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Review Article

Intraosseous Haemangioma of the Frontal Bone: A Review of Published Cases and Surgical Management

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Abstract

Background: Intraosseous haemangioma of the calvarium is a slow-growing diploic vascular lesion, most often frontal, that reaches the surgeon as a painless, hard swelling of the forehead.

Objective: To establish what the published literature will and will not support regarding frequency, preoperative diagnosis, surgical management and recurrence.

Methods: Narrative review of published case reports, pooled literature tallies and institutional surgical series.

Findings: The commonly quoted figures of 0.2% of bone tumours and 10% of benign skull tumours have no traceable denominator; a counted operative series found haemangioma in 9 of 133 calvarial lesions (7%). Frontal involvement accounted for 44.1% of 93 published skull cases. No recurrence occurred in 16 operated patients at mean follow-up of 76.2 months.

Conclusions: The evidence base is case-level. It supports en bloc excision with a small bony margin and immediate reconstruction; it does not support the quoted rarity, sex ratio or embolisation claims.

Keywords: Intraosseous haemangioma; Frontal bone; Calvarium; En bloc resection; Cranioplasty

Introduction

Intraosseous haemangioma of the calvarium is a slow-growing vascular lesion of the diploic space that most often presents as a painless, hard, fixed swelling of the forehead. The frontal bone is the site most frequently reported, and the lesion reaches the craniofacial surgeon precisely because it is visible. The literature on it is, however, of a particular and limited kind. It consists almost entirely of single case reports, supplemented by a small number of pooled literature tallies and two modest institutional series. This review sets out what that literature will and will not support.

The claim that opens most papers on this subject is that intraosseous haemangioma accounts for 0.2% of all bone tumours and 10% of benign tumours of the skull. That pair of figures is repeated across case reports and radiological reviews [1,2,3], yet no source attaches a series, a registry or any counted denominator to it, and the citation chain terminates in older secondary literature. The figures are also internally inconsistent. The introduction of one widely cited paper states that the lesion represents 0.7% to 1.0% of all bone tumours while its own discussion, a few paragraphs later, gives about 0.2% for the same quantity [4]. A further variant, less than 1% of bone tumours and around 7% to 10% of benign skull tumours, appears elsewhere [5]. These cannot all be correct, and none can be checked.

A counted substitute exists. In a single-centre review of 133 calvarial lesions taken to operation, haemangioma was the histological diagnosis in 9, or 7% of the series, alongside histiocytosis in 20, meningioma in 20, metastasis in 19, osteoma in 16 and fibrous dysplasia in 6 [6]. This figure has a denominator, a defined population and a stated method of ascertainment. It describes operated lesions rather than all skull tumours, and it should be preferred to the conventional 10% for that reason rather than despite it.

The Rarity Estimates and What They Rest On

Table 1: Published counts in this literature that carry a stated denominator, with the quantity counted and the result.			
Source	N	What was counted	Result
Nasi-Kordhishti 2021	133	Operated calvarial lesions, one centre	Haemangioma 9 (7%)
Yang 2016	93	Published skull cases, 1845-2015	Frontal 44.1%
Alexiou 2022	77	Patients in 51 studies	Mean age 32.7 y; F:M 1.4:1
Brandner 2018	51	Frontal-bone cases to 2016	Mean age 37.6 y; 31 women
Wang 2020	16	Operated cranial lesions	0 recurrences, mean 76.2 mo
Krishnan 2024	6	Operated cases over 8 years	Preop diagnosis in 2 of 6
Bruder 2009	77	Bone vascular lesions in the young	46 malformations, 31 tumours

The purpose of stating this at the outset is not pedantry. A figure without a denominator cannot be checked, cannot be updated as practice changes and cannot be compared with anything, and when the same figure is contradicted within a single paper it has ceased to carry information. Reviews of this entity have reproduced the convention for two decades [7,8], and the effect is a settled impression of precision that the underlying counts do not support.

Two other proportions in this literature carry real denominators. Cranial intraosseous cavernous malformations made up 1.15% of one institution's complete series of intracranial and intraspinal cavernous malformations treated between 2003 and 2016, a series that yielded 16 cranial cases [9]. Frontal involvement accounted for 44.1% of 93 skull cases collected from reports published between 1845 and 2015 [4]. Neither is an incidence estimate, and neither should be presented as one.

