

## Case Report

**Extracorporeal Management of the Arteriovenous Malformation of Mandible**Attresh Gyanander<sup>1</sup>, Singh Virender<sup>2</sup>, Dhamija Nidhi<sup>3</sup>, Solanki Ravinder<sup>4</sup>, Singh Pardeep<sup>5</sup>, Sheoran Kirtika<sup>6</sup><sup>1</sup>Oral and maxillofacial surgery<sup>2</sup>Oral and maxillofacial surgery, Prof & Head Dept. of oral surgery Post Graduate Institute of Dental Sciences Rohtak, Haryana<sup>3</sup>Pedodontics Govt. Dental College, Amritsar<sup>4</sup>Oral Surgery, Asst. Professor Dept. of oral surgery, Post Graduate Institute of Dental Sciences Rohtak, Haryana<sup>5</sup>Post graduate Student, Prosthodontics, Post Graduate Institute of Dental Sciences Rohtak, Haryana<sup>6</sup>Post graduate student, Periodontics, Post Graduate Institute of Dental Sciences Rohtak, Haryana**\*Corresponding author**

Dr. Pardeep Singh

Email: [pradeepsheokand7@gmail.com](mailto:pradeepsheokand7@gmail.com)

**Abstract:** Intraosseous vascular malformations are very rare and pose a challenge for maxillofacial surgeon because of difficulty in their radiological diagnosis and appropriate management. Different authors have suggested an array of treatment modalities with varying degrees of success, but there is no complete consensus on a suitable treatment. Extracorporeal resection offers an advantage of preserving the outer cortical bony framework, thereby avoiding additional reconstructive procedure. This technique has given good results in our case and can emerge as a good alternative for the management of arteriovenous malformation.

**Keywords:** Arteriovenous malformation, Congenital, Extracorporeal, Intraosseous

**INTRODUCTION**

Arteriovenous malformations (AVMs) are vascular mal-development characterized by the presence of abnormal channels joining the arterial to the venous circulation without the interposition of a normal capillary bed [1]. All intraosseous AVM are moderate to high flow associated with central vascular and peripheral vascular channels.

Although more than 50% of all congenital haemangiomas are located in the head and neck, the incidence of intraosseous vascular malformations is very low [2]. The clinical presentations of these congenital abnormalities are variable and range from an asymptomatic birthmark to life-threatening exsanguinating hemorrhage [3]. Primary goal of any surgery for vascular lesion is twofold one is to eradicate the disease part and second is to harmonically balance the facial appearance [4]. In case of mandible inferior alveolar vessel is the primary vessel associated with collateral supply of sublingual and facial artery [5]. Traditionally segmental / hemimandibular resection is the only surgical treatment available which further demands primary or secondary reconstruction [6]. Extracorporeal resection offers an advantage of preserving the outer cortical bony framework, thereby avoiding additional reconstructive procedure. Few cases

have been mentioned in literature using this technique; hereby we are discussing our experience of management of AV malformation of mandible through extracorporeal technique.

**CASE REPORT:**

A 19 years old systemically healthy girl came to our department with the complaint of swelling on lower side of gums from last 1 month. She noticed the swelling 1 month back which was associated with bleeding while eating and such episodes recur at an interval of 6-7 days, swelling increases gradually in size, she had undergone extraction of her right 1<sup>st</sup> molar around 6 years back with the complaint of pain in the same region. On examination the face was asymmetrical with a diffuse extra oral swelling present lateral to angle of the mouth. Small petechiae patches were present extraorally on right side of face & in the palate. Intraorally a small swelling was present over right side of mandibular gingiva extending from right 1<sup>st</sup> premolar to right 1<sup>st</sup> molar region, occluding the occlusal surface and extending upto the lingual sulcus (Figure 1). Swelling was 1 X 1 cm in size, non tender, pulsatile, reddish blue in colour, solid in consistency, non fluctuant. Teeth were found to be displaced buccally in right premolar- molar region with erythematous gingiva on the lingual aspect, and on

palpation pulsation was evident on the erythematous gingiva.



