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Cavernous haemangiona of the nasal bones

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Abstract
A case report of a cavernous haemangioma arising in the nasal bones is described, together with a discussion of the relevant literature. The condition, although rare, can be reliably diagnosed pre-operatively.

Introduction

Cavernous haemangioma arising in the nasal bones is rare, with only 25 cases recorded in the world literature to date (Dorfman *et al.*, 1971; Bridger, 1976; Zizmor *et al.*, 1978; Schvarcz, 1979; Kanter *et al.*, 1985; Kaplan *et al.*, 1991). We report a further case together with a review of the condition, and its diagnosis, pathology and management.

Case report

A 56-year-old male gardener presented with a spherical mass 2 cm in diameter on the right side of the bridge of his nose. Six years previously he had struck his nose with a rake and initially complained of swelling and tenderness which resolved to leave a shallow fossa. This persisted for two years, when a firm smooth swelling was noticed at the site. This gradually increased in size, particularly in the six months prior to presentation.

On examination, there was a soft, cystic, compressible lesion on the right side of the nose, with a palpable bony excrescence at its margin (Fig. 1). A bruit was audible within it. The lesion was not tender. It partially obstructed the visual field of the right eye. Radiographs of the region show a local bony erosion, with a reticular appearance within the lesion suggestive of spicule formation, together with bony thickening at its margin with nasal bones (Figs. 2 & 3). Examination with a doppler ultrasound probe demonstrated pulsatile vascular flow within the lesion.

At operation, the mass was exposed using a transverse incision; an intra-nasal muscosal extension was apparent. The lesion, its bony crater and its mucosal extension were excised. Septal mucosa, a free bone graft and a glabellar flap were used to close the defect, with a good cosmetic result.

Histopathological examination of the 20 × 15 × 12 mm specimen revealed an ellipse of skin and subcutaneous tissue with underlying bony tissue whose cut surface demonstrated multiple dilated blood filled spaces giving a 'honeycomb' appearance (Fig. 4). Microscopically there were multiple thin walled cavernous blood filled vessels lying between thin trabeculae of lamellar bone, with minimal extension into subcutaneous tissue (Fig. 5). The overlying skin was unremarkable. Histological and immuno-histological stains confirmed the lesion to be a cavernous haemangioma of the bone.

Discussion

Haemangiomas of bone are rare, accounting for 0.7 per cent of primary bone neoplasms (Bridger, 1976; Dahlin, 1986). Most are solitary lesions, and of the 80 patients with solitary haemangiomas in the series reviewed by Dahlin (1986), 53 were in the cranium or vertebrae, 11 in the maxilla or mandible, and the remainder were in the limbs or ribs. None were found in the nasal bones. In a series of 45 patients with haemangiomas of bone, Sherman and Wilner (1961) found one in the nasal bones. Osborn (1959) reviewed over 50 patients with haemangiomas of the nose seen over an 11 year period, and none involved the nasal bones. There have been only 26 nasal bone haemangiomas recorded in the world literature to date, including the present case. The first was reported by Nievert and Bilchick (1936) and a

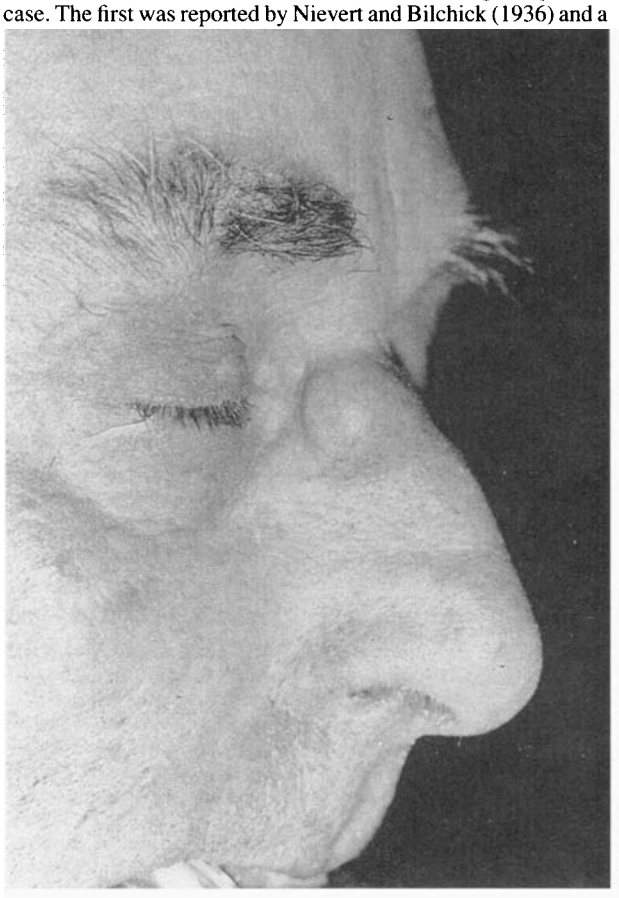

Fig. 1
Pre-operative appearance of cavernous haemangioma of the nasal bones.

