Intraosseous cavernous haemangioma of the nasal bone: A case report and literature review

Nika Vafaei[1] and Carlo Ferretti[2]

Abstract

Intraosseous haemangioma is very rare, accounting for less than 1% of all osseous tumours, and it is rarer still in the nasal bone. To the best of our knowledge only 33 cases have been reported in the literature to date. Consequently it is a differential diagnosis that is often overlooked. Clinicians should be familiar with the clinical features and radiologic appearance of this tumour in order to include the diagnosis in a differential. Intraosseous haemangioma requires excision, and if necessary reconstruction depending on the extent of osseous destruction. We present a review of the literature and add a case report of the presentation, treatment and reconstruction of an intraosseous cavernous haemangioma occurring in the nasal bridge.

Key words: Haemangioma; intraosseous; nasal bone.

Introduction

Haemangioma is a benign vascular tumour, believed to be a hamartoma, mesenchymal in origin.[1,2] There are 2 types of haemangiomas, capillary and cavernous, the capillary haemangioma being more common. Haemangioma of bone is uncommon, and is exceedingly rare in the nasal cavity, and when it occurs, it is predominantly in the septum.[2] The first case report of a haemangioma occurring in the nasal bone was described by Neivert and Bilchik in 1936.[3] A review published in 1976 reported 17 cases and a subsequent review in 1992 added a further 9 cases.[2,4] Since then we have found a further 7 cases reported in the literature (Table 1).[2-11] Cavernous haemangioma that occurs in the nasal bone presents as a slow growing bony hard mass covered by mucosa within the cavity. The adjacent tissues are usually uninvolved.[7] Cavernous haemangioma of the nasal cavity can present with a history of recurrent epistaxis and nasal obstruction, however this is uncommon with those occurring in the nasal bone. The radiographic appearance of nasal bone tumours is often "sun-burst" due to thickened linear trabeculations which radiate from a central radiolucent core.[4] The computed tomography (CT) appearance of these lesions is variable with some appearing as a heterogenous soft tissue mass, whilst others as a solid homogenous mass filling the nasal cavity.

For soft tissue haemangiomas various treatments have been proposed, including radiotherapy, embolization, cryotherapy, corticosteroid treatment, sclerosing solutions and resection using YAG laser.[12] For intraosseous haemangioma, surgical resection of the tumour with a cuff of surrounding uninvolved tissue has been found to be most effective. The role of preoperative angiography and embolization is controversial in intraosseous haemangiomas, as often a definite blood supply cannot be identified, and the benefits of embolization are minimal when compared with the potential risk.[13]

We present a case of intraosseous cavernous haemangioma occurring in the nasal

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Table 1. Summary of reported cases (after McAllister 1992)

Case#	Author	Age	Sex	Duration	Trauma	Follow Up
27	Karacoaglan, N et al 1997	19	F		No	1 yr
28	Adanali 2001	62	F			1 yr
29	Hazra, T.K. 2001	16	F		No	13 yrs
30	Layoun, W 2003				Yes	
31	Kargi, E 2005	60	M		Yes	1 yr
32	Stankovic, M 2008	32	F		No	1 yr
33	Cintra, BB 2008	49	M		No	14 mnths

bone treated by resection and reconstructed with a diced cartilage filled fascial pouch inserted between 2 struts of rib graft.

Case Report

A 67 year old female presented with a slow growing swelling on the bridge of the nose (Figure 1a & b). She first noted it 5 years ago, and she could not recall an injury which may have been causally related. She had the lesion debulked 4 years ago. Recently the swelling started increasing in size resulting in significant deformity over the nasal bridge; however no nasal obstruction or epistaxis was reported. Examination revealed a diffuse swelling on the bridge of the nose measuring approximately 2cm x 1.5cm. The overlying skin was normal in colour and texture, with a vertical scar down the bridge of the nose from the previous surgery. The swelling was non-tender, bony hard and well circumscribed. No neurological fall out was noted. Computed tomography revealed a heterogeneous mass approximately 3cm in diameter with osseous expansion of the nasal bridge, resulting in significant thinning of the outer cortex. The lesion appeared as a mass with trabecular coarsening that radiated in a spoke-wheel type pattern (Figure 2a & b).

