Arteriovenous Malformation of Face

Abstract

Arteriovenous malformations (AVMs) are rare congenital vascular malformations accounting only 1.5% of all vascular anomalies with 50% occurrence in the oral and maxillofacial region. It usually results from birth defects of the vasculature. A literature search revealed only few case reports of AVMs in the facial region. Lack of meticulous diagnosis, scarcity of knowledge, and paucity of literature can result in their exsanguinations leading to fatal hemorrhagic incidents after various dental procedures such as tooth extraction, surgical intervention, puncture wound, or blunt injury in involved area. The present case describes the accidental diagnosis of asymptomatic high-flow AVMs in the facial region of pediatric patient reported primarily for the treatment of periapical abscess. This case report is unique because although there was no history of bleeding episodes, thorough examination and investigations diagnosed it as high-flow vascular malformation. It is important for the dental practitioner to be aware of AVM which may be present in the head and neck region that can produce fatal bleeding episodes during various dental procedures. Proper diagnosis of AVMs through complete history, precise clinical examination, and advanced imaging modalities can help in preventing serious life-threatening complications.

Keywords: *Arteriovenous malformations, maxillofacial region, vascular anomalies*

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Introduction

Arteriovenous malformations (AVMs) are rare congenital vascular lesions occurring anywhere in the body. These can be life threatening due to potential massive hemorrhage. Based on endothelial characteristics, Mulliken and Glowacki[1] (1982) classified vascular lesions into (1) hemangioma – vascular tumor and (2) vascular malformations. Both categories of vascular lesions have different etiologies and clinical features. Hemangiomas are vascular tumors demonstrating endothelial hyperplasia that enlarges by rapid cellular proliferation. These are normally absent at birth but proliferate during the 1st year of life and then involute. Vascular malformations are congenital structural malformations with normal rate of endothelial cell turnover, which are present at birth but usually become noticeable in the later age. Rapid enlargement of the malformations is usually triggered by trauma or hormonal changes during puberty or pregnancy. The enlargement of these lesions is due to the change in pressure and flow, dilatation of vascular channels, shunting, and collateral proliferation rather than cellular proliferation.[2] Furthermore, based on blood flow characteristics, vascular malformations can be divided into low-flow lesions and high-flow lesions. Low-flow lesions include capillary, lymphatic, and venous malformations whereas high flow includes arterial and AVMs.[3]

Hemangiomas are formed due to a failure of differentiation in the early stages of embryogenesis however, vascular malformations are caused due to disturbance in the late stages of angiogenesis resulting in the persistence of embryonic arteriovenous anastomosis.[4] The shunt from high-pressure to low-pressure compartment can produce a pulsating mass and a characteristics bruit. The vein emerging from the shunt becomes dilated and thickened, and increased flow rate accounts for observed dilatation of the arteries.[5]

AVMs are rarely seen, only accounting for 1.5% of all vascular anomalies, and 50% of the lesions are located in the oral and maxillofacial region.[6] AVMs can easily be misdiagnosed and produce the substantial oral bleeding. Because of severe life-threatening complications associated with vascular malformations, dentist should be aware of fatal outcome so that necessary investigations should always be carried out before performing any oro-dental procedure.

How to cite this article: Kumar A, Mittal M, Srivastava D, Jaetli V, Chaudhary S. Arteriovenous malformation of face. Contemp Clin Dent 2017;8:482-4.

