Mandibular Arteriovenous Malformation with Unusual Radiographic Appearance

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KEY WORDS
Arterio-venous malformation; Mandible; Treatment;

ABSTRACT
Arteriovenous malformation (AVM) of head and neck is a rare vascular anomaly but when present, it is persistent and progressive in nature. It can also represent a lethal benign disease due to massive hemorrhage. There are several indications for treatment, including age, location, extension and type of vascular malformation. Endovascular therapy can effectively cure most lesions with limited tissue involvement. Surgery can be used in selected cases in combination with embolization. Here, we present a rare case of arteriovenous malformation of mandible with floating tooth appearance in an 11-year-old boy patient. Given the spectrum of imaging presentations seen and the crossover with other lesions, microscopic histopathological examination is the gold standard for diagnosis.

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Introduction
All vascular lesions until 1980’s were referred to as hemangiomas. Classification of vascular lesions based on endothelial characteristics into hemangiomas and vascular malformation was done by Mullikin et al. [1] in 1982. In 1996 International Association of the Study of Vascular Abnormalities modified the classification as vascular tumors or vascular malformations [2]. Vascular malformations based on blood flow are classified into low flow lesions such as capillary, lymphatic malformations and venous malformations also high-flow lesions such as arteriovenous malformations (AVMs) and arteriovenous fistulas [1]. Differentiation criteria between AVM and hemangioma are based on the clinical and histological characteristics. Most hemangiomas are not usually present at birth; they are characterized by a rapid growth phase with endothelial cell proliferation. Shortly after birth, they grow rapidly often faster than the child’s growth, which then show a gradual involution. On the other hand, AVMs are present at birth and enlarge proportionately with the growth of the child and remain throughout the life. They characterized by a normal rate of endothelial cell turnover and are the result of direct and permanent communication between arteries and veins [1,3]. AVMs are benign lesions that can arise in the any area of the body. Rarely, AVMs can arise within hard tissue structures such as bone/jaws and give rise to a highly unusual spectrum of symptoms [4-5]. Jaw intraosseous arteriovenous malformations have a high risk of life threatening hemorrhage either spontaneously or after oral surgeries [6-7]. In this paper, we report the case of mandibular intraosseous arteriovenous malformations in an 11-year-old boy with floating tooth appearance in radiographic feature.

Case Presentation
An 11-year boy with complaint of a painless swelling of the right posterior mandible and tooth mobility in that region for one month was referred to Department of oral and maxillofacial surgery, Shahid Beheshti University of Medical sciences (Iran, Tehran. The patient did not report a history of trauma or medical problems. Clinical examination was normal. In the intraoral examination, a fluctuant swelling extending from first premolar to first molar with erythematous mucosa was seen. Cone beam computed tomography (CBCT) revealed an ill-defined
unilocular radiolucency that extended from first molar to the distal of first premolar with floating tooth appearance (Figure 1). Bone destruction was seen in lingual and buccal crest which caused perforation in some areas. It seemed that mandibular canal was intact (Figure 2). On aspiration blood was seen. Due to clinical, radiographic and aspiration findings, central giant cell granuloma, leukemia and osteosarcoma were suggested as a differential diagnosis. Under local anesthesia, excisional biopsy of the lesion was done; it completely excised with two teeth and was submitted for the histologic examination. The gross was an irregular, creamy-brown soft tissue with elastic consistency, measuring 2.5 X 2X1 centimeters.

Histopathology sections showed a vascular lesion composed of a varying mixture of arteries, veins and associated small vessels within a fibrous background. Mixed inflammatory cells infiltration, giant cells, hemorrhage and curetted bone are seen (Figure 3a and b). The lesion is covered by keratinized epithelium which in some areas ulcerated and replaced by fibrinopurulent membrane (Figure 4). According to histopathologic and radiographic findings, the diagnosis of arteriovenous malformation was done. There was no recurrence during a 6-month follow-up (Figure 5).

