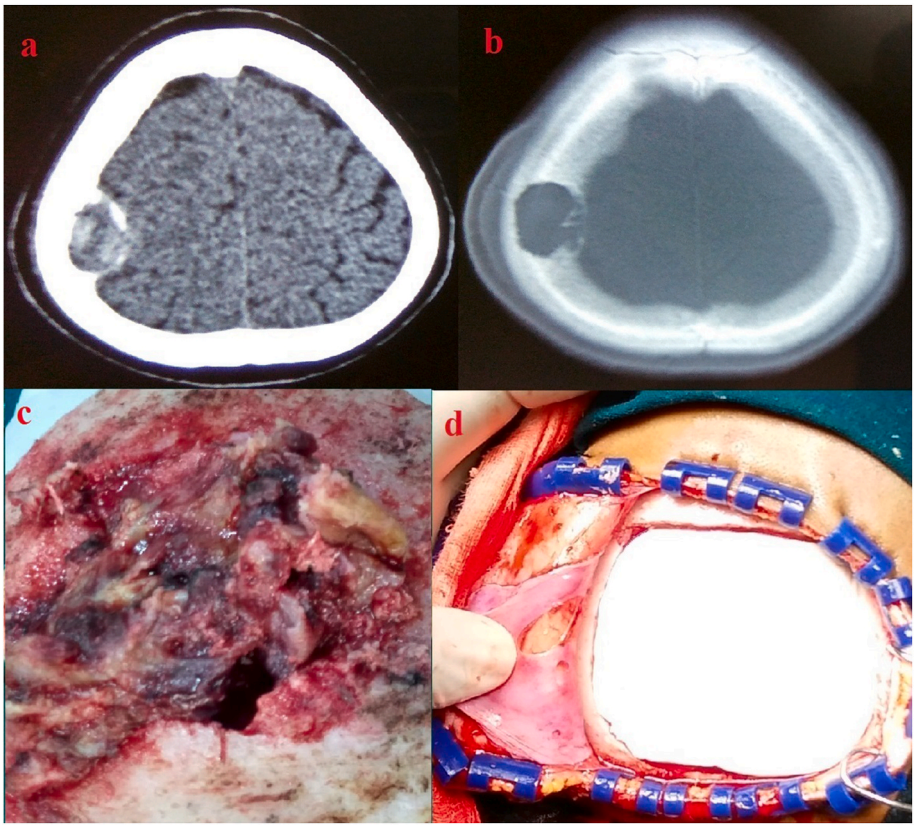

Fig. 4. Axial CT scan image (a) showing an expansile intradiploic lesion with erosion of the inner cortex of the right parietal bone and (b) bone windows showing a punched-out lesion with extracalvarial swelling as well. Intraoperative photographs (c) showing necrotic tumour destroying bone that was partially removed piecemeal during craniotomy and (d) cranioplasty done using bone cement.