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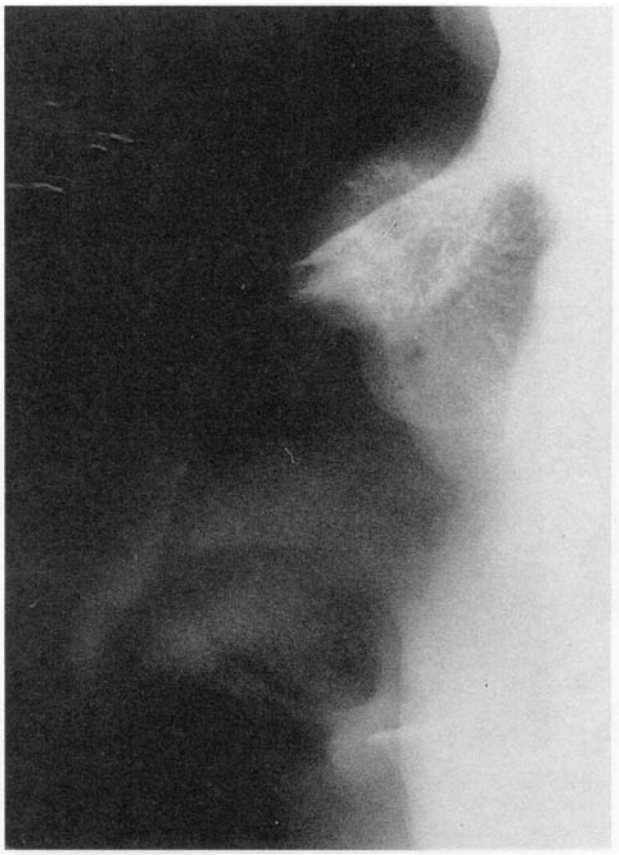

Fig. 2

Anteroposterior skull X-ray demonstrating the lesion associated with the right nasal bone.

systematic review was undertaken by Bridger (1976) of the 17 cases reported by that time. Table I summarizes the patients reported since then.

Clinical features

Nasal bone haemangiomas characteristically present as a slowly enlarging mass at the root of the nose, and usually reach between 1 cm and 2 cm in size at diagnosis. The tumour was unilateral in 11 cases (four on the left and seven on the right), mid-line in five cases, and unspecified in nine. Local discomfort is frequently but not invariably present, particularly on palpation. Symptoms of nasal airway obstruction or epistaxis are usually absent, and have been described in only one patient (Zizmor *et al.*, 1978). There are 15 females among the 24 patients whose sex is recorded in the literature, giving a female to male ratio of nearly 2:1 which is similar for haemangiomas of different sites (Dorfman *et al.*, 1971). The age at diagnosis was available for 23 of the 26 known cases, with a mean age of 42.7 ± 11.3 years (SD) and a range from 14 to 59 years. The duration of the symptoms until presentation ranges from two months to eight years, with a mean of 2.3 years among the 17 patients in whom it is specified.

Physical examination reveals a bony swelling at the bridge of the nose, usually with uninvolved mobile overlying skin. The patient whom we present had a centrally compressible area, which refilled on release of pressure, surrounded by a bony crater. The lesion also had an intra-nasal muscosal extension which has been described in only two other cases (Zizmor *et al.*, 1978); more usually the mucosa and nasal airway are not involved.

Investigations

The X-ray appearances of the lesion are characteristic, with thickened linear trabeculations which may radiate from a central core, within an oval radiolucent area, giving a 'sunburst' pattern (Griffith, 1967). A 'honeycomb' appearance of trabeculation is frequently seen. These features have been described on CT scans of the tumours (Kanter *et al.*, 1985), where a typical 'polka dot' pattern may also be observed. CT scans may be helpful in defining the extent of the haemangioma, and in demonstrating the presence of intact soft tissue planes and periosteum. Doppler ultrasound may assist in the diagnosis of a haemangioma by confirming the presence of pulsatile vascular flow within it.