Discussion
Vascular malformations are abnormal communications which lined by inactive endothelium. Although these conditions are congenital in nature, they show an additional growth when disturbed by trauma, infections or endocrine fluctuations [8]. Deepa et al. [9] reported that the mean age of presentation of intraosseous arteriovenous malformations in jaws (j-AVMs) is 19 years. In this case, child was asymptomatic until 11 years after that tooth mobility was observed. J-AVMs are very rare conditions but have an increased tendency of life-threatening due to hemorrhage either spontaneously or after surgery, therefore one should appreciate their part of a differential diagnosis when considering options for unexplained symptoms and pathology [4,10]. The female to male ratio is 2:1, while in our case is male, with peak incidence between the ages of 10 and 20 with extremes at 3 months and 74 years of age. Area of head...
and neck consists of 50% of vascular lesions with only a small percentage occurring in the jaws. The incidence is twice more common in the mandible than maxilla, as we saw in this case [11]. AVMs have slow growth nature, for this reason, they are asymptomatic for long time and can be seen at any age [12]. They usually present with non-specific symptoms bruit, dental loosening, swelling of soft tissues, change in skin and mucosal color and paresthesia of the lower lip or chin [13,14]. Although this case has no history of dental visits, but the appearance of jaw swelling and tooth mobility has been observed suddenly during one month. Due to presence of teeth, complex morphology of the jaws and vital structures in maxillofacial area, diagnosis and management of jaw AVMs are more difficult than other intrabony AVMs [6]. If occurring in proximity to teeth, AVMs can cause mobility and displacement, in keeping with their slow-growing nature, like in the present case. However, there have been reports of root resorption occurring [15]. In our case, bone destruction is seen in buccal and lingual crest, noticeably osseous erosion of the alveolus with apparently floating teeth, but root resorption is not seen. Clinically, J-AVMs are observed after extractions, biopsy procedures or while brushing [8]. The radiographic appearances of these lesions are variable and can be similar to other lesions, so definitive diagnosis based only on imaging is difficult [16]. Differential diagnosis of J-AVMs includes hemangioma, Langerhans’s cell histiocytosis, Ewing’s sarcoma, simple bone cyst, aneurysmal bone cyst, ameloblastoma, osteosarcoma, ameloblastic fibroma, odontogenic myxoma, central giant cell granuloma, odontogenic keratocyst, dentigerous, fibrous dysplasia and metastatic malignant tumors [8,17-18]. Despite of unilocular radiolucent lesion in our case, most commonly, these lesions show a multilocular radiolucent defect. The individual lesions may be small (honeycomb appearance) or large (soap bubble appearance). In other case the lesion may
present as an ill-defined radiolucent area or a well-defined, cystic-radiolucency. Large malformations may cause cortical expansion, and occasionally a sunburst radiographic appearance is produced. Angiography can be helpful in demonstrating the vascular nature of the lesion [19].

In few cases, markedly obvious osseous erosion of the alveolus with apparently floating teeth were also evident [20], such as present case. These features can be seen in other lesions like ameloblastomas, osteosarcomas, myxomas and or fibrous dysplasia [21]. Biopsy and histopathological examination for precise diagnosis of AVMs is needed. Features are fairly consistent in AVM histology, proliferative epithelial cells and fibrous tissue are often seen surrounding the lesion and the remaining bony trabeculae are interspersed with vascular structures and endothelial cells [4, 22], as the present case which the remaining bony trabeculae are interspersed with vascular structures (Figure 3b).

AVMs demonstrate a therapeutic challenge because of their hemodynamic characteristics and their modality of growth. Elimination of lesion, hemorrhage control during surgery and preventing of recurrence are main goals for treatment of AVMs. Radiotherapy may be only treatment option for inaccessible AVMs, however any bony deformity or effects on adjacent structures can only be controlled and corrected through surgery [23]. Treatment also depends on size of lesion and degree of involvement of vital structures. Direct surgery, with or without embolization of major afferent (feeding) vessels is the accepted treatment of choice. For lesions that require resection, radiographic embolization often is performed one to two days earlier than surgery process to minimize blood loss [19]. The present case, depending on the size of lesion, was also treated with direct surgery without embolization.

Radical excision of the large lesion allows for a curative purpose and generally immediate reconstruction is undertaken with a tissue graft [24].

The patient has signed informed consent for the surgical procedure and necessary information for reporting this case.

Conclusion
Here, we presented the rare case of mandibular intraosseous arteriovenous malformation with floating tooth appearance in an 11-year-old male patient. Given the spectrum of imaging presentations seen and the crossover with other lesions, microscopic histopathological examination for precise diagnosis is gold standard.

Conflict of Interest
Not declare

Reference