Beyond these, the numbers thin quickly. Only four sources in this field report a cohort with a stated size: a systematic review of 77 patients drawn from 51 studies [10], a frontal-bone literature review of 51 patients [11], a surgical series of 16 [9] and a surgical series of 6 [12]. Everything else is a single report or a narrative synthesis [7,13,8]. There is no basis in this material for pooled

estimates, meta-analytic aggregation or comparative effectiveness.

Nomenclature, Pathology and the Instability of Case Tallies

Histologically, three patterns are described: cavernous, with clusters of dilated vascular spaces separated by fibrous septa; capillary, with numerous small lumina and little septation; and mixed. Skull lesions are predominantly cavernous, whereas vertebral lesions are more often capillary [4]. Intraosseous capillary haemangioma is exceptionally rare, with only 5 documented cases in the head and neck [11], and mixed cavernous and capillary histology has been reported in a frontal lesion [14]. Microscopically the cavernous channels are separated by residual trabeculae of cancellous bone, lined by a single layer of flat, bland endothelial cells without mitotic activity, engorged with erythrocytes, and lacking an elastic lamina on elastic staining [11]. Calcification within the lesion takes the form of amorphous deposits, phleboliths, which are the commonest, or rarely metaplastic ossification [12].

The terminology is unsettled and this has practical consequences. Under the ISSVA framework, haemangioma denotes a true vascular neoplasm, and what is called cavernous haemangioma is in most cases a venous malformation [15]. A reappraisal of 77 vascular lesions of bone in patients from 3 months to 25 years of age, applying ISSVA criteria, reclassified 46 as malformations and only 31 as tumours [16]. A dedicated review has argued the point directly for bony vascular anomalies [17]. The consequence is that the same lesion appears in the literature as primary intraosseous cavernous haemangioma, calvarial haemangioma, intraosseous cavernous malformation [9] and intraosseous vascular malformation [14]. Any tally built on one search term undercounts, which is the most plausible reason the published counts of 93, 77, 51 and 76 cases cannot be reconciled with one another.

Anatomical Distribution Within the Skull

The best-counted site distribution comes from a tally of 93 published skull cases: frontal 44.1%, temporal 12.9%, parietal 12.9%, occipital 11.8% and cranial fossa 5.4%, with the residual scattered across sphenoid, zygoma, ethmoid, clivus and orbital rim [4]. Frontal lesions outnumber parietal by roughly 3.4 to 1 in this material.

The ordering is nevertheless disputed. Several reports state that the frontal bone is commonest and the parietal second [3,14], one states the reverse [18], and a third ordering places temporal ahead of parietal [2]. Only the 93-case tally attaches a denominator to its ranking, and it is a count of publications rather than of patients. Frontal and large lesions are the ones most likely to be photographed, operated on and written up, so publication bias plausibly inflates the frontal share.

Age and Sex Across the Counted Cohorts

Four cohorts report age or sex against a stated N. The mean age was 32.7 years among 77 patients pooled from 51 studies, with a female-to-male ratio of 1.4 to 1 [10]. Restricted to lesions in or including the frontal bone, 51 patients reported between 1894 and December 2016 had a mean age of 37.6 years, with 31 of 51 women and no laterality preference; frontal lesions measured 1 to 6 cm, while parietal and occipital lesions reached 8 cm [11]. In a surgical series of 16, there were 10 women and 6 men, with a mean age at operation of 38.7 years and a range of 1.9 to 63 years [9]. A series of 6 operated over 8 years comprised 3 men and 3 women, of whom 3 were under 18 [12].

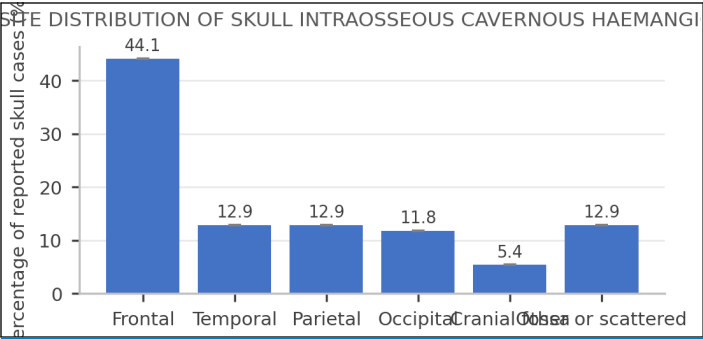


Figure 1: Site distribution of primary intraosseous cavernous haemangioma of the skull, from a pooled tally of cases published between 1845 and 2015, which counts reports rather than incidence [4] (N = 93).