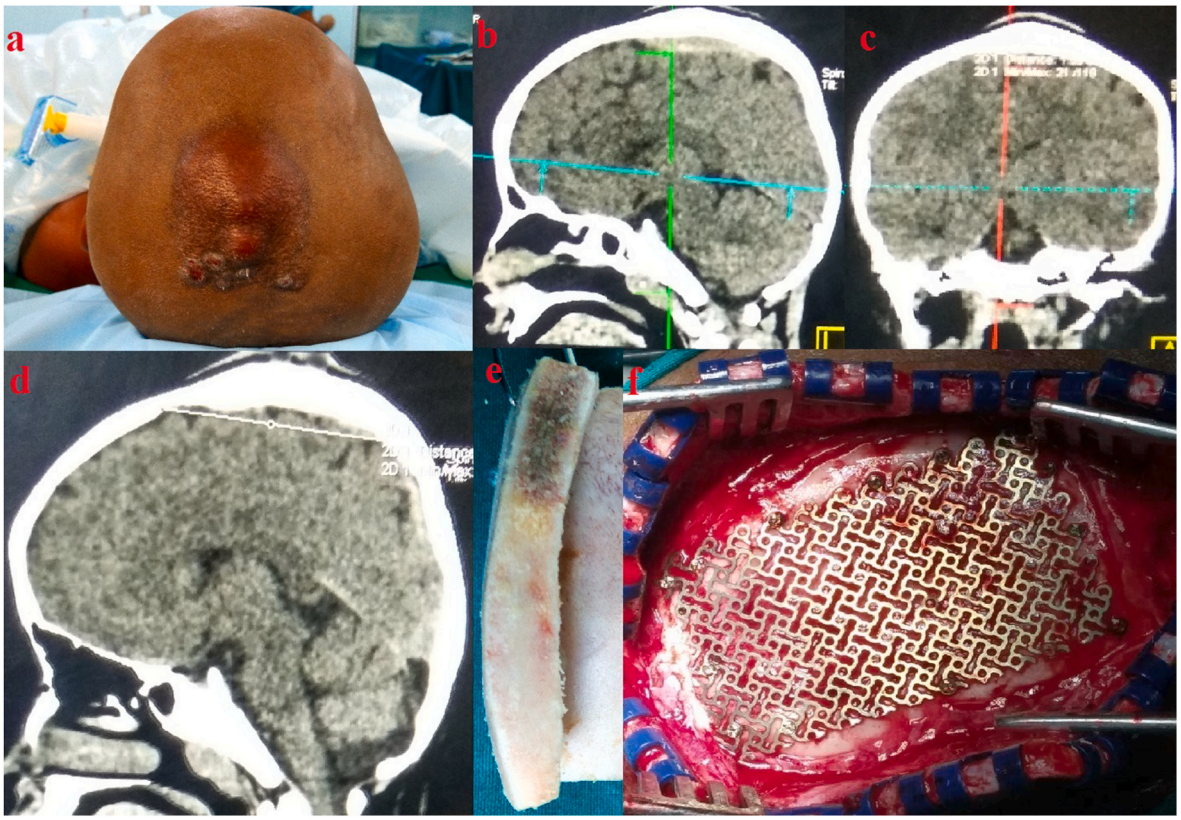

Fig. 5. Clinical photograph (a) showing a patient with a discoloured swelling in the vertex who had been subject to scarification as a form of treatment and (b,c,d) CT images showing thickened sclerotic calvarium. Operative images section of the bone after craniotomy (e) showing a yellowish white mass with intact inner and outer cortices and (f) cranioplasty done with a titanium mesh.

