



Case Report

Central hemangioma—A case report and review of literature

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ARTICLE INFO

Article history:

Received 26 April 2010

Accepted 20 October 2010

Keywords:

Vascular lesions

Hemangioma

Vascular malformation

Angiography

Direct transosseous puncture embolisation

Transmandibular curettage

ABSTRACT

Intraosseous vascular lesions of the maxillofacial region are rare, can be challenging for diagnosis considering various other jaw lesions. They are subdivided into hemangiomas and vascular malformations. Hemangiomas are usually located in soft tissues. Intraosseous hemangioma is a quite rare condition, comprising less than 1% of all intraosseous tumors. This article describes a case of central hemangioma of the mandible, angiography was used for diagnosis and treated with surgical excision. The newer technique, transmandibular curettage via proximal osteotomy without complete resection was used. This technique is less invasive but effective and helpful in preserving the normal anatomy and function.

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1. Introduction

Vascular lesions of the maxillofacial region are classified as either: (i) hemangiomas or (ii) vascular malformations [1]. Hemangiomas are the most common cutaneous tumor of infancy and demonstrate rapid growth followed by a slow spontaneous involution or regression within five to seven years. Unlike hemangiomas, vascular malformations enlarge proportionately with the growth of the child and do not undergo spontaneous involution [1]. Vascular malformations can be subdivided based on blood flow rate: “slow flow” (capillary, venous, lymphatic or mixed) versus “fast flow” (arteriole, arteriovenous, fistulae or shunt) subtypes.

Intraosseous hemangioma is a rare condition, comprising less than 1% of all intraosseous tumors. It mainly occurs in the vertebral column. Mandible is a very infrequent location although possible. The female:male ratio is 2:1 and the peak of incidence is between the second and fifth decades of life [2,3]. Its origin is not defined. Some authors believe it is a true neoplasm, whereas others state it is a hamartoma resulting from proliferation of intraosseous mesodermal cells that undergo endothelial differentiation [2].

An eighteen years old female patient reported to Department of Oral and Maxillofacial Surgery with growth in the lower jaw. Main concern of the patient was the slow growing swelling which was causing change in her appearance. We report a case of slow growing, expansile lesion in right side of the body of mandible which was diagnosed as central hemangioma of the mandible.

2. Case history

An 18-year-old female patient reported with a complaint of swelling in the lower right side of the jaw since one and half years (Fig. 1a). Swelling was slowly increasing in size, which was of cosmetic concern to the young female. Clinical examination revealed approximately 2 cm × 1.5 cm, diffused swelling on the right lower side of the face. Swelling was hard on palpation and no pulsations or bruits were noted. Skin over the swelling was normal and no neurological deficit was noted. Intra oral examination showed vestibular swelling at number 44, 45 and 46 teeth region, which was hard on palpation. Buccal cortical plate expansion was noted with no change in the lingual side. Slight mobility of number 45 tooth was noted, otherwise all other teeth were clinically normal.

Diagnostic panoramic radiograph revealed ill defined radiolucent area at the periapical region of premolar teeth (Fig. 1b). There was frank blood noted on aspiration through the swelling. Because of close proximity of mental neurovascular bundle second aspiration was done to rule out accidental vascular puncture. Repeat aspirations also yielded frank blood. Provisional diagnosis of vascular malformation was done and planned for angiography to identify the feeder vessels as well as to study the topography of the lesion.

3. Diagnostic angiography

A transfemoral angiography of the external carotid artery was performed. Scan revealed intraosseous lytic lesion in the right side of the body of mandible measuring approximately 17.0 mm × 15.4 mm, with expansion and erosion of buccal cortical plate (Fig. 2a). There was evidence of bony perforation both buccal

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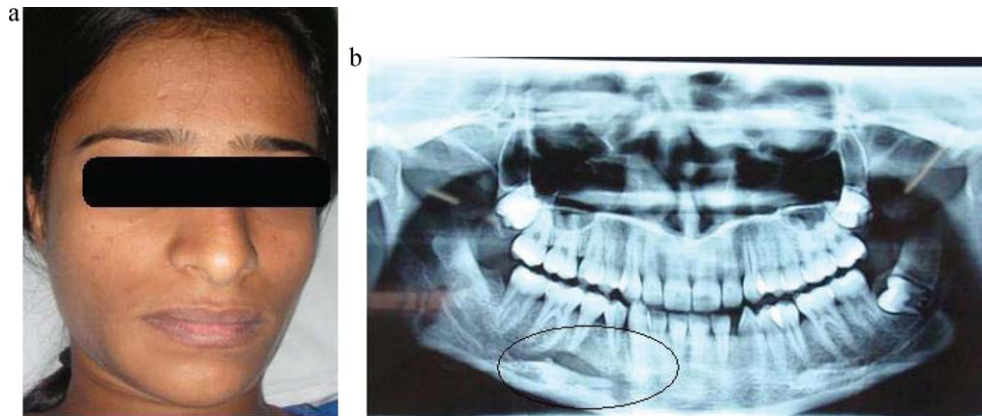


