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Clinical Record

Extranasopharyngeal angiofibroma of the cheek

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Abstract

Angiofibromas rarely localize in extranasopharyngeal sites. The most common site for extranasopharyngeal angiofibromas is the maxillary sinus. The ethmoid and sphenoid sinuses, nasal septum, middle and inferior turbinates, conjunctiva, molar and retromolar region, and larynx are other sites where extranasopharyngeal angiofibromas have been reported. Only one case of buccal extranasopharyngeal angiofibroma has been reported to date. We present a case of buccal extranasopharyngeal angiofibroma that was excised completely following embolization and we also review the literature.

Key words: Angiofibroma; Vascular Neoplasms; Cheek

Introduction

Angiofibromas are highly vascular and locally aggressive tumours despite their benign histopathology and they most commonly arise in the nasopharynx of adolescent males. They constitute 0.5 per cent of all head and neck neoplasms.[1] Angiofibromas usually arise from the posterolateral wall of the nasal cavity, where the sphenoidal process of the palatine bone meets the horizontal ala of the vomer and the pterygoid process.[2]

These tumours may rarely localize in extranasopharyngeal sites. To date, 56 extranasopharyngeal angiofibromas have been reported.[3] The most common site for extranasopharyngeal angiofibromas is the maxillary sinus. In a Medline search, we could find only one reported case of buccal extranasopharyngeal angiofibroma.[4]

In this article, we report a buccal extranasopharyngeal angiofibroma and review the literature.

Case report

A 17-year-old male presented to the out-patient clinic complaining of left buccal swelling of six months' duration. An incisional biopsy performed in another medical centre four weeks previously had been reported as 'capillary haemangioma'. Otolaryngological examination revealed a smooth-bordered, soft, painless, 5 × 3 cm mass in the left buccal region. The mass was covered with normal skin. There was a 1.5 cm incision scar related to the biopsy of the mass, in the left gingivobuccal sulcus. Results from anterior rhinoscopy, nasopharyngeal endoscopy and other otolaryngological examinations were within normal limits.

Doppler ultrasonographic examination showed significant blood flow and a low resistance flow pattern in the mass. Computed tomography (CT) showed a contrast-enhanced, smooth-bordered mass adjacent to the anterior border of the mandibular ramus, extending into the masticator space between the mandible and the maxilla, and extending into the left pterygopalatine fossa and inferior orbital fissure (Figure 1a). Magnetic resonance imaging (MRI) (Figure 1b) and digital subtraction angiography showed a highly vascular mass in the left masticator buccal space resembling a capillary haemangioma. Its blood supply was from the external carotid artery. The mass was embolized (Figure 2).

During surgery, the mass was seen to be adherent to the anterior wall of the maxilla and the masticator muscles posteriorly and to the zygomatic arc superiorly. Its size was 5 × 3 cm. The mass was excised after a superficial parotidectomy.

The localization of the mass and the branches of the facial nerve can be seen in Figure 3. The histopathological report was 'angiofibroma'.

Discussion

In a Medline search, we could find only one case of extranasopharyngeal buccal angiofibroma.[4] Our case will be the second in the English literature.

Some papers report buccal extension of extranasopharyngeal angiofibromas. Irby[5] reported an extranasopharyngeal angiofibroma originating in the infratemporal fossa and extending into the buccal region. Nasopharyngeal angiofibromas may also extend into the nasal cavity, maxillary sinus and from there into the cheek.[6] In our case, both nasopharynx and infratemporal fossa were free of tumour and the angiofibroma primarily involved the buccal region.