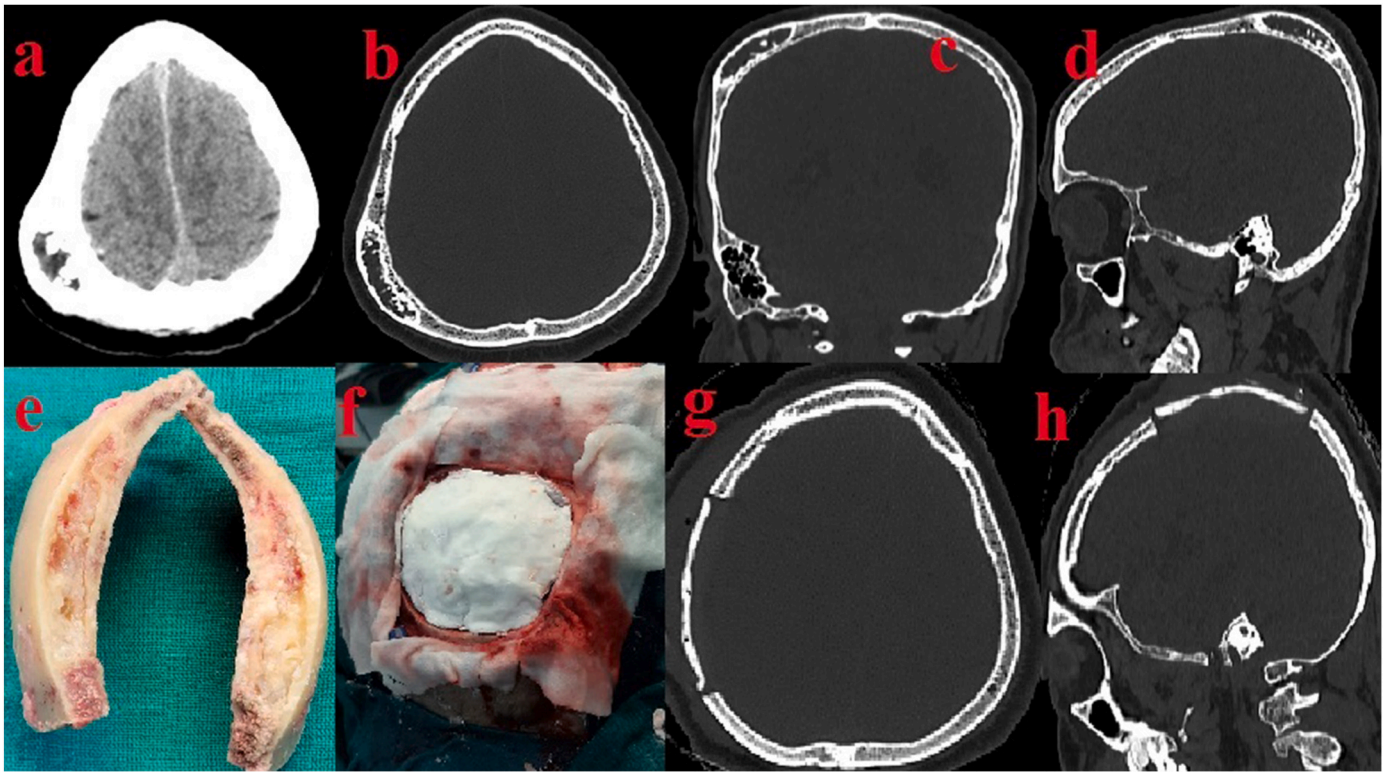

Fig. 6. CT images axial (a) showing a lytic expansile mass in the right parietal bone. Bone windows (b,c,d) in axial, coronal and sagittal planes showing perilesional sclerotic bone with intralesional calcifications and intact outer and inner tables. Intraoperative photographs of the bone sectioned following craniotomy (e) showing a yellowish red well marginated mass and (f) cranioplasty with bone cement. Postoperative CT bone windows in axial (g) and sagittal (h) planes showing complete removal. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Table 2
Summary of cases of Calvarial Hemangiomas described in literature.