Fig. 1. (a) Patient with diffuse swelling below right oral commissure with normal overlying skin. (b) Pantamography indicative of diffused radiolucent lesion at the right mandibular premolar region.

and lingual side suggesting feeder vessels. Three dimensional reconstructed (3D) images of angiography showed unusually small facial artery with the branches entering the mandible (Fig. 2b). These branches were identified as supplying the lesion. The scan although did not show any evidence of contrast medium enhancement in the lesion, was conclusive of the buccal and lingual vessels entering the bone and the inferior alveolar vessel was yet another feeder to the lesion.

4. Treatment

Considering the location of the lesion in the anterior part of the mandible surgical excision of the lesion was planned. Patient was taken under hypotensive anesthesia. It was planned to approach the lesion perorally. Crevicular incision was made on the right side of the mandible. Mucoperiosteal flap was raised on both buccal and lingual side, ligating and cauterising the feeder vessels.

A bony window was made on the buccal cortex at the periapical area of lower right third molar region to reach the mandibular canal. The vessel in the canal was identified and electrocoagulated to render the lesion bloodless. Thus the vascular supply to the lesion was controlled from all the sources. Expansion of the buccal cortex was noted over the lesion with perforations. Mental neurovascular bundle seen emerging from the lesion was clamped and ligated (Fig. 3a). Deroofing of the lesion was done from the buccal aspect. Bony cavity with the epithelial lining was excised and surgical wound closed after achieving haemostasis (Fig. 3b).

Second premolar was extracted as it was left with no bone support. The lesion was approached systematically controlling all the blood supply, which minimised blood loss during surgery avoiding blood transfusion.

5. Histopathology

Diagnosis of central capillary hemangioma was confirmed by histopathological study with evidence of proliferating endothelial cells forming small to large blood vessels (Fig. 4a and b).

6. Discussion

Central haemangioma of the jaws is an uncommon lesion that is often difficult to diagnose. Pathogenesis of central haemangioma is still debatable and several theories are postulated. Some authors describe hemangiomas as congenital lesions whereas others believe that the inferior dental canal is the origin of the lesion, based on its widening in the majority of these patients [4,5]. There was no evidence of any lesion since birth in our case and patient noticed slowly enlarging swelling at the age of 16 years. The literature suggests that the lesion may present at any age, but was most commonly discovered in the second decade of life [6]. The most frequent location of hemangioma is the molar premolar region [5].

The literature contains vague and conflicting reports on the clinical and radiological features of central haemangioma. The initial diagnosis is usually complicated because of the absence

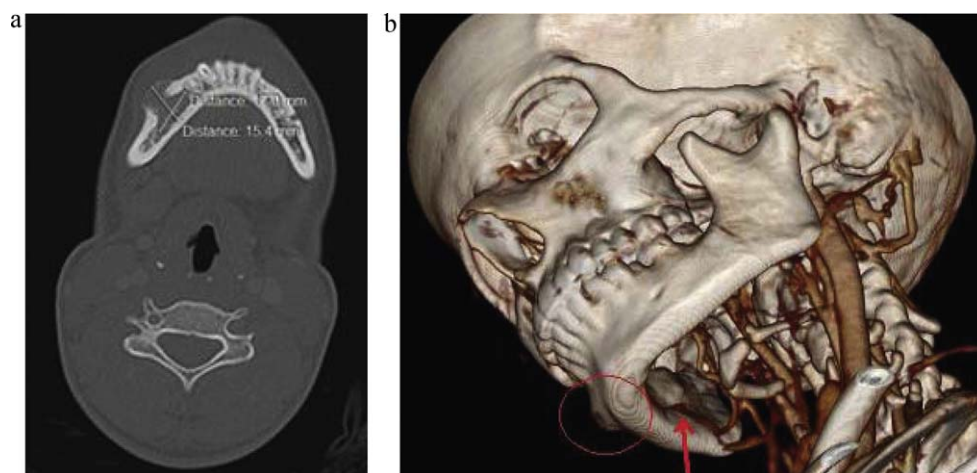


Fig. 2. (a) Computed tomography scan showing erosion of buccal cortex with respect to premolar molar region. (b) Angiograph showing feeder vessel on lingual aspect.

