Case Report

Arteriovenous malformation of mandible: Extracorporeal curettage with immediate replantation technique

Virendra Singh, Pramod Kumar Bhardwaj

Department Oral & Maxillofacial Surgery, Government Dental College, PGIMS, Rohtak, Haryana, India

Address for correspondence:
Dr. Pramod Kumar Bhardwaj, Department Oral & Maxillofacial Surgery, Government Dental College, PGIMS, Rohtak, Haryana, India.
E-mail: prm2525@gmail.com

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ABSTRACT

Arteriovenous malformations of jaw are extremely rare conditions that can result in disastrous complications, if handled carelessly. Although various treatment modalities have been advocated in the literature, there seems to be no complete consensus on a suitable treatment in these cases. This report highlights the importance of correct diagnosis and early treatment in management of vascular malformations. Extracorporeal curettage followed by immediate replantation yielded good results in our case and this technique can emerge as a valid alternative, especially in developing countries.

Key words: Arteriovenous malformations, extracorporeal curettage, jaw

Introduction

Arteriovenous malformations (AVMs) of the mandible are rare and challenging entities. The lesion can present with a variety of clinical findings depending on the severity of malformation. Interosseous AVM often remains undiagnosed until a dramatic bleeding incident occurs, which is usually a result of dental manipulation. AVMs have a high propensity to bleed, which may be life threatening.[1]

Various treatment modalities have been described for treatment of high flow AVM, most of them should be adjunct to surgery. Resection of the mandibular fragment containing lesion has been considered essential for complete healing.[2] Curettage of the resected fragment with immediate replantation reduces the morbidity associated with the procedure and the difficulty of reconstruction. This seems to be a good approach.

Following the above logic, here we present a case where AVM of mandible was treated with extracorporeal curettage of lesion and immediate replantation of segment.

Case Report

A 16-year-old female patient reported to emergency department with a massive bleeding episode after extraction of tooth, which could only be controlled after biting on gauze piece. Bleeding stopped only after applying sustained local pressure over the extraction site. History revealed that left mandibular second premolar was carious and associated with persistent pain and swelling in the region for the past 2 years. When swelling did not subside even after taking antibiotics, local dentist extracted the tooth without taking any radiographs, resulting in massive uncontrolled bleeding. On detailed examination after 12 hours, a soft, pulsatile, tender swelling with audible bruit was noted in left submental region [Figure 1]. The overlying skin was normal in color and there was no paresthesia of the involved region. Intraoral examination revealed extraction socket of 35; mobile 34, 36. Marginal gingival bleeding was also noted i.r.t 33, 34, 36, 37 with buccal expansion of mandible. Rest of the physical examination was unremarkable and other blood investigations were normal. At this stage, the patient refused treatment and returned after few months; meanwhile, swelling had increased to the

present size and the patient had suffered from two bleeding episodes.

Colored ultrasonography of soft submental swelling revealed high flow lesion with multiple feeders. Orthopantmograph (OPG) showed extrusion of 33, 34 with displacement and tilting of adjacent teeth. The ill-defined moth eaten type radiolucency involved left mandibular parasymphysial body region; also inferior alveolar canal was dilated and tortourous with enlarged mental foramen [Figure 2]. Axial, coronal sections of computed tomography (CT) scan revealed multiseptae central bony destructive lesion with expansion of bicortical plates without perforation [Figures 3a, b]. Needle aspiration from some distance away from the lesion resulted in syringe full of bright red blood. The patient was diagnosed to be a case of AVM mandible.