Author/Year	No of Cases	Age/Sex of patient	Location	Presentation	Imaging	Treatment and Outcomes
Schneider et al[40] (1973)	1	13/F	Frontal involving sphenoid	Progressive swelling with headaches, dizziness, and temper tantrums	No CT or MR findings. Angiogram showed feeders from MMA. X rays showed a globular bony tumour involving both trabeculae with no stalk.	First operation partial removal due to torrential intraoperative haemorrhage. Re-operated 1 year later after ECA control in neck was taken
Peterson et al[35] (1992)	1	64/M	2 lesions – left frontal and left occipital	Frontal and occipital headaches	Radiolucent skull lesions on X rays	NA
Kumar et al[27] (1996)	1	45/F	2 lesions - left temporal and occipital	Swelling for 20 years with headache, loss of sensation on right side of the face, ataxia, decreased visual acuity for 4 months	X rays - expansile osteolytic mass in occiput with inner and outer table erosion and soft tissue calcification with sunburst pattern in left temporal lesion. CT showed expansion of diploic space with cortical breech and heterogenous contrast enhancement	Total excision of both lesions with removal of involved thinned out dura and duroplasty
Pastore et al[33] (1999)	1	20/M	Left frontoparietal	Painless swelling for 4 weeks	X rays/CT – intradiploic lesion with "honeycomb" pattern with breech of inner table of skull	Total excision with CP No recurrence at 2 years
Yoshida et al[47] (1999)	1	4months/ M	Left parietal	Palpable mass since birth	CT - expansile mass with breech of inner table. MRI – heterogeneous hyperintensity on T1 and T2 sequences	Total excision with no recurrence at 6 months
Corr et al[19] (2000)	1	28/F	2 lesions – right occipital and right frontal	Painless progressive swelling for 7 years	CT - spiculated sunburst appearance. MRI – T1 heterogenous and T2 hyperintense. Angiogram – fed by MMA and OA.	Biopsy confirmed lesion. Outcomes NA
Khanam et al[25] (2001)	2	30/F 51/M	Right greater wing of sphenoid extending to right frontal Right occipital and incidental left parietal	Headaches for 1 month Palpable right occipital mass with mild pain for 12 years	CT scan - partially calcified lesion MRI - T1 isointense and T2 and FLAIR hyperintense lesion with homogeneous contrast enhancement with some dural enhancement X rays - radiolucent lesion with a radiating trabecular pattern in the right occipital bone CT scan – well defined lesion with mixed lytic and sclerotic patterns with bicortical breech. Incidental right parietal lesion also seen with similar appearance MRI - mixed intensity expansile lesion on T1- and T2-sequences with heterogeneous enhancement and no dural involvement	Total excision Excision of outer cortex of occipital lesion with curettage of material. Inner cortex was intact. Parietal not operated.
Ajja et al[12] (2005)	1	31/F	Left parietal	Painless progressive swelling over 1 year	X ray/CT – osteolytic lesion in left parietal bone with destruction of outer table	Total resection and CP
Namaa et al[6] (2008)	2	30/F 51/M	Right sphenoid wing Right occipital Incidental right parietal	Headache for 1 month Mild occipital pain for 1 month	CT – partially calcified bony lesion MRI – T1 isointense and T2 hyperintense with contrast enhancement X rays - radiolucent lesion with sunburst appearance CT - well defined mixed lytic - sclerotic lesion with cortical breech. MRI - mixed intensity lesion on T1- and T2-sequences with contrast enhancement.	Total excision Total excision of occipital lesion
Gordhan[22] et al (2009)	1	49/F	Left frontal	Intermittent left frontal headaches	CT – expansile, intradiploic, circumscribed mass with internal trabeculations. MRI – isointense on T1 and T2 with contrast enhancement. Scintigraphy - delayed radiotracer uptake	Total excision
Sasagawa[38] et al (2009)	1	55/F	Right frontal	Painless progressively growing swelling for 6 years	X rays/CT – osteolytic skull lesion MRI – T1 iso and T2 hyperintense lesion with contrast enhancement	Total excision with CP
Kang et al[24] (2009)	1	46/M	Left parietal at site of old CP for depressed fracture	Painless progressive swelling for several months	X-rays - radiolucent, circumscribed lesion CT - partially enhancing mass with mixed osteolytic and sclerotic patterns MRI – CP plate lifted up by a mass	Total excision with re-CP

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Table 2 (*continued*)