Extranasopharyngeal angiofibromas are rare tumours. They most commonly originate in the maxillary sinus.[7] The ethmoid and sphenoid sinuses, nasal septum, middle and inferior turbinates, conjunctiva, molar and retromolar region, and larynx are other sites where extranasopharyngeal angiofibromas have been reported.[3–8]

Angiofibromas are histologically benign tumours; however, they are locally aggressive. They may cause mortality due to haemorrhage and intracranial extension.[9]

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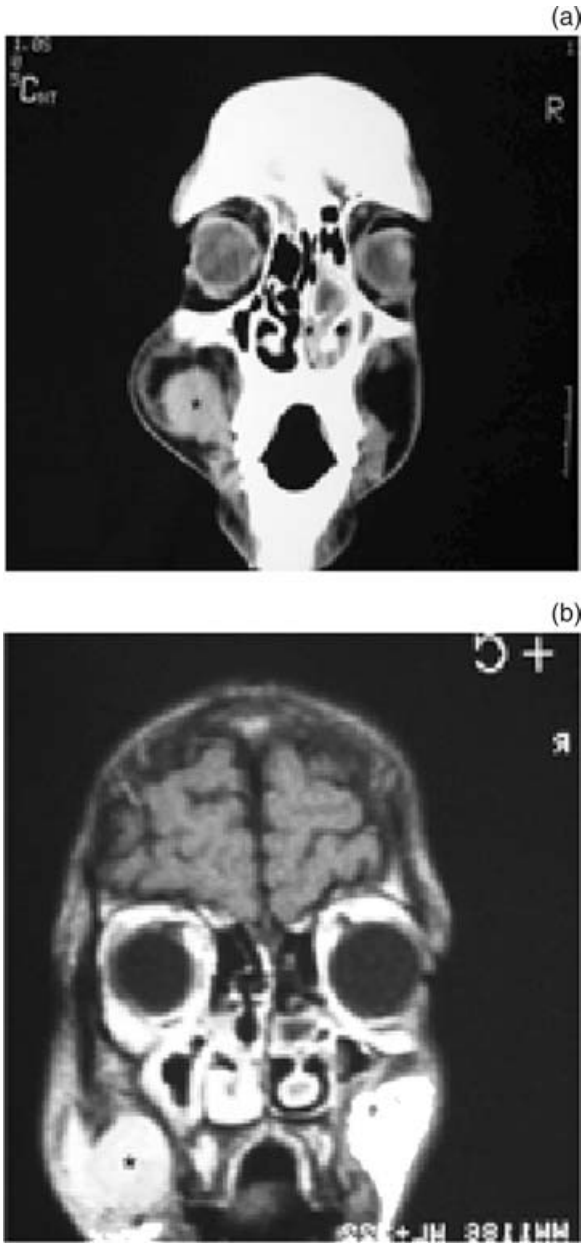

FIG. 1
(a) Computed tomography and (b) magnetic resonance imaging scans show the mass (*) in the left buccal region.