Differential diagnosis

The combined clinical presentation and radiological findings in haemangioma of the nasal bones are sufficiently characteristic to allow pre-operative diagnosis to be accurate. However other lesions may present a similar clinical picture. These include nasal dermoid cysts, distinguished by skin dimpling on physical examination, and which are without radiological features; and sebaceous cysts which have a punctum and are invariably intradermal and not attached to underlying structures. Malignant melanoma of the nasal mucous membrane or carcinoma of the ethmoid sinus may cause a swelling in the rest of the nose but are likely to be associated with additional clinical features. Osteosarcoma can rarely present as a swelling of the nasal bridge, but is distinguished radiologically by disruption of the periosteum. Fibrous dysplasia may present similarly, but has sharply demarcated bony inclusions and an 'eggshell'-like capsule on plain X-ray (Kanter *et al.*, 1985). Primary nasal meningioma can present as a slowly progressive tumour expanding the nasal bone and frontal process of the maxilla; nasal polypi, or rare nasal gliomas in neonates and infants may produce similar clinical appearances (Bridger, 1976). Giant cell tumours of the nasal bones and ossifying fibromas are other differential diagnoses.

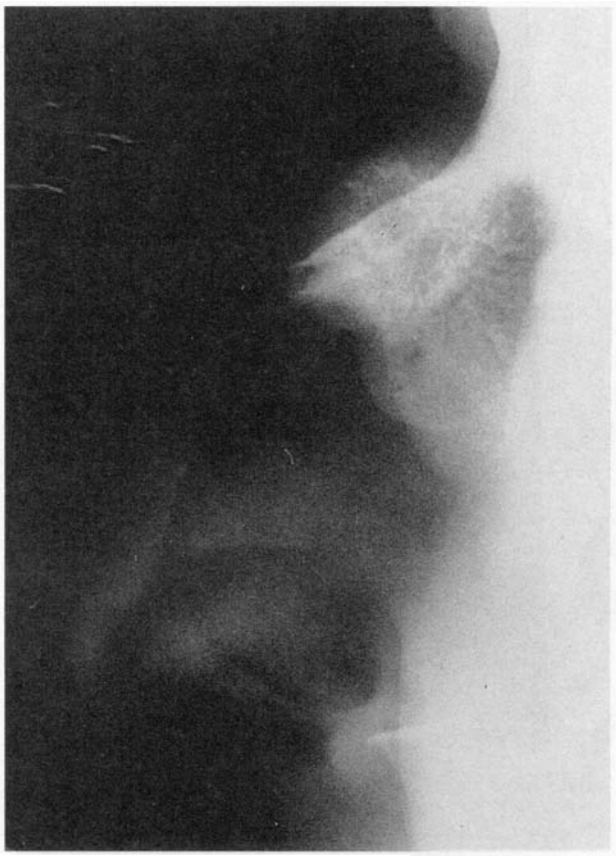

Fig. 3

Lateral X-ray of the nose demonstrating trabeculation within the haemangioma arising from the nasal bone.

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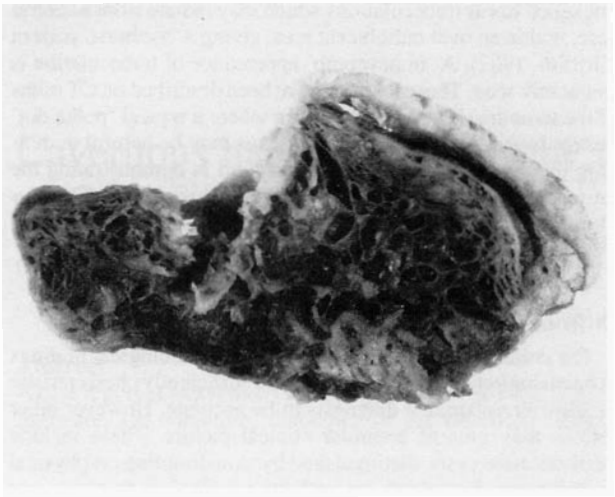

Fig. 4

Macroscopic 'honeycomb' appearance of the sectioned gross specimen.

Pathology and aetiology

Gross pathological examination showed a sponge-like red to brown spherical mass 1–2 cm in diameter, whose cross-sectional cut surface shows bony trabeculae between large blood-filled spaces giving a 'honeycomb' appearance (Fig. 4).