Author/Year	No of Cases	Age/Sex of patient	Location	Presentation	Imaging	Treatment and Outcomes
					that showed heterogenous enhancement on contrast with no dural involvement	
Satoh et al[39] (2009)	5	40/M 36/F 39/M 38/M 38/F	Left frontal (all cases)	Painless progressive swelling in all cases	X rays - honeycomb appearance 3 cases, sunburst pattern 1 case CT – expansile lesion with inner table breech in 2 cases and sunburst pattern in 2 cases	Total excision with CP using split calvarial graft
Nasrallah et al[31] (2009)	1	17/F	Left frontal	Palpable skull mass for 6–7 years	CT – expansile intradiploic mass with well-defined margins and coarse trabeculations MRI - No extraosseous extension and heterogeneous hyperintensity on T1 and T2 sequences with heterogenous enhancement on contrast MRA enlargement of the left MMA	DSA and embolization of the MMA followed by craniotomy surrounding the lesion, de-bulking of the bone flap leaving only outer table intact and replacing it. No recurrence at 38 months
Vural et al[45] (2009)	1	6 months/ M	Right parietal	Palpable skull mass since birth	X rays - asymmetric enlargement of the calvarium on right with nonspecific increased radio-opacity CT – diploic expansion with increased thickness of both tables with some defect in inner table MRI - T1 iso and T2 hyperintense mass with contrast enhancement	Total excision and CP
Hong et al[23] (2010)	5	34/F 50/F 52/F 72/M 9/M	Temporal Parietal Parietal Frontal Parietal	Incidental Dx Incidental Dx Incidental Dx Incidental Dx Painful progressively increasing mass	NA but all had osteolytic lesions of the skull	NA
Nair et al[7] (2011)	1	20/F	Left occipital	Headache, gait instability, visual blurring of 2 months duration	CT- hyperdense calcified lesion in left posterior fossa involving occipital bone. MRI - extra-axial T1 isointense and T2 hyperintense lesion with contrast enhancement with occlusion of left transverse sinus on MRV	Complete excision with postoperative right hemiparesis and cerebellar signs that resolved in 6 months
Tyagi et al[43] (2011)	1	28/F	Right parietal	Non tender swelling increasing over 15 years	CT – uniformly hyperdense lesion with erosion of both the inner and outer tables	Total excision with CP after 6 months. No recurrence at 3 years
Yucel et al[48] (2011)	1	4 months/ F	Right parietal	Progressively increasing swelling	CT - -soft tissue mass with expansion of diploic space and inner cortical breech	Total excision
Patnaik et al[34] (2012)	1	27/M	Left frontal	Progressively increasing swelling	CT - expansile lesion with intratumoral calcification destroying both tables of the skull. MRI- Contrast enhancement with adjacent dural enhancement.	Excision
Zhong et al[49] (2012)	1	16/M	Left temporal	Progressive swelling in left temple of 3 months duration	CT - expansile intradiploic lesion with well-defined margins with destruction of both inner and outer tables MRI – T1 hyper and T2 mixed intensity with mild heterogenous enhancement with mass effect on the brain	Total excision with removal of involved dura and CP followed by neurological deterioration due to operative site bleed and needed removal of bone flap and clot evacuation
Atci et al[1] (2013)	1	38/M	Left parietal	Intermittent localized headache and palpable mass for 2 years	CT - increase in the left parietal diploic space MRI – well defined expansile lesion	Total excision
Park et al[32] (2013)	1	39/F	Left frontal	Progressive enlargement of swelling for 1 year	X-ray - radiolucent lesion in the frontal bone CT - intradiploic osteolytic mass	En bloc resection with CP using split calvarial graft
Uemura et al[44] (2014)	2	34/F 51/F	Right frontal Right frontal	Progressive swelling in the right side of forehead for 2 years Progressive swelling in the right side of forehead for 1 years	CT – expansile, well demarcated lesion with honeycomb appearance X ray – punched out lesion CT – expansile lesion with intact inner table	En bloc resection with CP using split thickness calvarium En bloc resection with CP using calcium phosphate bone cement
Dutta et al[2] (2015)	1	40/F	Left frontal	Painless progressive swelling of left forehead for 2 years, proptosis for 7 months with dimness of vision and diplopia	CT -well-demarcated, contrast-enhancing, expansile osteolytic lesion involving both tables with sunburst pattern	Excision details NA
Verma et al[9] (2015)	1	9/F	Left temporal	Progressively increasing swelling for 2 years duration	CT- mixed density contrast enhancing extra-axial lesion with bony destruction MRI – T1 hypointense and T2 hyperintense lesion with areas of	Near total excision in 2 stages with stereotactic radiosurgery for small residue

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Table 2 (*continued*)