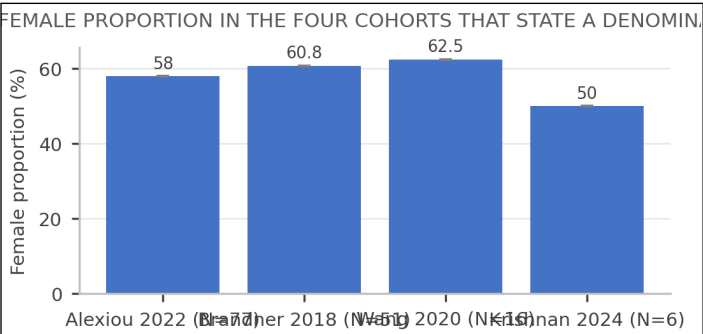


Figure 2: Female proportion in each of the four cohorts reporting a stated denominator, plotted separately because these overlapping reviews and institutional series must not be pooled [10,11,9,12] (N = 77, 51, 16 and 6).

These four are not poolable; they overlap and use different ascertainment. What they share is a modest female excess, between roughly 1.0 to 1 and 1.6 to 1. That is incompatible with the frequently quoted 3 to 1 female preponderance [2] and equally incompatible with the 2 to 1 or 3 to 1 male preponderance quoted elsewhere [4,19]. Neither of the steeper claims has a counted denominator behind it.

Imaging, Preoperative Diagnosis and Surgical Management

The clinical presentation is monotonous. A slowly growing, firm, immovable swelling is the commonest complaint [9], and in the verified frontal cases the history ran from a few months to ten years before presentation. Pain is variable, reported as dull facial and forehead pain in one case [11] and as intermittent headache in another with orbital roof and rim involvement [18], while other

patients presented for cosmesis alone [20]. Antecedent trauma is mentioned often enough to be recorded but rarely enough to remain unexplained: 2 of 6 patients in one series had a trauma history [12], and two published frontal cases describe growth beginning or accelerating after a head injury [21,3]. The great majority of intraosseous haemangiomas are asymptomatic and found incidentally, and those reaching publication are a selected minority.

Table 2: Published cases of frontal intraosseous haemangioma read for this review; not stated means the field was absent from the record, not that it was negative.

Author, year	Age, sex, site	Imaging; embolisation	Operation and reconstruction	Recurrence; follow-up
Nasrallah 2009	17 F, left frontal	Expansile, coarse trabeculations; MMA embolisation	Craniotomy, flap debulked, outer table preserved and replaced	None; 38 months
Park 2013	39 F, left frontal	Osteolytic, read as fibrous dysplasia; none	Bicoronal excision; split calvarial graft	None; 9 months
Bravo-Martinez 2019	58 F, bifrontal	CT sunburst, T2 high; MMA coils 2 days before	Bifrontal craniectomy with clear margin; cranioplasty	None; 5 months
Bird 2022	Adult F, right frontal, intradural	CT sunburst, 5.5 x 3.2 x 5.6 cm; considered, not done	Piecemeal debulking; dural patch, temporary titanium mesh	Not stated
Kim 2023	35 F, right frontal	Osteolytic, 1.1 x 0.7 cm; none	Partial resection via 3 cm hairline incision; none	None; 6 months
Lee 2024	59 F, left frontal	CT sunburst, 3.1 x 1.7 x 3.6 cm; none	En bloc, 0.5 cm margin; galeal flap and bone cement	None; 6 months
Gigov 2025	36 M, frontal	Dense mass about 3 cm; none	Re-resection; iliac crest autograft	Recurrence at 6 years