Surgical technique

The patient was taken to the operating room and elective tracheostomy was done to maintain airway postoperatively in case of massive swelling. Hypotensive general anesthesia was maintained producing systolic pressure of approximately 70 mm Hg. A wide exposure was attained with large submandibular incision extending to submental region. Dissection was done to identify common carotid artery. After achieving proximal and distal control, the external carotid artery was temporarily ligated above superior thyroid artery. Engorged vessels supplying to AVM were identified and ligated. Osteotomy cuts were given in buccal cortical plates away from the lesion, and using bone spreader, inferior alveolar artery was identified and ligated. Three units of blood were transfused during resection of the segment. Using extracorporeal technique, teeth were extracted in the involved segment, followed by curettage and hollowing of mandible with curretes, rotating handpiece and bur [Figures 4 and 5]. No graft was placed inside the cavity. Stabilization of reimplanted mandibular piece was done with three 2.5 mm miniplates [Figure 6]. Intermaxillary fixation was done on the contralateral side to achieve occlusion and strength. Postoperative course was uneventful. During follow up of 2 years, ultrasound revealed no new vascular lesion in the region while excellent facial form and function was maintained [Figures 7 and 8].

Figure 1: Soft, tender, pulsatile swelling in left mandibular region

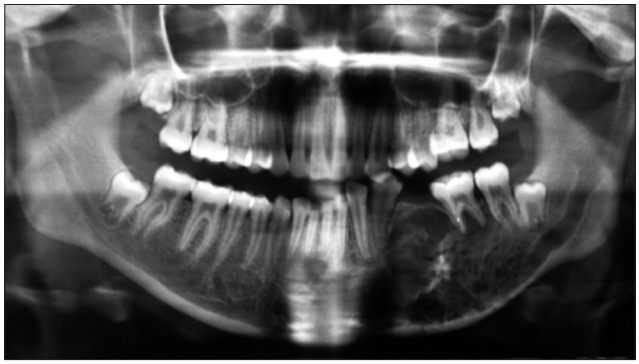

Figure 2: Preoperative OPG showing moth eaten irregular radiolucency in left mandibular parasymphysis body region with enlarged and torturous mandibular canal; adjacent teeth are displaced

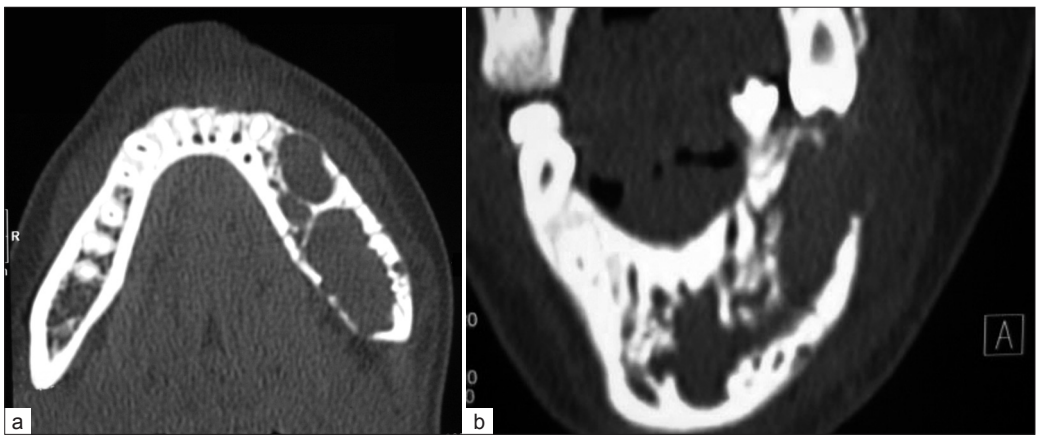

Figure 3a, b: Axial CT and Coronal CT section demonstrating widespread destruction and thinning of cortical plates.

Discussion

International Society for the Study of Vascular Anomalies has classified them as hemangioma (with endothelial proliferation) and vascular malformation (VM) (with normal endothelial tumor) relying on 1982 biologic classification of Mulliken and Glovacki[3] based on endothelial characteristics. The VMs can be further categorized as low flow lesions (capillary, venous, lymphatic malformations) and high flow lesions (AVMs, arteriovenous fistulae) according to blood flow characteristics.[4] AVMs are most common high flow lesions.