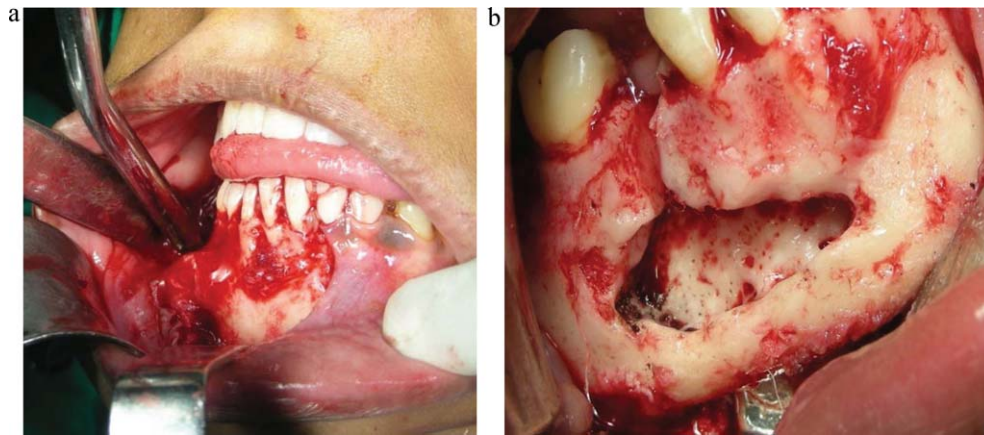


Fig. 3. (a) Exposure of lesion showing erosion of buccal cortex and in association with mental neurovascular bundle. (b) Clear bone cavity after excision of the lesion.

of symptoms and the unspecific radiological findings [4,7]. In our case clinical features except the presence of swelling were normal, number 45 tooth was mobile and had altered response on pulp vitality testing. There was no history of bleeding or altered neurological function. Although these lesions may present sign and symptoms including a slow growing bluish mass, discomfort, pulsatile sensation and mobile teeth [2,3]. In our case on panoramic radiograph there was ill defined radiolucency in the premolar region. This lesion should be considered in the differential diagnosis of any multilocular or unilocular radiolucent lesion of the jaws, especially when the mandible was involved and sometimes associated with a peripheral sclerotic border or a resorption of the neighbouring root teeth [5]. It is important to emphasise that some radiographic patterns, such as the spoke-like and sunray appearance, frequently described in other studies and textbooks, are actually extremely rare [6].

In the present case, angiography was helpful in identifying the feeder vessels. It also showed unusually small facial artery with branches entering the mandible. There was no evidence of contrast medium enhancement in the lesion. Some suggest preoperative arteriography is usually unnecessary because vascular flow cannot be identified in the majority of the cases [2–4,8,9] and angiographic aspects of some intraosseous lesions are not well-defined [10]. Histological variants of lesions such as low flow microfistulas and sclerotic vascular tissue may not be detectable through angiography [2]. Nevertheless, it should be performed together with a presurgical embolization in big lesions to minimize the surgical bleeding [11,12]. Therapeutic alternatives include: surgery, curettage and embolization [4,12]. Percutaneous embolization has been

defended by several authors, although technical risks are greater than benefits obtained [13].

Histologically, hemangioma can be divided into three groups: cavernous, is the most frequent one and is located in the mandible [4], capillary and mixed. The histological features are dependent on the stage of the lesion. In the proliferative phase, the lesion is highly cellular and contains plump proliferating endothelial cells and pericytes, with a high mitotic activity and numerous mast cells. Vascular channels are not prominent. In the involutive phase, the endothelial cells are flattened, the cell turnover is normal and there are few mast cells. Vascular channels filled with blood cells predominate, and the lesion is eventually replaced by fibrofatty tissue [14].