On plain films and CT the lesion is expansile, lytic and intradiploic, classically with radiating trabecular spicules described as a sunburst, sunray or spoke-wheel pattern, or as honeycombing [22,23,24]. No published series reports the proportion of lesions showing this sign against a

denominator; the sensitivity of the sunburst appearance has never been measured, and it should not be quoted as a percentage. Larger lesions display it more readily than small ones [12]. MRI signal varies with venous stasis and marrow composition, so that T1 may be high or low while T2 and FLAIR are commonly high [4,3]. A dural tail may be present and invites confusion with meningioma [25]; the 30% to 85% figure for the dural tail applies to meningioma and is itself described as unspecific [6]. Preoperative diagnosis is accordingly unreliable: calvarial haemangioma was suspected before operation in only 2 of 6 operated patients in one series [12], and a frontal lesion was reported by the radiologist as fibrous dysplasia [21]. Classical teaching that the inner table is spared is also imperfect, with inner-table violation found in 3 of 6 patients in one series [12] and frank intradural extension reported [26].

Surgical practice in the published frontal cases is summarised in Table 2. En bloc resection with a margin of normal bone is the dominant approach, with margins of 0.5 cm reported as sufficient and no recommended width established in the literature [11,4,14]. Among 51 pooled frontal cases, 4 were managed non-operatively and 5 had partial resection [11]. Reconstruction has used titanium mesh in 11 of 16 in one series [9], same-sitting cranioplasty in all 6 of another, with PMMA in 4 and mesh in 2 [12], and split calvarial autograft [21,18], bone cement [14] and iliac crest autograft [27] in single cases.

Preoperative embolisation is recommended by many authors for large or hypervascular lesions and considered unnecessary by others if an adequate bony margin is taken [11]. The feeders are usually branches of the middle meningeal, superficial temporal and posterior occipital arteries [3]. The evidence for benefit is not quantified: the only verified intraoperative blood-loss figure in this material is approximately 500 mL in a single non-embolised skull-base case [1], and no blood-loss volume is reported in any embolised case. No comparison

of blood loss with and without embolisation exists, and embolisation is not always safe, having been abandoned in one case because the dominant anterior falcine feeder anastomosed with the internal carotid circulation [26].

Recurrence, Follow-up and Unanswered Questions

The strongest recurrence datum is the absence of recurrence in 16 operated patients at a mean follow-up of 76.2 months, range 19 to 119 months, with complete resection achieved in 15 of the 16 [9]. Among 51 pooled frontal cases, 1 of the 5 who underwent partial resection required a further procedure [11]. Beyond these, the assertion that curettage carries a higher recurrence risk than en bloc excision is universal in the literature and unquantified: no comparative recurrence rate for curettage against en bloc excision has been published for any defined cohort.

The differential is broad and includes metastasis, meningioma, fibrous dysplasia, osteoma, eosinophilic granuloma and epidermoid, all of which occupy the same diploic space and several of which outnumbered haemangioma in the only counted calvarial operative series [6,13,23]. The practical consequence is that a solitary lytic frontal lesion in an adult may prompt a search for a primary tumour: staging imaging of chest, abdomen and pelvis was obtained before resection of one massive frontal lesion because metastasis remained in the differential [26], and elsewhere a calvarial haemangioma was misdiagnosed as skull metastasis from breast cancer and irradiated [20].

Follow-up duration is the more serious problem. The published frontal cases report follow-up of 6 to 38 months [20,14,21,18,28], while a frontal recurrence has been documented presenting 6 years after the index resection [27]. Reassurance drawn from a 6-month or 12-month scan therefore rests on an interval shorter than the demonstrated time to recurrence.

Conclusion

Intraosseous haemangioma of the frontal bone is a benign, slowly enlarging diploic lesion, usually cavernous, best understood in modern terms as a venous malformation. En bloc excision with a small margin of normal bone and immediate reconstruction is the treatment most often reported and is followed by recurrence in a small minority. The quantitative scaffolding around these statements is weaker than the confidence with which it is usually repeated. The rarity figures have no traceable denominator; the sex ratio, the site ranking and the value of embolisation are all asserted more firmly than the counts allow; and the follow-up in the published record is shorter than the interval over which recurrence has actually been seen. Reporting future cases with dimensions, margins, blood loss and follow-up stated in full would do more for this literature than another repetition of the same uncounted percentages.

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