Differential diagnosis included osteoma and fibrous dysplasia, although the site and age of the patient was uncommon for both lesions. A more common swelling on the midline dorsum of the nose is a dermoid cyst, however these lesions occur more frequently in a younger age group and present with a tell-tale 'pit' in the overlying skin.[2] Other differentials included parosteal osteosarcoma based on the radiographic "sun-burst" appearance; however the indolent

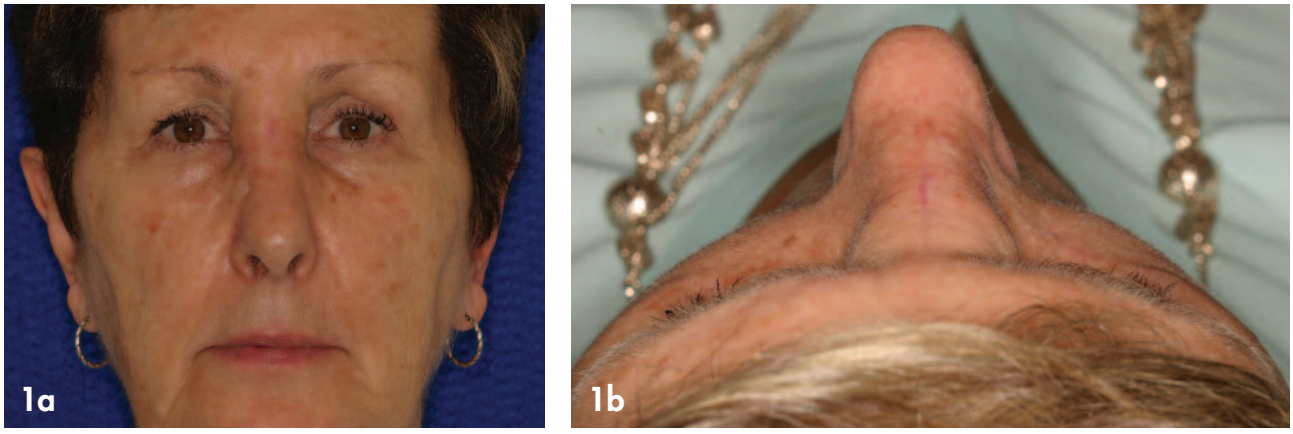


Figure 1a & b: Pre-operative photo showing frontal and superior view of diffuse swelling of nasal bridge.

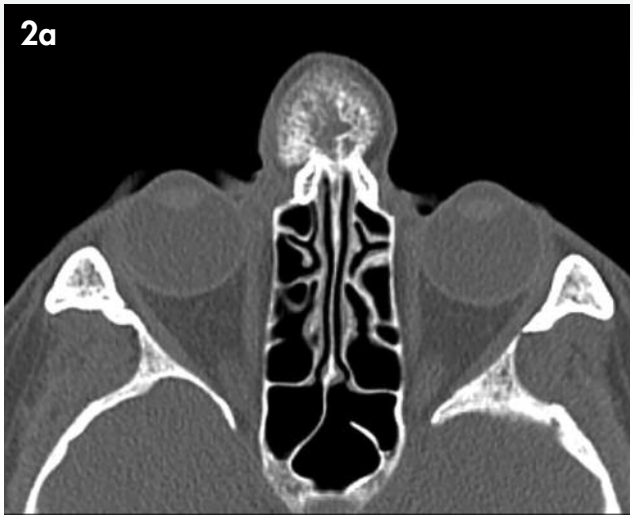


Figure 2a: Coronal cut of CT scan showing expansile osseous lesions with a 'sunburst' appearance, in the nasal bone.

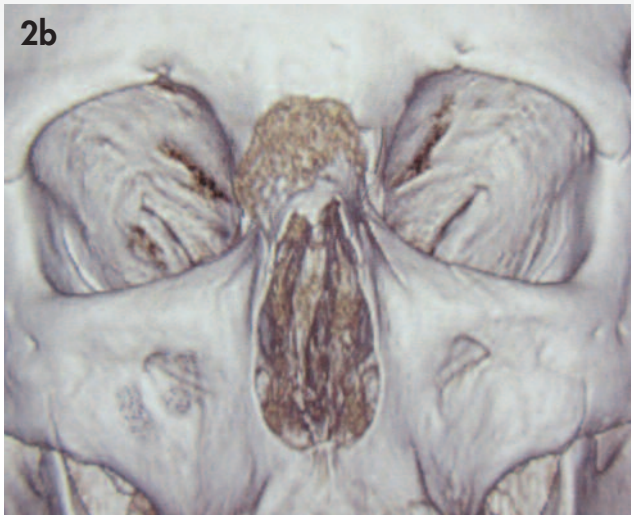


Figure 2b: 3D reformatted CT of lesion showing surface of nasal bridge expanded and pitted by expansile lesion.

