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CASE REPORT Open Access

Intraosseous cavernous hemangioma: presentation of a clinical case

Alejandra Arévalo Sáenz[1,2*], Natalia Frade Porto[1] and Manuel Pedrosa Sánchez[1]

Abstract

Background: Cavernous hemangiomas are benign tumors that exceptionally affect the cranial bones. The first description of this type of tumor was in 1845 by Toynbee. A review of the literature reveals less than 100 published cases and a growing trend every year. Total surgical excision is the treatment of choice, and the prognosis after complete excision is excellent, with a recurrence usually rare.

Case presentation: We present the case of a 57-year-old patient with a painless tumor of the left frontal bone, of slow growth and osteolytic characteristics from the neuro-radiological point of view. The lesion was excised en bloc by craniectomy, followed by cranioplasty. The anatomopathological diagnosis was intraosseous cavernous hemangioma.

Conclusions: Despite its low frequency, the diagnosis of intraosseous cavernous hemangioma should be considered in the presence of a slow-growing cranial tumor, with solid and painless characteristics, and its osteolytic nature confirmed by radiology. The treatment of choice consists in the complete resection of the lesion.

Keywords: Skull base, Craniectomy, Intraosseous cavernous hemangioma, Osteolithic, Benign tumor

Background

Primary intraosseous cavernous hemangiomas (PICHs) are a rare and infrequent tumor representing 0.7 to 1.0% of all bone tumors [1]. PICHs are usually found in the spine and rarely appear in the vault cranial, being 0.2% of cranial bone tumors [2]. The first description of this type of tumors was in 1845 by Toynbee. A review of the literature reveals about 100 published cases and a growing trend every year (Table 1) [3–77]. These tumors are seen mostly in middle age, with a peak around the fourth decade and a male/female ratio that ranges between 3:1 and 2:1 [2].

Case report

We are dealing with a 57-year-old patient, with no history of interest, referred to our Service for surgical assessment. It had presented, for 4.5 years, a small tumor in the left front region of 1.5 cm in diameter, which had slowly increased in size (Fig. 1c, d). During the examination, a mass of solid and hard consistency, painless and adhered to deep planes, was palpated under a normal-looking skin. The plain radiograph (Fig. 1a, b) and the CT (Fig. 1d) showed a left frontal intraosseous lesion with osteolytic characteristics with moderate aggressiveness. The radiological differential diagnosis included bone metastasis or plasmacytoma. The systemic studies of tumor tracking (blood count, hematological smear, tumor markers, proteinogram, and cervical-thoraco-abdominal CT) were negative. The percutaneous puncture with fine needle of the tumor was inconclusive for the diagnosis; only blood fragments were obtained. Finally, it was decided to surgically intervene the patient based on the clinical progression of the lesion, with its esthetic implications, as well as to obtain a definitive histological diagnosis. During the surgery, a bone-dependent tumor was identified, with multiple dilated vascular channels in its center, which expanded the external table. To avoid manipulation of the lesion, it was decided to include it in a piece of craniectomy with a circumferential margin of 1 cm of the seemingly healthy bone. The resulting bone defect was reconstructed by means of a cylindrical metametacrylate plasty, which was fixed to the surrounding bone with titanium miniplates. The postoperative period was uneventful. The definitive anatomopathological diagnosis was intraosseous cavernous hemangioma.

* Correspondence: praimale@hotmail.com
[1]Neurosurgery Service of the University Hospital of La Princesa, C/ Diego de León 62, 28006 Madrid, Spain
[2]Division of Neurosurgery, University Hospital La Princesa, C/ Diego de León 62, 28006 Madrid, Spain

Table 1 Review of the literature from 1845 to 2016 of the published cases of intraosseous hemangiomas