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Access this article online

Website: www.contempclindent.org

DOI: 10.4103/ccd.ccd_100_17

Quick Response Code:

Case Report

A 10-year-old girl reported to the Department of Pediatric and Preventive Dentistry, ESIC Dental College and Hospital, Delhi, India, with a chief complaint of pain and swelling in the upper right back tooth region for the last 4–5 days. The pain was constant, throbbing and spontaneous. No significant medical history was reported by patient and parents. On clinical examination, there was a swelling on the right side of midface in the maxillary region which was slightly tender. Intraoral examination revealed grossly decayed primary maxillary right second molar and swelling in the gingiva adjacent to the tooth. Slight pus discharge from gingival swelling was also seen. Hence, provisional diagnosis of periapical abscess with respect to primary maxillary right second molar was made, and antibiotics and analgesics were prescribed. The patient was recalled after 3 days for review. On review, gingival swelling was reduced in size whereas facial swelling was persistent but nontender. Primary maxillary right second molar was extracted and the patient was recalled after 1 week. On follow-up, complete healing at extraction site was observed with persistent facial swelling. On further history, patient's mother revealed that the swelling was there since many years but she did not remember the exact duration of swelling. There was also a history of fall from height at the age of 1 year. On further examination, slight bluish discoloration over the face was observed [Figures 1 and 2]. Provisional diagnosis of hemangioma or vascular lesion was made, and orthopantogram was advised which did not reveal any bony involvement [Figure 3]. Then, magnetic resonance imaging (MRI) angiography was advised. MRI angiography revealed ill-defined serpiginous altered signal intensity lesion in the right cheek in deep subcutaneous plan having multiple flow voids onT1-weighted and T2-weighted images. The lesion showed early arterial enhancement with multiple enhancing vessels directly draining into veins. Arterial supply was from the external carotid artery, and the lesion was draining into the right internal jugular vein [Figure 4]. MRI angiography findings were suggestive of AVM in the right cheek. Therefore, diagnosis of AVM (which was initially superimposed with periapical abscess) was made, and the patient was referred to Department of Pediatric Surgery for further management. Embolization of lesion was done, and the patient was scheduled for follow-up examination.

Discussion

AVMs are fast-flow vascular malformations, consisting of anomalous capillary beds between the arterial and venous system, thus causing shunting of blood. AVMs are the most aggressive form of vascular malformation which can lead to significant deformity and functional impairment.[2] Fast-flow vascular malformations usually become evident during childhood and puberty.[7] Kohout *et al*. in their study found that AVMs were present at birth in 59% of cases, in childhood 10% of cases, in adolescent 10% of cases, and in adulthood 21% of cases.[8] In the present case report, AVMs was present in childhood.

AVMs grow synchronously with the growth of the child. Puberty and trauma are found to have triggered the rapid growth of the lesion leading to the manifestation of symptoms.[9] Holt *et al*. reported that trauma leading to the AVMs may be penetrating, blunt, postsurgical, or inflammatory.[10] The enlargement occurs due to the change in the pressure and flow within the malformation, ectasia, shunting, and collateral proliferation rather than cellular proliferation.[11] In the present case report, symptoms were triggered after periapical infection in the tooth which probably leads to inflammation in the region. Although not definite, the onset of symptoms may be attributed to the infection and inflammation in the periapical region of the tooth.

Various diagnostic tools are available to diagnose vascular lesions such as ultrasound with color Doppler, computed tomography (CT) scan, MRI, and magnetic resonance angiography. Type of malformation can be confirmed by ultrasound with color Doppler examinations. CT scan with iodinated contrast identifies AVMs as a highly enhancing