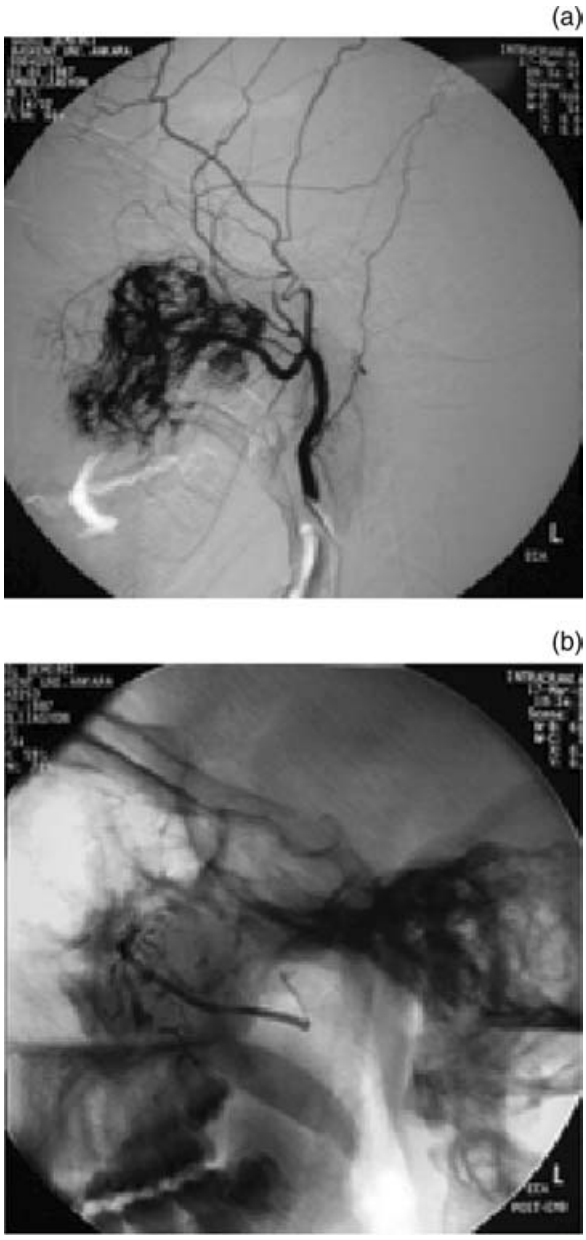

FIG. 2
(a) Digital subtraction angiography of the mass showing its arterial blood supply. (b) Post-embolization angiographic view.

The mean age of occurrence is 17 years for nasopharyngeal angiofibromas and 22 years for extranasopharyngeal ones. Nasopharyngeal angiofibromas occur almost always in men, although the female/male ratio for extranasopharyngeal angiofibromas is 1:3.[7] Our case was in a 17-year-old male.

The most frequent symptoms of nasopharyngeal angiofibromas are unilateral nasal obstruction, epistaxis and pain.[6,10] Extranasopharyngeal angiofibroma symptoms are related to their site of origin. Our patient did not have any symptoms except swelling in his cheek.

The most commonly employed radiological modalities are CT, MRI, MRI angiography and selective angiography of the external and internal carotid arteries. The size, extension, localization, vascularization of the mass and the involvement of the surrounding bony structures may be determined by these techniques.[2] We determined the localization and vascularization of our patient's mass using CT, MRI and digital subtraction angiography. Selective embolization of the mass enabled minimal blood loss during surgery.

After excision, the mass was measured at $5 \times 3 \times 2$ cm. In microscopic examination, many irregular blood vessels of different calibres, lined with one layer of endothelium, were seen in a fibrocollagenous stroma (Figure 4). The thick-bordered vessels contained an interrupted medial layer. Vascularity was more prominent in the periphery of the tumour and consisted of small vessels. The lesion was relatively more hypovascular in its central part, with larger vessels (Figure 5). The stroma between the vessels consisted of spindle-shaped and stellate fibroblastic cells that were scattered in an irregular, collagenous matrix. There were no mitoses. Masson trichrome staining revealed an incomplete muscular layer in the walls of large vessels (Figure 6a) and elastic van Gieson (EVG)

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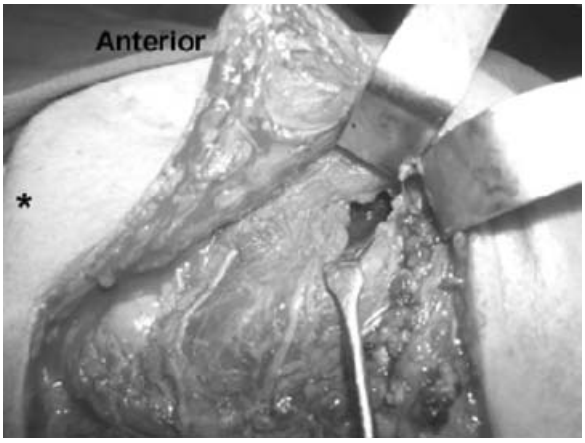

FIG. 3
The localization of the mass and the branches of the facial nerve. * = angle of the mandible.

staining showed that these areas did not contain any elastic fibres (Figure 6b). The histopathological diagnosis was 'angiofibroma'.

Angiofibromas may be mistaken for capillary haemangiomas histopathologically. The incisional biopsy was reported as 'capillary haemangioma' in our case. In fact, angiofibromas contain more erectile tissue and fibrous component compared with capillary haemangiomas, and these characteristics assist the differential diagnosis.