Frontal			Temporal	Occipital	Parietal	Skull base	Otros
Pilcher (1894) [3]	Relf et al. (1991) [17], × 2	Carrasco et al. (2009) [30]	Sargent et al. (1965) [38]	Peterson et al. (1992) [2]	Toynbee et al. (1845) [54]	Jackson et al. (1980) [64]	Chaterji et al. (1969) [66]
Wyke (1949) [4]	Peterson et al. (1992) [2]	Roel et al. (2012) [31]	Mangham et al. (1981) [39] × 3	Cervoni et al. (1995) [18]	Kumar et al. (1993) [55]	Glasscock et al. (1984) [40]	Schofield (1950) [67]
Gupta et al. (1975) [5]	Cervoni et al. (1995) [18], × 2	Park et al. (2013) [1]	Glasscock et al. (1984) [40]	Corr (2000) [48]	Yoshida et al. (1999) [56]	Mazzoni et al. (1988) [42]	Dickins (1978) [68]
McIntyre et al. (1997) [6]	Pastore et al. (1999) [19]	Xu et al. (2013) [32]	Suss et al. (1984) [41]	García-Marín et al. (2001) [49]	Heckl et al. (2002) [22]	Bottrill and Poe (1995) [65]	Inoue et al. (1982) [69]
Gross and Roth (1978) [7]	Sharma et al. (1999) [20]	Uemura et al. (2014) [33], × 2	Mazzoni et al. (1988) [42]	Khanam et al. (2001) [50]	Ajja (2005) [57]	Khanam et al. (2001) [50]	Suss et al. (1984) [41]
Fouad et al. (1979) [8], × 2	Suzuki et al. (2001) [21]	Murrone et al. (2014) [34]	Buchanan et al. (1992) [43]	Heckl et al. (2002) [22]	Paradowski et al. (2007) [58]		Tashiro et al. (1991) [70]
Shinno et al. (1986) [9]	Heckl et al. (2002) [22]	Chun et al. (2015) [35]	Fierek et al. (2004) [44]	Buhl et al. (2007) [26]	Naama et al. (2008) [28]		Slaba et al. (1999) [71]
Hook et al. (1987) [10]	Pottelbergh et al. (2004) [23]	Hsiao et al. (2015) [36]	Sasagawa et al. (2009) [29]	Gibson and Prayson (2007) [51]	Sasagawa et al. (2009) [29] × 2		Moore et al. (2001) [72]
Zucker et al. (1989) [11]	Politi et al. (2005) [24]	Yi Yang (2016) [37]	Silva et al. (2013) [45]	Baltazar et al. (2008) [52]	Rumana et al. (2013) [59]		Liu et al. (2003) [73]
Hoffmann et al. (1990) [12]	Cheng et al. (2006) [25]		Yang et al. (2014) [46]	Nair et al. (2011) [53]	Atci et al. (2013) [60]		Jeong and Rhee (2006) [74]
Hornig et al. (1990) [13], × 2	Buhl et al. (2007) [26]		Yetiser et al. (2014) [47]		Hsiao et al. (2015) [36]		Salunke et al. (2010) [53]
Sinnreich (1990) [14]	Nasser et al. (2007) [27]				Kilani et al. (2015) [61]		Moravan et al. (2011) [76]
Aurora et al. (1991) [15]	Naama et al. (2008) [28] × 2				Sarmast et al. (2016) [62]		Yu et al. (2014) [77]
Faerber and Hiatt (1991) [16]	Sasagawa et al. (2009) [29]				Brichacek et al. (2018) [63]		

This table shows all the cases published in the literature since 1846, making a difference between locations. The most frequent are the frontal ones, as our case. Other sites indicate the case where the lesion was located in sphenoid, zygomatic, ethmoid, clivus, orbital arch, etc. × 2 or × 3 refers to the number of cases published by the author

Discussion

The first case of cranial cavernous hemangioma was described by Toynbee in 1845 [54]. Since then, most of the publications in the scientific literature have been presented in a single clinical case format, with the exception of two reviews of extensive casuistry that constitute the main references on this entity [22]. PICHs of the skull are rare benign vascular tumors, accounting for about 0.2% of all tumors and 10% of benign tumors of the skull [59]. They occur most frequently in the spine and rarely in the skull. Of the 93 cases of cranial PICH reported in the literature from 1845 to 2015, 44.1% were located in the frontal bone, 12.9% in the temporal bone, 11.8% occurred in the occipital bone, 12, 9% in the parietal, and 5.4% in the cranial fossa; fewer cases have been reported in sphenoid, zygomatic, ethmoid, clivus, and orbital, etc. [37]. In the review carried out by Wyke, this distribution is supported [19].

They are usually unique lesions, although cases of multiple cranial cavernomas have been described [28]. They usually have a size at the time of diagnosis that ranges between 15 and 25 mm, although lesions of up to 8 cm diameter have been described [78]. Its origin is in the vessels of the diploic space, and their blood supplies are branches of the external carotid artery. The middle and superficial temporal arteries are the main sources of blood supply. Within the lesion, the capillaries are widely dilated and separated by fibrous tissue [77]. Its pathogenesis remains unknown. It was believed that it could be congenital, but this has not yet been proven. A previous trauma could also be an important etiology to consider [77]. The typical presentation is given by the presence of a hard, painless mass that slowly increases in size under an overlying intact skin. Sometimes they are associated with headache, which can be of high intensity when the hemangioma expands [79]. The most common clinical feature is a solid tumor in the skull, painful or painless.