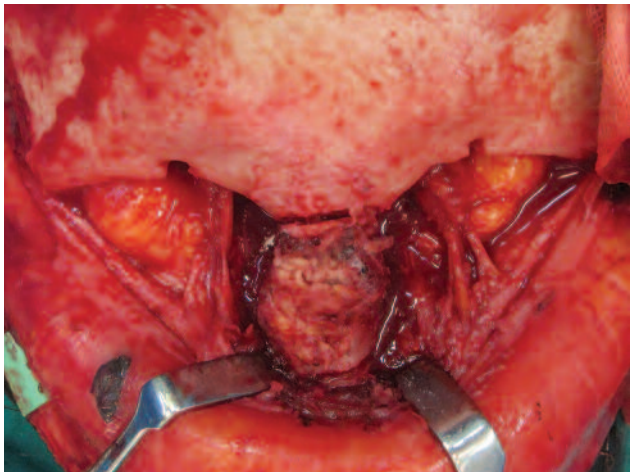
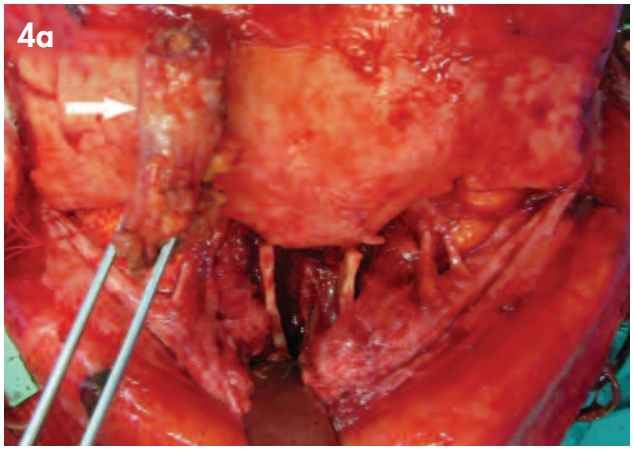


Figure 3: Lesion exposed via bicoronal flap.

behaviour of an intraosseous haemangioma is inconsistent with such a diagnosis, as was the patient's age. In addition, the intact periosteum is an important feature in distinguishing this benign neoplasm from an osteosarcoma in which there is a breach in the periosteum, and likely invasion of the surrounding soft tissues. A definitive diagnosis of intrabony haemangioma required histological examination of an incisional biopsy.

An incisional bone biopsy was performed under general anaesthesia via a midline incision over the bridge of the nose. Brisk bleeding was encountered and controlled by local methods. The biopsy confirmed the diagnosis of an intraosseous cavernous haemangioma. Definitive treatment was en bloc resection of the tumour with concurrent



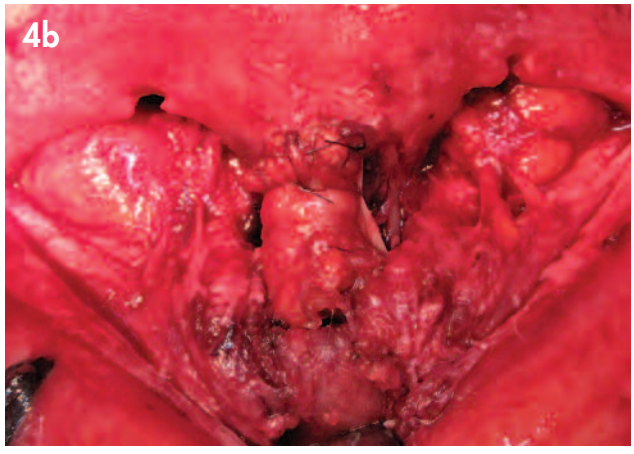
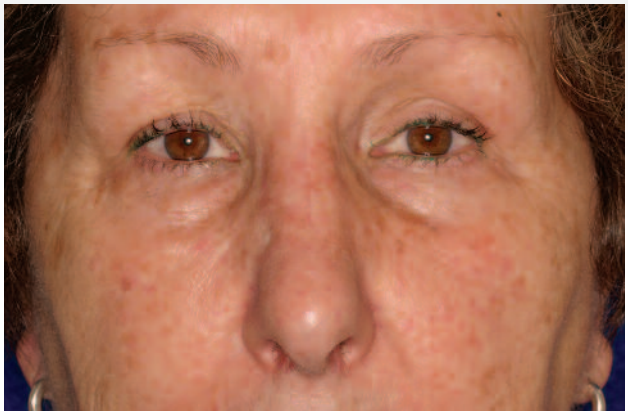


Figure 4a: Extant nasal bridge with cartilage struts in situ. Diced cartilage fascial pouch (arrow) to be secured between cartilage struts. Figure 4b: Cartilage filled fascial pouch secured to adjacent remaining nasal bone.