Author/Year	No of Cases	Age/Sex of patient	Location	Presentation	Imaging	Treatment and Outcomes
					intratumoral haemorrhage and heterogenous contrast enhancement	
Davern et al[21] (2015)	2	13/F 19/F	Right parietal Left parietal	Painless swelling growing under observation over 6 years Painless progressively enlarging swelling	CT - hyperdense mass involving the outer table of skull but not the inner CT - well demarcated expansile lesion with a central area of bony sclerosis	Outer table and diploe resected with preservation of inner cortex No recurrence at 1 year Total excision with delayed CP after 6 months
Kirmani et al[3] (2016)	1	40/F	Right parietal	Painless progressively increasing swelling for 2 years	X ray – AP/lat lytic lesion with honeycomb appearance CT - hypodense lesion with sharp, sclerotic margins ^{99m}Technitium (Tc) labelled bone scan showed only mild increase in tumour	Total excision with curettage of surrounding margins with CP
Nasi D[30] (2016)	1	60/M	Left frontal	Progressively increasing skull mass for 6 months Increasing headache, nausea, right sided weakness for 2 weeks	MRI – T1 iso and T2 hyperintense lesion destroying both tables with heterogeneous enhancement on contrast	Total excision of involved bone with removal of subgaleal and intradural component and CP
Mohindra et al[28] (2016)	1	50/M	Midline occipital	Progressively increasing swelling over 10 years	CT - intra-diploic tumour expanding both inner and outer tables with sunburst pattern MRI – T1 iso-intense and T2 hyper-intense with contrast enhancement	Total excision No recurrence at 6 months follows up
Sharma et al[41] (2016)	1	8/F	Midline frontal	Pain and swelling on forehead for 4 months	X-ray - lytic expansile lesion with sunburst pattern MRI – well circumscribed lesion eroding both tables, T2 heterointense with contrast enhancement with mass effect and edema in right frontal lobes	En bloc resection
Yang et al[46] (2016)	1	17/F	Left frontal	Progressively enlarging swelling for 2 years	X-rays - radiolucent skull mass CT - osteolytic intradiploic lesion	Total excision with no recurrence at 1 year
Srinivasan et al[42] (2016)	1	35/F	Midline parieto-occipital	Occipital headaches, visual blurring 1-month Fundoscopy - bilateral optic disc edema	CT - well-circumscribed mass with thickened trabeculae expanding the diploic space in a "starburst" pattern MRI - expansile midline mass with parietooccipital calvarium with mass effect on the occipital lobe, cerebellum, and torcula - T1 hyper and hypointense with enhancement on contrast MRV - compression of the torcula and narrowing of the distal SSS	Total excision with delayed CP
Prasad et al[5] (2017)	1	12/F	Left parietal	Painless hard progressively increasing swelling for 6 years	X rays/CT- expansile lytic lesion with bony trabeculae radiating from the centre of the mass MRI – T1 iso to hyper, T2 hyperintense with contrast enhancement indenting underlying brain with mass effect and midline shift	NA
Saenz et al[37] (2018)	1	57	Left frontal	Painless progressive enlargement of swelling for 4.5 years	X-ray - radiolucent lesion in the frontal bone CT - osteolytic mass with preserved inner table	Total resection with PMMA CP
Brichacek et al[18] (2018)	1	2/M	Right parietal	Progressively increasing swelling for 1 year	X rays - focal thickening of bone with sunburst appearance CT - bone thickening with coarsening of the bony trabeculae, minor irregularity of the outer table and unaffected inner table MRI – well circumscribed intradiploic mass enhancing on contrast	Diagnostic biopsy to confirm haemangioma followed by propranolol therapy for 3 years and 4 months Post-treatment MRI showed decrease in lesion size
Ilyas et al[50] (2018)	1	50/F	Occipital	Progressively increasing occipital swelling with tenderness	X rays - well-circumscribed, lytic lesion with sunburst appearance CT - mixed lytic and sclerotic expansile lesion, with destruction of both tables MRI – T2 hyperintense enhancing lesion with hypointense centre and multiple serpentine vessels on MRA	NA
Prasad et al[36] (2018)	1	12/F	Left parietal	Painless progressive swelling	X rays – well demarcated lytic lesion with honeycomb pattern	Total excision and CP with PMMA

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Table 2 (*continued*)