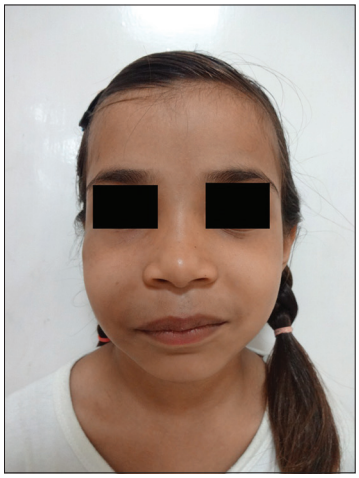

Figure 1: Frontal view

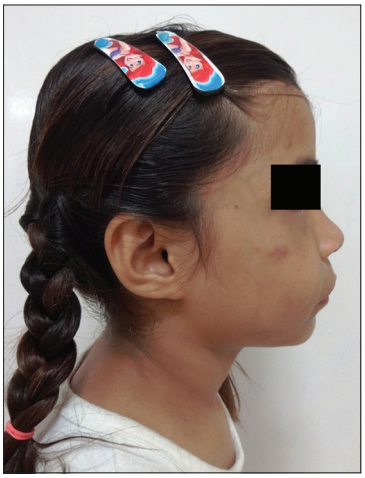

Figure 2: Lateral view

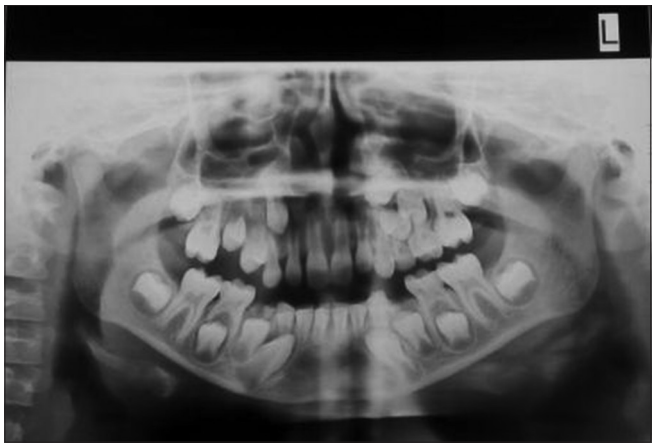

Figure 3: Orthopantogram

lesion and can demonstrate soft tissue enhancement as well as dilated feeding and draining vessels.[12] MRI can be used as an excellent technique in the diagnosis of vascular malformations either by itself or before angiography. It shows a good depiction of vascular structure, permitting the differentiation between high-flow and low-flow lesions. High-flow lesions show typical signal flow voids both in T1-weighted and T2-weighted sequences with the appearance of serpentine images.[13] In the present case, MRI was able to identify the kind of flow, i.e., high flow as well as the origin of the vessels, i.e., external carotid artery.

Management of AVMs is most difficult due to the replacement of normal tissue by the diseased vessels and high-flow rate of recurrence.[9] This management mainly consists of surgery, vascular embolization, or a combination of both. Surgical treatment consists of wide resection which is difficult and potentially hazardous due to significant blood loss during surgery.[14] The purpose of embolization is to occlude the vessels contributing to the lesion. Various materials such as polyvinyl alcohol particles, muscles, gel foam, cyanoacrylate, metal coils, and collagen[4] can be used for embolization. The combination of embolization and surgery is used to control acute hemorrhage allowing for excisional surgery to be performed afterward. In the present case, the patient was embolized and was scheduled for follow-up examination.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

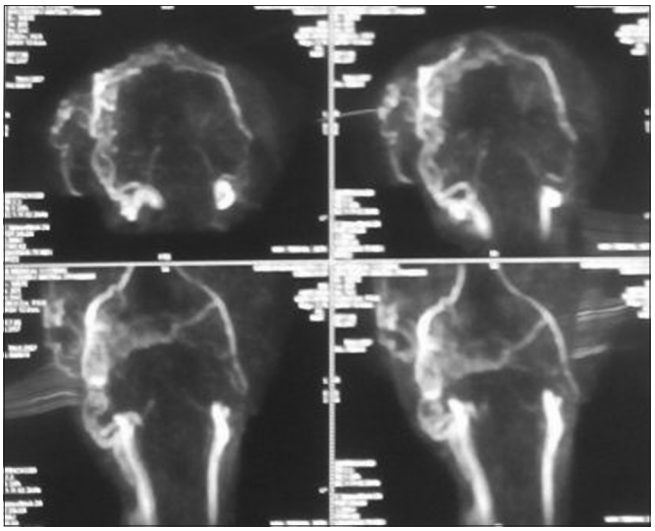

Figure 4: Magnetic resonance angiography

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