**Fig 1: Preoperative intraoral picture of the patient showing the pathology**

OPG showed widening of inferior alveolar canal of right side along with disorientation of trabeculae in relation to right 1<sup>st</sup> molar region. CT scan revealed a lytic lesion showing soft tissue density in it, on CECT there was enhancement of the lesion equal to the vessels. Right side lingual and facial arteries were slightly more pronounced than normal. Needle aspiration from some distance away from the lesion resulted in syringe full of bright red blood with pressure. The patient was diagnosed to be a case of AV malformation of mandible.

**TREATMENT:**

The patient underwent surgical treatment under hypotensive general anesthesia, via nasal intubation. A wide exposure was attained with large submandibular incision extending to the submental region; anomalous vessels were explored and ligated. Corticotomy on mesial and distal side of the pathology was done in the healthy bone. The corticotomy site was expanded using smith's bone spreader to free the inferior alveolar vessel which was ligated on both sides. As in mandible main feeder vessel was inferior alveolar artery, this helped in reducing the amount of blood loss. Now the mandibular segment was resected and the teeth were extracted from the involved segment using extracorporeal technique (Figure 2).



**Fig 2: Intraoperative photograph of the patient revealing resected pathological mandibular segment (extracorporeal technique)**

This was followed by curettage and hollowing of the mandible with curettes, rotating hand piece and bur to remove the pathological portion completely. No graft was placed inside the cavity, and remaining bone

segment was used for reconstruction. This was stabilized with a K wire first followed by reconstruction plate (Figure 3).



**Fig 3: Intraoperative photograph revealing fixation of basal bony segment**

One unit of blood was transfused during the resection of the segment and postoperative course was uneventful. Postoperative orthopantomogram of the

patient was done and revealed proper uptake of bony segment (Figure 4).



**Fig 4: Postoperative orthopantomogram revealing fixation with reconstruction plate along with K- wire.**

#### DISCUSSION:

An AVM occurs due to abnormal communications between arteries and veins without the normal intervening capillary bed.[1] Expansion of an AVM is the result of increased blood flow through the lesion (not cellular proliferation) and subsequent tortuosity of adjacent feeding arteries and dilation of veins (collateralization and recruitment) [7].

The incidence of intraosseous vascular malformations is very low. Primary skeletal involvement occurs in the tooth-bearing bones, which are invariably expanded by the malformation [8]. Vascular lesions of the jaw have an overall 2:1 female:male occurrence, with peak incidence in the second decade [9, 10]. They are twice more common in the mandible as compared to the maxilla [11]. In our case also it was found in a 19 year old female patient in right mandibular region.

AVM are present at birth, but they may not be clinically evident or are misdiagnosed as a haemangioma [12]. They rarely expand in infancy or early childhood; more typically they grow proportionately starting in the late childhood. Usually, years go by before they pose a threat to the patient as the signs and symptoms of their fast flow nature become obvious. Nevertheless, rapid expansion can occur due to variety of factors like hormonal changes associated with pregnancy or puberty, local trauma, attempted excision or ligation [8]. Spontaneous bleeding from the gingiva in the molar region may reflect the presence of a life threatening vascular malformation [6]. In our case the patient had episodes of bleeding due to local trauma to the gingiva in the mandibular molar region.

Clinical diagnosis is confirmed by US and Color Doppler examination. On OPG there is widening of inferior alveolar canal with disorientation of trabeculae while CECT shows prominent and tortuous

feeder vessels, similar findings were seen in our study also. MRI and/ or MRA define the extent of soft tissue involvement. Angiography shows variable degrees of arterial dilation and tortuosity, arteriovenous shunting, and dilated draining veins [13].

Various treatments modalities in different combinations have been suggested like use of sclerosing solution, ligation, embolization, packing, radiation, cryosurgery, bone wax packing in cavities followed by curettage and radical resection [6]. A widely accepted mechanism of recurrence, in AVMs is based on the belief that microfistulae always exist in the tissue adjacent to the AVM. Therefore, an incomplete resection leaves undetectable microfistulae that later can respond to changes in hemodynamics, leading to enlargement of the microfistulae and recurrence of the lesion [14]. Therefore, Resection of the lesion with margins in normal bone has been advocated as the treatment of choice. As resection of the mandible results in major cosmetic & functional deformities, this led to the concept of immediate reconstruction which was introduced by Weaver *et al.*; who used the patient's own prefrozen mandibular bone [15].