Author/Year	No of Cases	Age/Sex of patient	Location	Presentation	Imaging	Treatment and Outcomes
					CT – lytic lesion with destruction of both tables MRI – T1 iso to hypo and T2 hyperintense with contrast enhancement	
Ryu et al[10] (2018)	1	56/M	Right frontal	Increasing headache for 1 month	X rays – radiolucent skull defect MRI T1 iso and T2 hyperintense lesion destroying both tables and enhancing on contrast	Total excision with removal of intradural component, duroplasty and CP
Prasanna[8] et al (2019)	1	32/F	Right parietal	Progressively increasing non-tender swelling	CT - hyperdense osteolytic lesion with a characteristic spoke-wheel pattern MRI -hyperintense on T1and T2 sequences with contrast enhancement	Total excision
Akhter et al[13] (2019)	1	68/F	Frontal	Forehead discomfort with proptosis and diplopia	CT- well circumscribed, expansile mass with internal trabeculations and sclerotic margins MRI showed enhancing mass with dural involvement	Gross total removal with CP Asymptomatic at 4 months follow up
Kim et al[26] (2019)	1	60/M	Left frontal and left parietal	Painless mass left forehead expanding for 2 years	USG -well-defined hypoechoic lesion in subgaleal layer with internal septations and focal cortical discontinuity with intact inner cortex with normal doppler study Postop CT – another lesion incidental in left parietal bone	Partial excision of left frontal lesion
Bravo-Martinez et al[17] (2019)	1	58/F	Midline frontal	Painless mass of long duration progressively increasing for 1 year following trauma	X rays - expansile lytic lesion CT - well-demarcated, trabeculated mass with a sunburst pattern MRI - T1 mixed intensity and T2 hyperintense	Embolization of feeders followed by total resection and CP
Jha et al[11] (2021)	1	35/F	Right parietotemporal	Headaches and recurrent seizures for 2 years	CT - hyperdense, spiculated lesion breaching both tables MRI – T1 hyperintense with contrast enhancement Tumour blush on DSA	Total excision with delayed CP
Cui et al[20] (2021)	1	6/M	Left temporal	Hallucinations for ½ year	CT - circumscribed hyperdense mass with wagon wheel appearance MRI - T1 hypointense and T2 hyperintense with contrast enhancement CTA/DSA - abundant blood supply from ECA	Embolization followed by Total excision with CP
Nagamine et al[29] (2021)	1	5 month/F	Left occipital	Progressively increasing swelling	CT - Hyperdense lesion eroding calvarium MRI - T1 mixed and T2 hypointense lesion not enhancing	Completely excised with no recurrence at 1 year follow up
Bantan et al[15] (2021)	1	29/M	Left occipital and bilateral parietal	Left occipital progressively increasing mass since childhood with recurrent seizures and recent onset headache, vomiting and decreased sensorium for 1 day	CT - parietal and occipital expansile lytic bony lesions involving both tables with internal trabeculations and spiculated pattern MRI - T1 and T2 hyperintense enhancing on contrast and showing haemorrhage on GRE sequences Left parietal lesion invaded the SSS Left occipital erosion of both tables with mass effect on occipital lobe MRV absence of flow in distal SSS and left transverse sinus Empty sella and distended optic nerve sheaths seen	DSA and embolization of left occipital and left MMA followed by total excision of occipital and left parietal lesions with removal of dura and intradural part and titanium mesh CP
Bird et al[16] (2022)	1	38/F	Midline frontal	Painless progressive swelling for 7 months	CT – well -circumscribed mass with sunburst pattern expanding through the inner and outer tables MRI – T2 hyperintense heterogeneously enhancing lesion with mass effect on brain DSA - tumour supplied by STA and anterior falcine artery	Total excision of lesion with involved dura and intradural component with duroplasty and titanium mesh CP No recurrence at 6 months
Anagnostou et al[14] (2022)	1	59/F	Left frontoparietal	Palpable mass, personality changes, emotional and cognitive dysfunction	CT - expansile lytic mass with contrast enhancement eroding both tables of skull MRI - T1 hypointense and T2 hyperintense lesion DSA tumour blush	Embolization followed by total excision Satisfactory at 6 months

Abbreviations AP- Anteroposterior, Lat –lateral, CP- Cranioplasty, CT – Computed tomography, CTA – CT angiography, DSA – Digital Subtraction Angiography, ECA – External carotid artery, FLAIR – Fluid attenuation inversion recovery, GRE – gradient echo, MMA – Middle meningeal artery, MRA – Magnetic resonance angiography, MRI – Magnetic Resonance Imaging, MRV – Magnetic resonance venography, NA-not available, PMMA- Polymethyl methacrylate, OA-occipital artery, STA – superficial temporal artery, SSS – superior sagittal sinus, USG -ultrasonography.

history of trauma as reported by the patients[17] though they do not attribute this to be causal in the formation of the tumour.

Two types of CH are described – the commoner sessile type with expansion of the diploic space and a globular type that extends out of the confines of the bones into the surrounding space.[4,8,9] Our series had 3 cases of each type.

While smaller asymptomatic lesions with classical imaging findings may be kept under observation, indications for surgery include cosmetic deformity, pain, diagnostic dilemma and mass effect on the brain. Pain was present in 4 of our patients and progressively increasing scalp swelling in 4. Progressively increasing painless swelling with or without local tenderness or mild pain is the commonest presentation reported in literature too.[6,12,19,22,24,25,33,38,39,45] However, there are reports of patients presenting with features of raised intracranial pressure[7,15,27,42,51] as well as emotional disturbances,[14,20,40] ataxia,[27] diplopia and proptosis[2,13] and seizures.[11,15] In our series one patient had no symptoms related to the CH nor any history of progression of the lesion but after an initial period of conservative treatment for 2 years her relatives wished that the lesion be removed.

