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CASE REPORT

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Intraosseous haemangioma of the mandible: a case report

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Abstract An unusual case of an endangering intraosseous haemangioma affecting the mandible in early childhood is presented. The early diagnosis was unclear until a surgical biopsy was performed. MRI confirmed the diagnosis. Successful emergent arterial embolisation and intralesional steroid injection were undertaken to control troublesome bleeding prior to medical treatment.

Keywords Conventional radiography · MR imaging · Embolisation · Mandible · Haemangioma

Introduction

Haemangiomas are endothelial cell vascular tumours. They are commonly located in cutaneous soft tissues and their natural history is of rapid postnatal growth (proliferative phase), followed by slow spontaneous regression (involuting phase), which can take years [1]. Although the majority require no specific treatment, 10–20% are the so-called 'endangering' type, requiring specific treatment. We report a rare presentation of an intraosseous haemangioma that caused troublesome oral bleeding and required catheter embolisation for control of bleeding.

Case report

A 10-month-old previously well girl presented with painless left facial swelling. On examination, there was hard swelling and expansion of the left mandible in the canine-to-molar region, which was non-tender. The overlying mucosa was normal and the child was afebrile. General examination was otherwise unremarkable and there was no significant past medical history.

Radiography of the mandible showed a left-sided, lytic expansile lesion displacing the teeth (Fig. 1). A ^{99m}Tc bone scan demonstrated increased focal uptake in the lesion. Unenhanced CT confirmed the lytic expansile nature of the bony lesion, together with some internal septations. The lesion encroached onto adjacent teeth but did not involve them; the bony cortex was intact and there was no abnormality of the perimandibular soft tissues.

Surgical biopsy of the lesion was performed under general anaesthesia. At operation a mucosal flap was reflected over the affected alveolar ridge. No perimandibular soft tissue abnormality was seen. On incision of the cortex there was brisk haemorrhage with substantial blood loss requiring resuscitation. Haemostasis was obtained using bone wax and Surgicel and the procedure was abandoned. Histology of the small amount of tissue obtained showed bony trabeculae with intervening small and large channels lined by plump endothelial cells. The endothelium was strongly positive for CD31 and histology was compatible with intraosseous haemangioma. There were no postoperative complications.

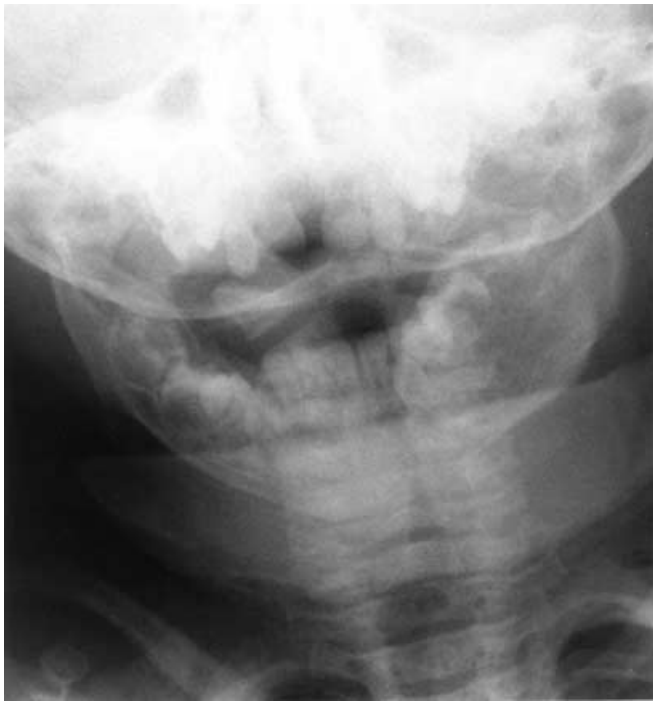

Fig. 1. Radiograph showing expansion of the left mandible with distortion of the dentition