The cranial CT with a bone window is the diagnostic modality of choice, since it surpasses the sensitivity of simple radiography and allows bone to be defined in a superior way to MRI, giving a detailed image of the cortical and trabecular bone. Although the appearance in the CT can vary, the characteristic image consists of a

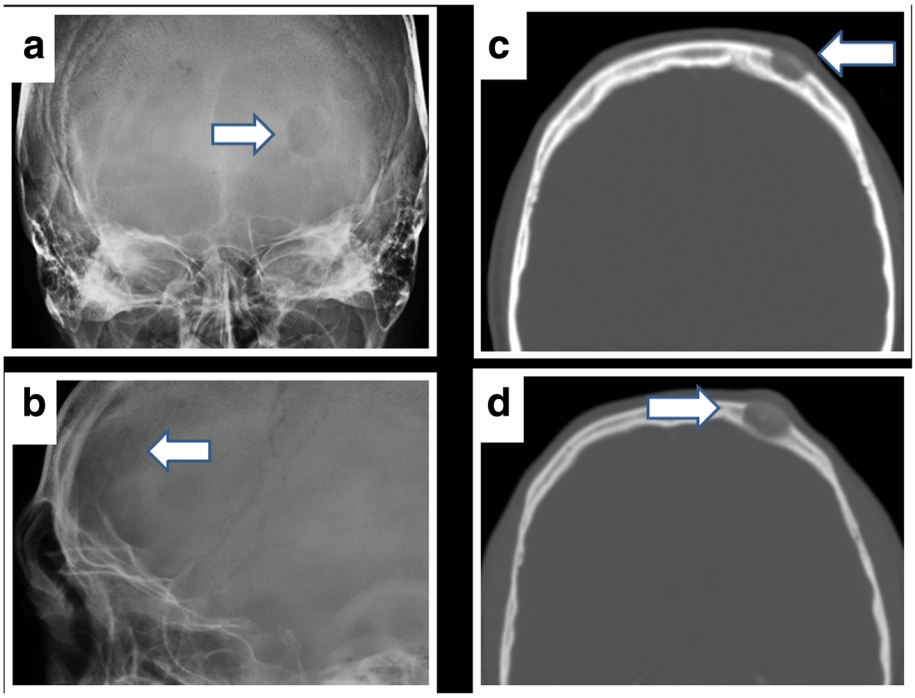

Fig. 1 Radiological images of a left frontal intraosseous lesion. **a** Simple X-ray of the skull, in the antero-posterior view and **b** in the lateral view of a radiolucent lesion (white arrows), rounded, with well-defined edges, located in the left frontal bone. **c** First CT cerebral with bone window performed on the patient 3 years ago. In localization of the left frontal region, a lytic bone lesion is observed that discreetly expands the diploe both in its internal and external table, with well-defined contours in its medullar region, being worse defined there could even be minimal solutions of continuity in the internal table. A poorly defined trabecular bone pattern was observed inside the lesion. The findings are compatible with a moderate aggressiveness. **d** In the last CT cerebral, it was appreciated how the lesion had increased in size. Produces a slight insufflation of the frontal calotte with loss of the cortex of the external table. Its behavior is of low radiological aggressiveness

lytic lesion, oval or rounded, expansive, and well delimited, with trabeculae that radiate from a common center in its interior in the tangential cuts, giving sometimes an appearance of honeycombing in the axial cuts [21]. It frequently invades and expands the external table, respecting the periosteum. Usually no signs of reactive hyperostosis are identified at their margins [22]. The cortex can undergo a great expansion leaving a thin bone layer, but in almost all cases, the periosteum remains intact and usually without reactive sclerosis at the margins. The angiography of these lesions, typically the largest, demonstrates a hypervascular lesion, but without drainage veins. Preoperative embolization may be useful in some cases [80]. The differential diagnosis includes other slow-growing, expanding bone lesions, such as osteoma, aneurysmal bone cyst, giant cell tumor, and multiple myeloma [81]. The radiological characteristics that help the differential diagnosis are shown in Table 2.

The natural history of these pathologies has not yet been described. Considering that osseous cavernous hemangiomas grow progressively, without spontaneous involution, their surgical treatment is usually recommended for several reasons: progression of the painful clinic, cosmetic implications, and, although with low frequency, avoidance of complications such as hemorrhages or nerve damage cranial, depending on the location of the lesion [22]. In 1923, Cushing designed the one that represents the treatment of choice of cranial cavernous hemangiomas to the present day: en bloc resection of the lesion, including a circumferential margin of the healthy bone [11]. On the other hand, the possibility of recurrence is avoided by including a margin of safety [77]. Most authors recommend total surgical excision to treat the mass effect and neurological compromise, to improve an esthetic deformity, and to obtain a definitive diagnosis [11]. The surgical approach becomes more difficult for those with extension to the base of the skull. Radiotherapy should be reserved for those lesions that are considered unresectable or in the case of recurrent tumors. This therapeutic modality stops tumor growth and reduces vascularization, but does not modify the size of the lesion and carries the risk of malignancy or the appearance of de novo malignancies [39].