When the decision is taken to proceed with surgery, complete resection[3–5,7,8] is the treatment of choice. Curettage of the lesion alone results in greater blood loss and a propensity for recurrence.[4–6] Hence most authors have mentioned complete resection as the procedure that they carried out but there are reports of curettage alone along with removal of the outer table[21,25,26] particularly in cases where the inner table was preserved and also craniotomy followed by debulking of the diploe and replacement of the outer layer.[31] Atci et al[1] have recommended taking margins of the adjacent non-involved bones during surgery while Namaa et al[6] advocate resection with a 1 cm margin of normal bone to prevent recurrence and decrease intraoperative haemorrhage as the sinusoids in the tumour are not disturbed. As all our patients with CHs were excised with tumour free margins, we did not face troublesome bleeding in any of them. We performed immediate cranioplasty (CP) following tumour excision in all our patients. In 4 cases we used polymethylmethacrylate (PMMA) bone cement while in 2 cases titanium mesh fixed with screws was employed to cover the skull defect. Materials for CP mentioned in literature vary from split calvarial graft[32,39,44] to titanium mesh[15,16] and bone cement[36,37,44] Most authors describe resection and CP at the same sitting.[12,14,15,20,33,39,45] However, there are reports of staged cranioplasty too[11,21,42] usually due to torrential haemorrhage or doubt regarding the nature of the lesion.

Though we did not embolize any of our patients preoperatively, this has been described as a useful adjunct to minimize blood loss in various studies[3,5,8,14,15,17,20] and they have been reported to be fed by the branches of the superficial temporal artery, middle meningeal artery, occipital artery or anterior falcine artery.[9,11,16,19,31,40] Radiotherapy has a role in incomplete resections[3,4] and may arrest further growth without decreasing size[6,8] but was not required in any of our cases as all of these were exclusively in the calvarium and amenable to total resection.

3. Conclusion

CHs are uncommon skull tumours that are usually asymptomatic, commonly presenting with scalp swelling and occasionally with pain. Rarely they may present with neurological deficits. Classical radiological findings include sunburst sign and honeycombing appearance, but this may not be found in all cases and the final diagnosis is then clinched only by histological examination. Both CT scan and MRI are mandatory to define extent of spread when planning for surgery. Angiography and embolization of vascular or large lesions may aid in minimizing blood loss during excision. Wide local excision and cranioplasty is easy to perform in the cranial vault (unlike in the skull base) and is curative.

CRediT authorship contribution statement

Prasad Krishnan: Conceptualization, Data curation, Methodology, Visualization, Writing – review & editing, Writing – original draft. **Rajesh Bhosle:** Conceptualization, Data curation, Visualization, Writing – original draft, Writing – review & editing. **Shamshuddin Patel:** Conceptualization, Supervision, Writing – original draft, Writing – review & editing. **Dimble Raju:** Conceptualization, Methodology, Supervision, Writing – original draft, Writing – review & editing. **Rafael Cincu:** Conceptualization, Supervision, Writing – original draft, Writing – review & editing. **Luis Rafael Moscote-Salazar:** Conceptualization, Data curation, Supervision, Writing – original draft, Writing – review & editing. **Amit Gupta:** Conceptualization, Methodology, Resources, Writing – review & editing. **Amit Agrawal:** Conceptualization, Methodology, Supervision, Visualization, Writing – original draft, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Abbreviations

AP	anteroposterior
Lat	lateral
CP	Cranioplasty
CT	Computed tomography
CTA	CT angiography
DSA	Digital Subtraction Angiography
ECA	External carotid artery
FLAIR	Fluid attenuation inversion recovery
GRE	gradient echo
MMA	Middle meningeal artery
MRA	Magnetic resonance angiography
MRI	Magnetic Resonance Imaging
MRV	Magnetic resonance venography
NA	not available
PMMA	Polymethyl methacrylate
OA	occipital artery
STA	superficial temporal artery
SSS	superior sagittal sinus
USG	ultrasonography

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