This young female patient was followed up for more than 1 year without any structural functional and aesthetic abnormality. We believe that immediate reincorporation of the patient's own resected, curetted segment, without freezing or autoclaving, is a safe, convenient, and effective alternative to treat vascular malformation. This procedure primarily restore near exact form, function, and symmetry without adding to the surgical burden and obviating the need for space maintainers, bone harvesting, and future major reconstructive operations.

#### REFERENCES

1. Van Den Akker HP, Kuiper L, Peeters FL. Embolization of an arteriovenous malformation of

- the mandible. *Journal of Oral and Maxillofacial Surgery*. 1987 Mar 1; 45(3):255-60.
2. Tonner PH, Scholz J. Possible lung embolism following embolization of a hemangioma with fibrin glue. *Der Anaesthetist*. 1994 Sep; 43(9):614-7.
  3. Mohammadi H, Said-Al-Naief NA, Heffez LB. Arteriovenous malformation of the mandible: report of a case with a note on the differential diagnosis. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*. 1997 Sep 1; 84(3):286-9.
  4. Hoey MF, Courage GR, Newton TH, Hoyt WF. Management of vascular malformations of the mandible and maxilla: review and report of two cases treated by embolization and surgical obliteration. *Journal of oral surgery (American Dental Association: 1965)*. 1970 Sep 1; 28(9):696-706.
  5. Hamparian AM. Blood supply of the human fetal mandible. *Developmental Dynamics*. 1973 Jan 1; 136(1):67-75.
  6. Schneider C, Wagner A, Hollmann K. Treatment of intraosseous high flow arteriovenous malformation of the mandible by temporary segmental ostectomy for extracorporeal tumour resection: a case report. *Journal of Cranio-Maxillofacial Surgery*. 1996 Oct 1; 24(5):271-5.
  7. Mulliken JB: Vascular anomalies. In Aston SJ, Beasley RW, Thorne CHM, editors: *Grabb and Smith's plastic surgery*, Philadelphia, 1997, Lippincot-Raven.
  8. Kohout MP, Hansen M, Pribaz JJ, Mulliken JB. Arteriovenous malformations of the head and neck: natural history and management. *Plastic and reconstructive surgery*. 1998 Sep 1; 102(3):643-54.
  9. Willis RA, *Pathology of Tumors*. St Louis, MO: CV Mosby; 1948.
  10. Stanley E. *A treatise on diseases of bone*. Philadelphia, PA: Lea and Blanchard Co; 1849.
  11. Lamberg MA, Tasanen A, Jääskeläinen J. Fatality from central hemangioma of the mandible. *Journal of oral surgery (American Dental Association: 1965)*. 1979 Aug; 37(8):578-84.
  12. Marler JJ, Fishman SJ, Upton J, Burrows PE, Paltiel HJ, Jennings RW, Mulliken JB. Prenatal diagnosis of vascular anomalies. *Journal of pediatric surgery*. 2002 Mar 1; 37(3):318-26.
  13. Gold L, Nazarian LN, Johar AS, Rao VM. Characterization of maxillofacial soft tissue vascular anomalies by ultrasound and color Doppler imaging: an adjuvant to computed tomography and magnetic resonance imaging. *Journal of oral and maxillofacial surgery*. 2003 Jan 31; 61(1):19-31.
  14. Leipzig B, Yau PC. Massive congenital arteriovenous malformation of the pterygomaxillary space. *Otolaryngology--Head and Neck Surgery*. 1982 Jan; 90(1):48-51.
  15. Weaver AW, Smith DB. Frozen autogenous mandibular stent-graft for immediate reconstruction in oral cancer surgery. *The American Journal of Surgery*. 1973 Oct 1; 126(4):505-6.