Table 2 Differential diagnosis of skull vault lesions

Lesion	Clinical features	Radiology	Treatment
Osteoma	Osteomas are slow-growing lesions that are normally completely asymptomatic. A few may be associated with Gardner syndrome	CT: small, well-defined round, or oval dense and homogeneous lesions; homogeneous low signal intensity on T1WI; variable appearance on T2WI; not enhance after gadolinium administration	Not require surgical treatment unless the location or size of the lesion affects the adjacent structures (orbit, sinus, brain)
Aneurysmal bone cyst	Mainly in children and adolescents; may be secondary to other underlying lesions like fibrous dysplasia, chondroblastoma, and osteosarcoma	Sharply defined expanded osteolytic lesion with thin sclerotic borders, although the tables appear disrupted when the expansion is significant	The traditional treatment is complete surgical excision
Myeloma	Bone pain, deterioration of health, or abnormalities on blood or urinary test (e.g., high erythrocyte sedimentation rate, anemia)	Multiple small, roundish osteolytic lesions that are relatively uniform in size with sharp and non-sclerotic margins. On MRI, the signal intensity of the lesions is nonspecific; a "salt and pepper" appearance or diffuse bone marrow replacement may be noted	Treatment depends on the stage of the disease. The most common treatments are based on chemotherapy or grafting of hematopoietic cells
Langerhans histiocytosis	Clinical features are variable, from asymptomatic lesions to painful swellings	CT: unequal involvement of the inner and outer tables; appearance of having beveled edges. The lesion center may contain a sequestrum, representing residual intact bone. MRI: usually strongly enhance after gadolinium administration	Single lesions: conservative treatment (surveillance or systemic corticosteroids). More diffuse or aggressive forms: surgical excision, radiotherapy, and chemotherapy
Skull metastasis	Usually secondary to the breast, lung, prostate, kidney, and thyroid cancer; generally asymptomatic; may be revealed by a painful swelling	Mostly multiple, well circumscribed osteolytic lesions, which generally extend into the adjacent soft tissues. Usually homogeneously enhanced on enhanced MRI, but heterogeneous enhancement, peripheral ring enhancement, or lack of enhancement (sclerotic lesions) can be observed	Surgical treatment may be possible when there is only 1 metastasis, especially if without any neoplastic context. Radiotherapy is another alternative
Intraosseous meningioma	Predominantly seen in women in the fifth and sixth decades of life and often revealed by painless and expanded swelling	CT: osteosclerotic lesion with destructive irregular and spiculated borders. Low signal intensity on T1WI; variable signal intensity on T2WI; not enhance. Meningeal enhancement is rare and is explained by adjacent dural irritation or invasion, but the center of the tumor growth is outside the dura	Surgical resection of the lesion is required. The therapeutic decision depends on the possibility of resecting the lesion and on the patient's health
Epidermoid and dermoid cyst	Painless subcutaneous swelling; discovered mainly during the third and fourth decades; predominantly occur laterally in the parietal or frontal bone	CT: well-demarcated osteolytic lesions with sclerotic borders; may tend to expand into both the inner and outer tables; homogeneously hypodense. MRI: fluid-like signal intensity on T1WI and T2WI and high signal intensity on DWI; usually do not enhance	Treatment of these cystic lesions is surgical, usually without recurrence

This table shows the main clinical and radiological characteristics of possible differential diagnoses

Conclusions

Cranial cavernous hemangiomas are bony tumors of nature, which, in the absence of typical radiological features, are usually surgically treated under suspicion of another type of bone neoplasm. The treatment of choice is a complete resection by craniectomy, including healthy bone margins of safety, with good prognosis and little recurrence. In the presence of subtotal resections or progression, radiotherapy could be a valid option.

Abbreviations

CT: Computed tomography; DWI: Diffusion-weighted imaging; MRI: Magnetic resonance imaging; PICHs: Primary intraosseous cavernous hemangiomas; T1WI: T1-weighted imaging; T2WI: T2-weighted imaging

Availability of data and materials

Please contact the author for data requests.

Authors' contributions

The individual contributions of authors to the manuscript are the following: AAS has made the elaboration of the manuscript, the description of the clinical case, the review of the literature and the surgery of the presented case. NFP has made the elaboration of the images and the table. MPS has made the revision of the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient/parent/guardian/relative of the patient.

Competing interests
The authors declare that they have no competing interests.

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