Histology shows thin walled, endothelial-lined, cavernous

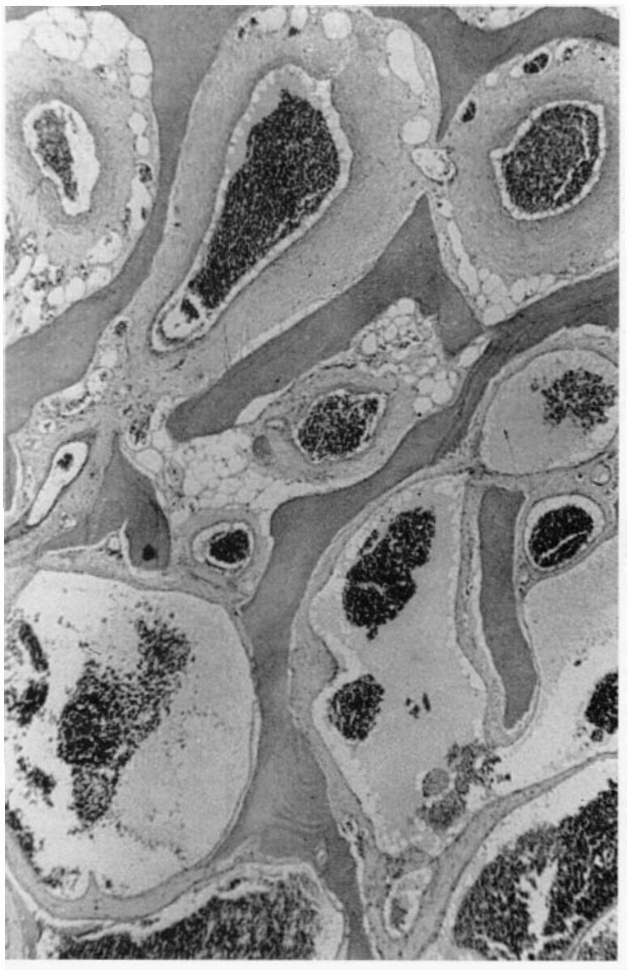

Fig. 5

Photomicrograph (×140) of the specimen demonstrating endothelial-lined blood-filled spaces within bony trabeculae.

TABLE I

SUMMARY OF REPORTED CASES (AFTER BRIDGER 1976)

Case no.	Author	No. cases	Age	Sex	Duration of symptoms	Situation of tumour	History of trauma	Management of defect	Follow up
18	Dorfman *et al.* (1971)	1	44	M	6½ years	Not specified	Not specified	No graft	Not specified
19			42						
20			43						
21	Zizmor *et al.* (1978)	4	46	2M	Not specified	Not specified	None	No graft in 3 cases cartilage graft in one case.	1–6 years
22			52	2F	'slowly enlarging mass'				
23	Schvarcz (1979)	1	47	M	Not specified	Right side	Nasal fracture prior presentation	No graft	8 months
24	Kanter *et al.* (1985)	1	36	M	1½ years	Left side	Vague: occupation Optometrist	No graft	1 year
25	Kaplan *et al.* (1991)	1	20	F	2 years	Left side	None	No graft	18 months
26	McAllister *et al.*, (1992)	1	56	M	4 years	Right side	Injury to right side of nose 6 years prior to presentation	Bone graft	3 months

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blood-filled spaces separated by trabeculae of lamellar bone of variable thickness. This produces a characteristic pattern of 'honeycomb', 'soap-bubble' or radiating 'sunburst' bony spicules, which are visible on gross examination and on radiographs and CT scans (Zizmor *et al.*, 1978).

The aetiology of cavernous haemangiomas is unknown. Local trauma at some time prior to presentation has been implicated as an associated factor but its influence remains unknown. Among the 26 reported cases a definite history of previous trauma is present in seven, suspected in a further three, not mentioned in two, and absent in 11.

Treatment and prognosis

Haemangiomas in most sites usually respond well to conservative surgical procedures (Dahlin, 1986). Radiotherapy may be used to treat haemangiomas at inaccessible sites, such as the vertebrae where surgery is usually confined to laminectomy for cord decompression. Haemorrhage may be significant in primary surgery for haemangiomas in long bones, but this has not been reported to be a problem in haemangiomas of the nasal bones, with the exception of one patient with a giant carvernous haemangioma at this site who required a blood transfusion (Schvarcz, 1979). A number of authors state that it was possible to excise the nasal bone tumour in one piece without undue difficulty and this was the experience in the patient we present. In eight cases the resulting defect was filled with a bone or cartilage graft, and in 11 cases no graft was used. In the remainder this part of the management is not mentioned. A satisfactory cosmetic result is reported in all cases, with some described as excellent. It is probable that organization of the haematoma in the defect and osteogenesis from retained periosteum contributes to this where a graft is not used. While the follow-up period in the patient we present is insufficient, there has been no recurrence of the tumour in any of the 20 patients in whom follow-up information is provided, and this varies between nine months and nine years.

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