Tillaux[11] suggested that nasopharyngeal angiofibromas originated from the fibrocartilaginous barrier in the lower border of the sphenoid bone, in front of the atlas. Brunner[12] described this structure as 'fascia basalis' because he found no cartilage in it. Later, Hiraide and Matsubana reported a case of angiofibroma located in the anterior third of the nasal septum and showed that this tumour originated from the periosteum of the perpendicular lamina of the ethmoid bone, away from the fascia basalis.[13] Following this and other reports concerning other extranasopharyngeal angiofibromas, the most common theory explaining their site of origin is the presence of ectopic tissue.[10] Our case of buccal extranasopharyngeal angiofibroma also supports the theory of ectopic tissue as the site of origin since the tumour was located primarily in the buccal region away from the nasopharynx and the nasal cavity.

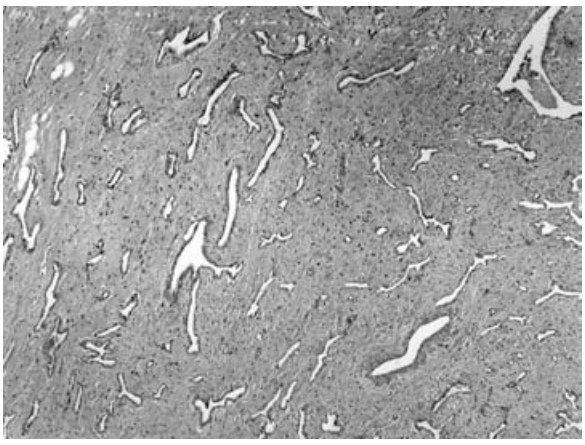

FIG. 4
Irregular vascular structures in a fibrocollagenous stroma (H & E; ×40).

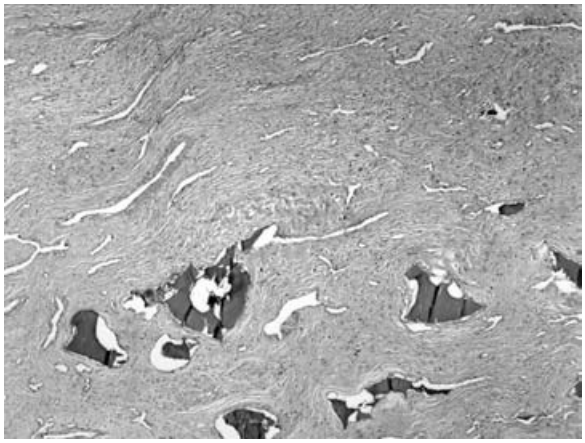

FIG. 5
Large vascular structures located in the central part of the lesion, which are filled with the material used for vascular embolization (H & E; ×40).

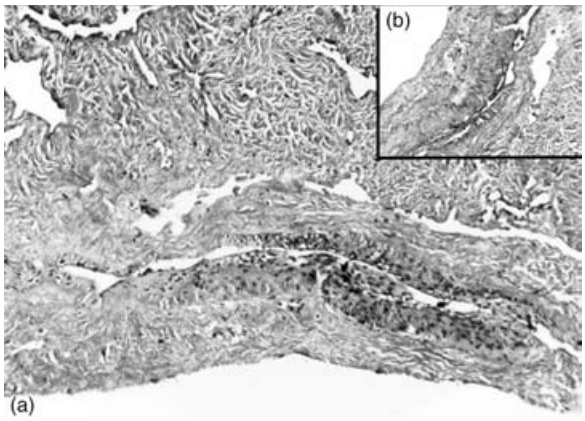

FIG. 6
(a) The incomplete muscular layer in the vascular wall (Masson trichrome staining; ×100). (b) Absence of elastic fibres in areas where no smooth muscle bundles exist in vessel wall (EVG staining; ×100).

In conclusion, it must be kept in mind that angiofibromas originate in the extranasopharyngeal regions of the head and neck. They must be included in the differential diagnosis of vascular extranasopharyngeal masses.

- **This case report describes an angiofibroma occurring in the buccal area of a 17-year-old male**
- **Angiofibromas occurring in sites outside the nasopharynx are very rare. The case was treated by surgical excision following arterial embolization**
- **The radiological, clinical and pathological features are discussed**

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