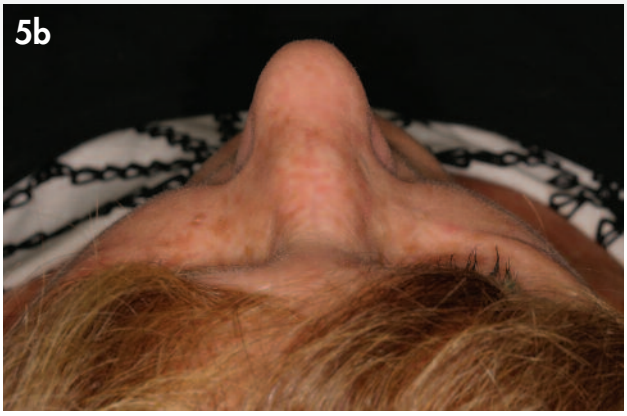


Figure 5a & b: Frontal and superior view 18 months post-operative.

reconstruction. The lesion was exposed via a bicoronal flap and, as planned, resected with a small margin of uninvolved bone (Figure 3). Resection was not accompanied by vigorous bleeding. The right 6th costal cartilage was harvested with a portion of pectoralis fascia. Two struts were cut from the costal cartilage and the remaining cartilage was morselized. The fascia was wrapped around a 5cc syringe and the free end sutured to form a pouch into which the morselized cartilage was inserted (Figure 4a). The 2 struts were secured to the extant nasal bone bilaterally, and the cartilage filled fascial pouch was placed between the 2 struts to form the bridge of the nose and secured with sutures (Figure 4b).

Histological examination of the resected mass showed numerous large cavernous vascular channels interspersed between the bone trabeculae. There was extensive destruction of the bone. The vascular channels were lined by flattened endothelial cells and were filled with red blood cells.

The patient has been followed up for 18 months and post-operative course has been uneventful. The nasal bridge was successfully reconstituted and maintained (Figure 5a & b).

Discussion

Haemangioma of the nasal bone is very rare. To the best our knowledge only 33 cases have been reported in the literature to date. Although many patients have a history of local trauma, the causal relationship remains doubtful. Despite being uncommon, the clinical and radiographic features of nasal bone haemangioma are fairly characteristic. Clinically these tumours appear as a slowly enlarging painless mass at the base of the nasion. These tumours may result in local discomfort, but airway obstruction or epistaxis are usually absent.[2,4] From the previous case reports and reviews, it occurs more frequently in females with a female to male ratio of 2:1, and the mean age at diagnosis of 39 ±12 years (range: 16-62). These bony tumours are distinct from soft-tissue haemangioma, as the intranasal mucosa is generally intact, as is the nasal passage and overlying skin. The radiographic features are characteristic as a translucent area within the nasal bone in which spicules of bone radiate outwardly from the central area.,[2,4] CT scan aids in determining delineation, remodelling, and destruction of bone trabeculae. Although the "sun-burst" appearance may give the impression of a more aggressive neoplasm, the clinical history would often preclude such a diagnosis. Definitive diagnosis can be made histologically based on the incisional biopsy.

Choice of the surgical approach for tumour resection depends on the location and size of the tumour. Reconstruction is required if the size of the post resection defect will leave an unacceptable cosmetic deficit. The reconstruction of the bridge of the nose may be accomplished by several means. The technique using diced cartilage graft for nasal rhinoplasty and reconstruction has been described and used successfully since the 1960's.[14] The advantages of using this method of reconstruction is its ease of preparation, lack of rejection as it is autogenous, and easier graft manipulation to appropriate shape. Technical problems with this method include overcorrection, visibility of graft, and junctional step-offs.[14]

Given the rarity of these lesions, as well as the limited differential diagnoses of a slow-growing bony mass in the nasal bones, clinicians should bear in mind the possible

diagnosis of an intraosseous haemangioma. Contrast CT would assist to determine the nature of the lesion in order to identity high or low flow lesions.

In conclusion, intraosseous haemangioma should be considered as a differential diagnosis when a slow growing, bony mass is noted in the nasal bone. Treatment of choice is surgical resection, and reconstruction with autologous grafting for nasal defects, yields good functional and aesthetic results.

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