In contrast, histologic evaluation of vascular malformations shows no evidence of cellular proliferation, but rather a progressive dilatation of vessels of abnormal mural structure. Vascular malformations are lined by flat, quiescent endothelium, lying on a thin single laminar basement membrane. Mast cell counts are normal throughout the evolution of vascular malformations [1,15].

Resection of the mandibular fragment containing the lesion has long been considered essential to complete healing [16–19]. Curettage of the resected fragment with immediate replantation reduces the morbidity associated with the procedure and the difficulty of reconstruction [19,20]. Complete cure by arterial embolization is difficult, either with particles or glue, and may lead to complications [21–23]. Selective arterial embolization, followed by surgical treatment, is still the most modern conventional approach [17], but still results in functional deficits [24]. The direct transosseous puncture of the vascular bed has been proposed [16,25]. This procedure

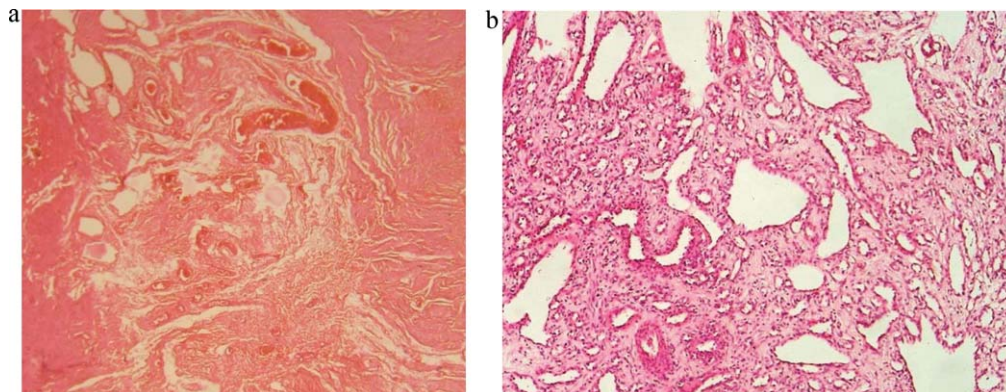


Fig. 4. (a) Evidence of numerous proliferating small to large blood vessels, with areas of numerous bony trabeculae. (b) At higher magnification blood vessels lined by endothelial cells in few vessels endothelial cells appearing as hobnail pattern.

controls the acute hemorrhagic phase, but does not eliminate the risk of a recurrence. Moreover recently it was reported that, blood loss was not significantly decreased by embolization [26–28].

The newer technique, transmandibular curettage via proximal osteotomy without complete resection, however, is a less invasive but effective method to treat small intraosseous vascular lesions of the mandible that have not invaded the soft tissues. Less blood loss, preservation of the bone and periosteum, more favorable postoperative esthetics and better function are among the advantages of this technique when indicated [27,29].

Azzolini et al. [30] presented an exclusively intraoral treatment by extracting the teeth and then cleaning the underlying cavity through the alveolar process and packing the mandible with oxidized cellulose after superselective intra-arterial embolization (SIAE). A modified technique was reported on the treatment of arteriovenous malformations in the mandibular body [31]. The technique reported consists of SIAE and piecemeal removal of the lesion through burred holes made in the cortex. Bone wax packing (BWP) of bone cavities was used to control hemorrhage of the lesions in emergency and carried out curettage 2 months later. This technique is reported as simple and safe. Bone tissue, dental embryo, erupted tooth, and lower alveolar nerve may be preserved in most cases. It reduces morbidity by maintaining the continuity of the mandible [32].

The management of these lesions depends on symptom control and aesthetic concerns. In the present case, a decision was made to remove the lesion due to its increasing size and cosmetic deformity. Though various therapeutic options have been described for soft tissue venous malformations, surgical excision remains the mainstay for purely intra-bony lesions, if necessary, augmented with preoperative embolisation [3,5,8,12,33–38].

In the current case, all the feeder vessels were ligated including the feeder from inferior alveolar canal by bur hole deroofing method. The lesion was exposed through the buccal cortex and was excised through this opening completely. Number 45 tooth was extracted. So, we employed the newer technique, transmandibular curettage via proximal osteotomy without complete resection.

In conclusion, intraosseous vascular lesions of the maxillofacial region are rare and can be challenging for diagnosis considering various other jaw lesions. We consider transmandibular excision and ligation of feeder vessels as less invasive and effective method to treat central hemangiomas of the mandible. There were preservation of continuity of jaw bone, more favorable postoperative esthetics and better function.

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