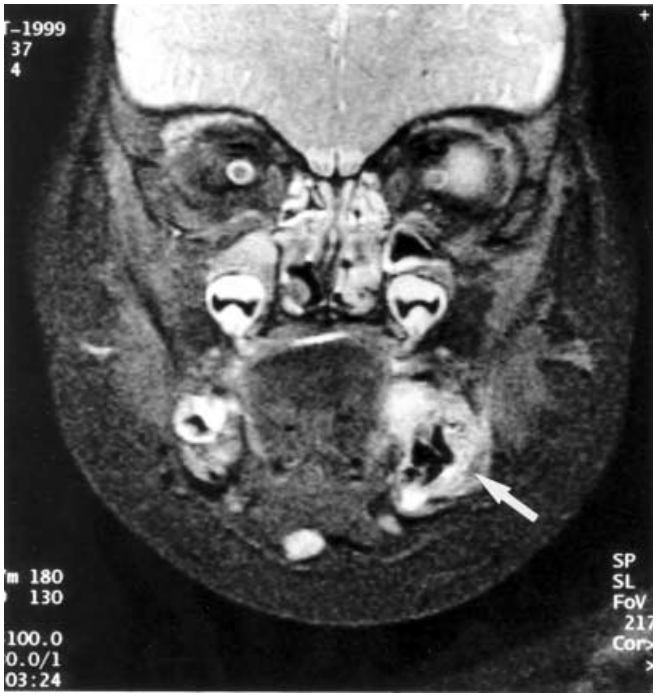

Fig. 2. Coronal, T1-W, post-gadolinium fat-saturated MRI showing an intensely enhancing parenchymal lesion in left mandible (*arrow*) with central flow voids

The child remained well for the next 6 months, but suffered from frequent oral bleeding from the site of the lesion. The opposing upper molar, which was encroaching onto the lesion, causing it to bleed, was subsequently extracted. MRI confirmed a proliferating haemangioma. The lesion was isointense on T1-weighted spin-echo and hyperintense on T2-weighted fast spin-echo images, with intense parenchymal contrast enhancement and flow voids present in the centre of the lesion (Fig. 2).

Oral bleeding from the gum overlying the haemangioma became increasingly troublesome, and the child was found to be anaemic (haemoglobin 9.2 g/dl). In order to control bleeding, catheter angiography and embolisation of the lesion with direct intralesional injection of steroid were performed under general anaesthesia. Left external carotid arteriography confirmed abnormal hypervascularity in the left mandible consistent with a proliferative haemangioma (Fig. 3a). The haemangioma was predominantly supplied from the lingual artery (sublingual branch), the facial artery (jugal branch) and the internal maxillary artery (inferior dental branch). The inferior dental arteriogram showed pooling of contrast medium in the mandible due to bleeding around a tooth socket (Fig. 3b). Arterial embolisation (with a coaxial system) was carried out with a 4-F guiding catheter in the common carotid artery. Polyvinyl alcohol (200-μm size) was used in the lingual and facial branches and absorbable gelatin sponge and a single micro-coil in the internal maxillary branch. Left external carotid arteriography immediately after embolisation showed significant reduction in vascularity (Fig. 3c). The haemangioma was then directly punctured with a 20-G needle from an intra-oral approach, a contrast study performed and intralesional steroids (triamcinolone 40 mg and betamethasone 4 mg) injected.

Treatment with oral prednisolone was commenced following the procedure and continued for 3 months (2 mg/kg per day for 6 weeks and then on a reducing dose). The lesion significantly decreased in size within days of embolisation and has continued to decrease in size with normal eruption of the first and second molars in the left mandible. Currently, 18 months since initial presentation, the child remains well with no significant troublesome rebleeding.

Discussion

Haemangiomas are the commonest tumours of infancy and childhood, occurring in approximately 12% of infants. Most are cutaneous in location although visceral sites such as the liver are well recognised [1, 2]. The incidence of intraosseous haemangiomas is unclear and, to date, primary mandibular haemangiomas have been rarely reported in infancy and early childhood [3, 4]. Mandibular haemangiomas in this age group more commonly arise from adjacent soft tissue and involve the mandible secondarily [3]. Our case reports the rare occurrence of a primary mandibular haemangioma.

The current classification of vascular anomalies into vascular tumours (predominantly haemangiomas) and vascular malformations by Mulliken and Glowacki [5] has allowed improved understanding of these anomalies. Haemangiomas are endothelial cell tumours while vascular malformations are channel disorders due to errors in vascular morphogenesis. This classification, which is supported by clinical, histological and biochemical differences, has good correlation with imaging findings (including MRI and catheter angiography), allowing differentiation of haemangiomas from vascular malfor-