Intraosseous VMs of the maxillofacial region sometimes give rise to dental emergencies and may cause disfigurement, morbidity and even death.[5,6] The proximity of the teeth can prove disastrous, like in our case where extraction of tooth resulted in massive hemorrhage. A review of fatal cases by Lamberg and others[5] shows that in most instances, exsanguination is the result of dental extractions, the dentist being unaware of existence of the AVM.

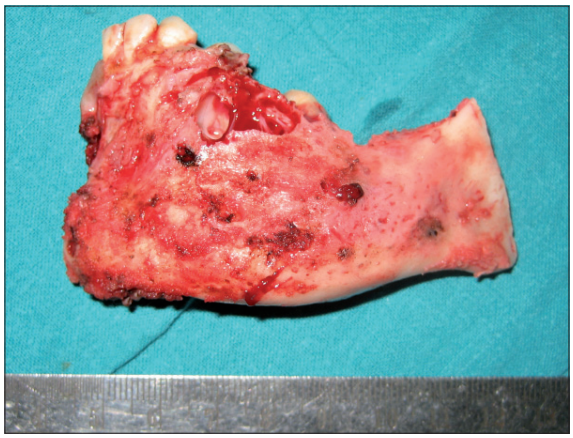

Figure 4: Resected mandibular segment containing focal lesion

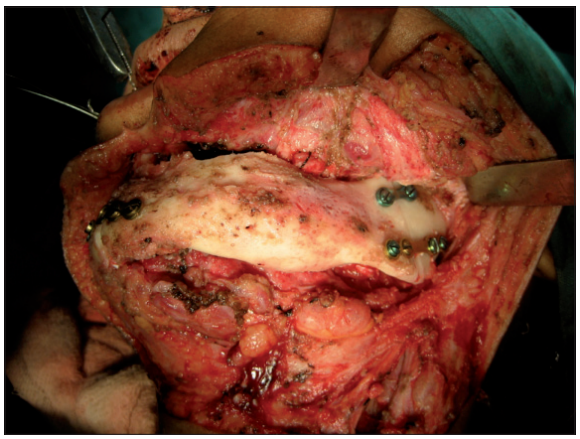

Figure 6: Reimplantation of mandibular segment and stabilization with miniplates

Vascular lesions of the jaws have an overall 2:1 female:male occurrence, with peak incidence in the second decade.[7,8] In this female patient of 16 years of age, persistent pain and mandibular swelling from the past 2 years were main concerns initially. Although more than 50% of vascular lesions occur in the head and neck region, only a small percentage of these occur in jaws.[8-10] They are twice as common in the mandible as in the maxilla.[5] Mandibular VM usually appears during adolescence, with extremes at 3 months and 74 years of age.[11] VMs are caused by a disturbance in the late stages of angiogenesis (truncal stage) and result in the persistence of AV anastomosis present during embryonic life. VMs, which usually present as developmental anomalies from birth, develop in proportion to physical growth. The increase in size of these VMs is asymptomatic and imperceptible at an early age and is promoted by local hemodynamic factors. The blood shunted to the malformation causes the lesion to grow, which in turn causes increased shunting of the blood, hence leading to a vicious cycle. This overgrowth can result from hormonal imbalances, vasomotor disturbances, infections or trauma.[12]

As has been seen in this lesion, it can present with a

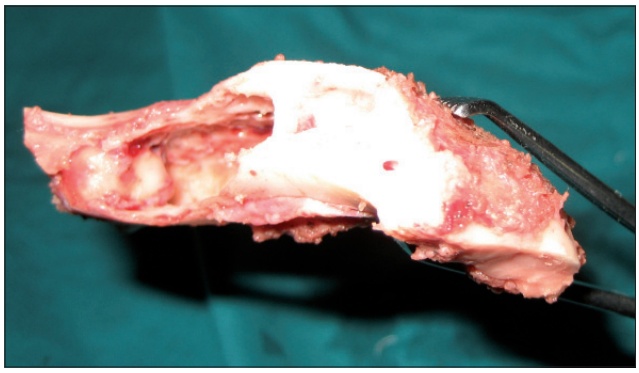

Figure 5: Extracorporeal technique of extraoral removal of teeth and curettage of lesion

Figure 7: Postoperative OPG showing good alignment of mandibular lower border and fixation with plates at symphysis and angle

Figure 8: Postoperative radiograph showing maintenance of normal anatomical contour

variety of clinical manifestations depending on the severity of the malformation; however, occasionally asymptomatic cases have also been reported in patients with AV malformation.[12,13] Some of the signs and symptoms reported are soft tissue swelling,[14] paresthesia,[13,15] pain of variable intensity,[16-20] teeth mobility and migration,[12,16,18,19,21] discoloration of overlying skin and intraoral mucosal surfaces,[5,15,17,21,22] facial asymmetry,[19,23] local pulsation,[5,12,19] noticeable bruit,[5,15,18,19,24] erythematous gingival and bleeding around the teeth[18,25] and bone resorption with palpable thrill as well as resorption of the roots in the affected area with no evident tooth-related cause or periapical pathoses.[13,18,19] Systemic findings like blurred vision, epistaxsis, paresthesia and cardiac abnormalities (murmur, hypertrophy and failure) have been reported.[25,26,27]

The radiographic appearance is quite variable and therefore unreliable as a sole basis of diagnosis. Gelfand[28] has summarized three typical radiographic appearances:

1. A sunray appearance created by trabecular bone between the vessels and osetolytic lesion.
2. A soap bubble or honeycomb appearance with occasional punched out areas.
3. An appearance described as ill-defined radiolucency.

Other radiographic findings may include cortical expansion as well as unilocular or multilocular cystic areas. Phleboliths, root resorption and lack of lamina dura have also been described.[28] According to Stafne,[29] radiographic appearance can resemble any destructive lesion of bone.

Numerous treatments in varying combinations and various degrees of success have been employed, including ligation,[25,30] embolization,[31,32,33] radical resection,[31] use of sclerosing solutions,[34] curettage and packing,[35] radiation,[24] bone wax packing in cavities followed by curettage, in two cases observation[35,36] and even cryosurgery.[37,38]

Embolization followed by surgical treatment is still the modern conventional approach.[38] Embolization reduces blood flow, allowing excision to be performed subsequently within 48 hours–2 weeks.[38,39] Embolization is also not without risk as embolic complications, allergic reactions, avascular necrosis of bone,[24] delayed root development,[40] defective mandibular growth have been reported. Resection of mandible can result in a variety of disabilities including impairment of speech articulation, salivary control, difficulty in swallowing, trismus and deviation of mandible toward the surgical side during functional movement.[41] This led to the concept of immediate reconstruction which was introduced by Weaver *et al*. who used the patient's own prefrozen mandibular bone.[42,43] Motamedi *et al*,[44] used this technique in AVM without freezing or autoclaving. The hollowed cortical shell acts as autogenous inductive tray. Greene *et al*,[18] advocate filling of particulate marrow in the cavity. As digital subtraction angiography and embolization treatment modality are not available in our institute, we had to rely on traditional method of careful ligation of feeder vessel to prevent excessive blood loss, and intraoperative blood infusion was done to prevent any complications.

This young female patient was followed up for almost 2 years and a good esthetic, facial symmetry and functional status was maintained. We have arrived at similar conclusion that this technique is safe, convenient and effective alternative to treat such vascular malformations and restore near exact form, function and symmetry without obviating the need for space maintainers, bone harvesting, future major reconstructive operations and keeping cost factor in check. Resection of involved portion followed by curettage and reimplantation can emerge as a valid alternative to secondary reconstructive surgery.

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