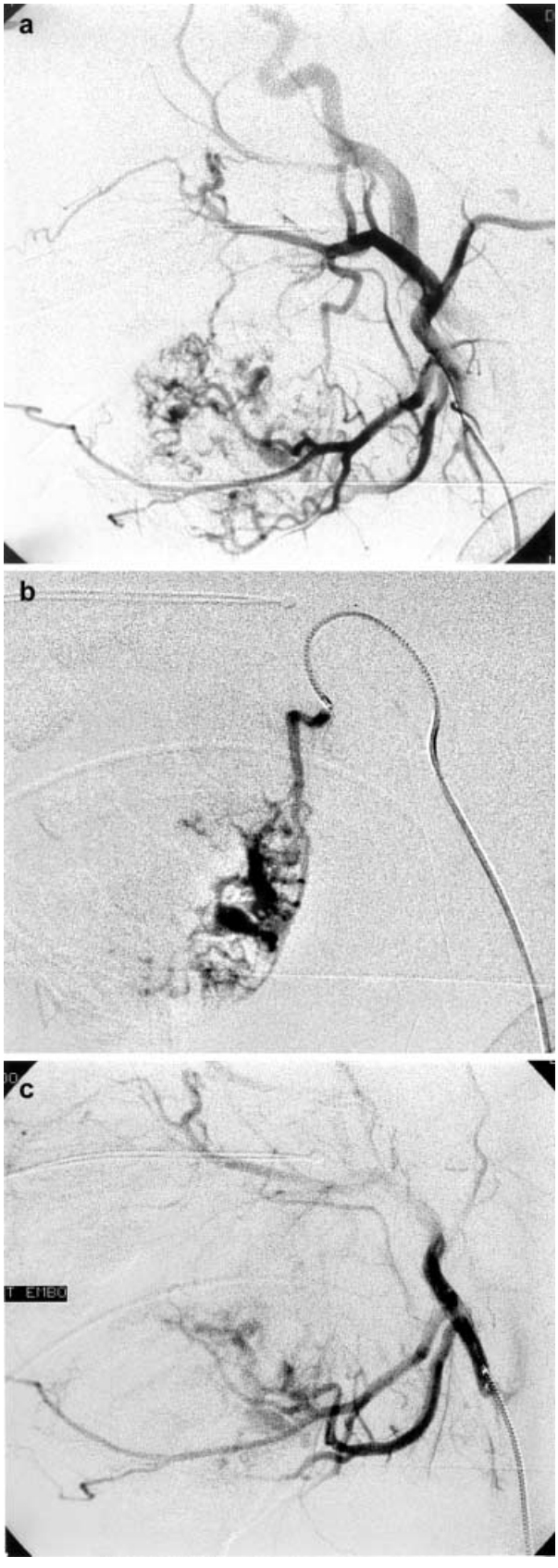

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Fig. 3a–c. **a** Left external carotid arteriogram (prior to embolisation) demonstrating the hypervascular haemangioma. The facial artery (jugal branch), lingual artery (sublingual branch) and internal maxillary artery (inferior dental branch) feeders are shown. **b** Selective left inferior dental arteriogram. Pooling of contrast medium is present around a tooth socket at the site of clinical bleeding. This vessel was successfully embolised with absorbable gelatine sponge and a single micro-coil. **c** Post-embolisation, left external carotid arteriogram showing significant reduction in vascularity of the haemangioma. Super-selective embolisation has ensured patency of the main trunks of the facial, lingual and internal maxillary arteries

mations and the subtypes of the latter [6]. Using this classification, the majority of mandibular haemangiomas, which are reported predominantly from adult patients, are most likely malformations [4]. Ten to twenty percent of childhood haemangiomas are the so-called 'endangering' form because, during proliferation, rapid growth may endanger adjacent structures and vascular shunts can lead to complications associated with these biologically active lesions. Our case illustrates a rare location for an endangering haemangioma at a primary intraosseous site.

Vascular anomalies may have associated alteration in bone contour, hypertrophy and demineralisation with bony destruction. Bone alteration possibly occurs as a result of physiological factors related to alteration of bone blood flow [7]. Demineralisation and bone destruction are more commonly observed in high-flow anomalies, which could explain the radiographic changes in our patient. Rapid growth and high vascular flow are features seen in these biologically active endothelial tumours during proliferation.

Lytic lesions in the mandible are rare at this age and can be seen in other conditions including osteomyelitis, eosinophilic granuloma, reparative granuloma and dentigerous cysts [3]. As plain radiographic findings in these lesions are non-specific, further imaging, such as MRI, should be undertaken. MRI can easily establish the diagnosis of a proliferating haemangioma and had this been undertaken early in our case, then surgical biopsy may have been avoided and appropriate treatment, such as with steroids, commenced. MRI typically shows a well-circumscribed, lobulated, hypervascular, uniformly enhancing mass lesion with dilated feeding and draining vessels, peripherally or centrally [1].

Arterial embolisation is rarely required in endangering haemangiomas, but can be useful to control complications, e.g. bleeding, and is undertaken when complications are unresponsive to medical management [8, 9]. In our case it was a useful therapeutic option providing emergent control of bleeding and stabilisation of the patient prior to commencing medical treatment.

When a young child presents with an expansile lytic mandibular lesion, the rare intraosseous presentation of

a haemangiomas should be considered. MRI can confirm the diagnosis and appropriate treatment commenced, thereby reducing the morbidity associated with an endangering haemangioma.

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