Venous malformation of mandible – Case report of extracorporeal curettage with immediate replantation

Pritish Patnaik, Shreyas Sorake, Ashish Shetty, Lida Mary Nidhin Philip*, Athiramol C K

Email: lidmjg87@gmail.com

Abstract

Vascular anomalies are a rapidly evolving multidisciplinary field that combines several surgical and medical specialties. A maxillofacial surgeon plays an essential role in the management of patients affected with such lesions in the head and neck region. According to the World Health Organization, these lesions are considered as benign vasoformative neoplasms of endothelial origin. This article reports a case of venous malformation of mandible in a 6-year-old child, which presented as a post extraction complication.

Key words: Vascular anomalies, mandible benign tumours, post extraction complication

Introduction

Intraosseous vascular lesions are quite rare conditions, comprising less than 1% of all intraosseous tumours. It mainly occurs in the vertebral column. It rarely involves the mandible. Peak incidence is noted between the second and fifth decades of life, and females are twice more affected than males. Diagnosis and treatment has been hindered by a confusing and largely descriptive nomenclature. The term “intraosseous hemangioma” has been used indiscriminately for intraosseous lesions, including both hemangiomas and vascular malformations. However, the major difference between them is the existence of an increased endothelial cell turnover (vascular neoplasms), which is ultimately determined by identifying mitoses on histopathology. The term “hemangioma” usually refers to the common infantile hemangioma. There is no evidence of this occurring in bone, although it can deform adjacent skeleton or cause minor osseous overgrowth. The authors describe a case of intraosseous venous malformation of the mandible in a paediatric patient.

Case report

A young 6-year-old female was presented with excessive bleeding from extraction socket of lower left deciduous molar. History revealed the extraction of lower left deciduous first molar tooth done by general dentist a day prior due to mobility and swelling since six months.

Post extraction, there was considerable bleeding from the socket which was arrested with pressure packing. Few hours later, the patient complained of excessive bleeding from the extraction socket.

Pritish Patnaik1, Shreyas Sorake2, Ashish Shetty3, Lida Mary Nidhin Philip3*, Athiramol C K4

1Medical officer (Dental) Ganjam, Odisha
2Reader, Department of Oral and Maxillofacial Surgery, A J Institute of Dental Sciences, Mangalore, Karnataka
3Senior Lecturer, Department of Oral and Maxillofacial Surgery, A J Institute of Dental Sciences, Mangalore, Karnataka
4Post graduate student, Department of Oral and Maxillofacial Surgery, A J Institute of Dental Sciences, Mangalore, Karnataka

*Corresponding Author

How to cite this article: Pritish Patnaik, Shreyas Sorake, Ashish Shetty, Lida Mary Nidhin Philip, Athiramol C K. (2019). Venous malformation of mandible – Case report of extracorporeal curettage with immediate replantation. 4(1), 34-37.
profuse reactionary haemorrhage from the socket with swelling gradually increasing in size.

On extra oral examination, 5cmX4cm diffuse bony hard swelling over lower left part of the face was noted. (Fig. 1) Intraorally, the patient had a pressure pack over the socket which when released resulted in immediate haemorrhage, and hence was replaced with a fresh haemostatic pressure pack. OPG revealed an ill-defined lesion in the body of left mandible with a soap bubble appearance, with neither root resorption nor root flaring. (Fig. 2).

Contrast enhanced computed tomography (CECT) exhibited a large 40x25x12mm bony cystic lesion at left mandibular angle region extending into body region with internal hyperdense soft tissue component. CT angiography was not done as the patient was not willing to bear the expenses. Patient was then planned for surgery under high-risk consent. After careful nasotracheal intubation, without disturbing the pressure pack, cervical incision was placed from chin to mastoid.

Subplatysmal flap was raised, Sternocleidomastoid muscle retracted posteriorly to expose the carotid sheath at the level of the greater cornu of the hyoid bone. External carotid artery distal to the carotid bifurcation was identified. This was done to achieve hemostasis during the excision of the lesion. Dissection was then carried to the body of the mandible. Facial, inferior alveolar and mental vessels were identified, ligated and divided. Controlled digital compression was applied over the external carotid. Mandible body was completely exposed to the field.

Osteotomy cuts were placed anterior and posterior to the lesion and much away from it. Mandible was resected en-bloc along with the pathologic tissue contiguous to the excised segment. (Fig. 3) After resection of mandibular segment, hemostasis was applied by local measures. Extracorporeal curettage was done (Fig. 4) and affected bone were burred out.

Soft tissue specimen was sent for histopathological examination. The remaining cortical segment was re-implanted as free graft and stabilized with 2.5 mm low profile titanium locking recon plate secured with 2X8mm screws (Fig. 5). Drain placed over the wound and layered closure was done. Patient was extubated with stable vital and recovered.
uneventfully. The histopathological examination of the curetted material demonstrated fibro-collagenous tissue with numerous blood vessels with nil mitotic activity. Follow up after six months exhibited good facial form and function with no signs of avascular necrosis of the graft. The patient remains under follow-up without significant findings to the day.

Figure 5: Re-implantation of remaining cortical segment and stabilize using screws

Permission was taken from the institutional ethics committee to publish the case report.

Discussion

Understanding the various vascular anomalies is of immense significance for their accurate diagnosis and subsequent appropriate treatment. Many a time, authors reporting cases on intraosseous vascular anomalies fail to understand and distinguish the binary classification terming all the lesions under an umbrella term “haemangioma”. The authors call such lesions “tumours”. Yet, the histological sections usually demonstrate slow-flow, abnormal vascular channels lined by quiescent endothelium. The correct diagnosis in all of these patients is intraosseous venous malformation. In 1996, the International Society for the Study of Vascular Malformations authorized the binary classification. The binary classification (tumours and malformations) of vascular anomaly into tumour and malformation is straightforward and easy to understand. Tumours include haemangioma, haemangiobenodtheiomas and angiosarcomas, whereas malformations may be slow flow (capillary, lymphatic, venous) or fast flow (arterial/combined).

Usually, these lesions remain asymptomatic although may present signs and symptoms including a slow growing mass with or without discoloration, discomfort, pulsatile sensation and mobile teeth. Intraosseous vascular lesions may prove disastrous as seen in this case, where the mobility of deciduous tooth associated with swelling was extracted without definite clinical diagnosis. Hence, mobile teeth should not be empirically diagnosed as periodontitis or simple periapical pathology.

Disturbances in the truncal stage of angiogenesis is responsible for the persistence of arterio-venous anastomosis present during embryonic life. The vascular lesions present as developmental anomalies from birth develop in proportion to physical growth. They are asymptomatic and their growth is promoted by local hemodynamic factors. A vicious cycle starts with the growing lesion causing increased blood supply that in turn enhances its growth. Hormonal imbalances, vasomotor disturbances, infections or trauma might also have a contributory effect.

Advances in imaging have led to the unnecessary exposure of many lesions. Ultrasound and Doppler analysis are useful in establishing the flow dynamics of a lesion. MRI has a major advantage over CT or angiography in differentiating hemangiomas from the surrounding structures, but its high cost and limited availability are its limitations. Imaging in this case was also restricted to contrast enhanced CT for cost reasons. Angiography and subtraction angiography has a specific but limited role in the diagnosis of vascular lesions. It is restricted to lesions requiring therapeutic endovascular intervention. Selective embolization as a single treatment modality is rarely successful because of rapid establishment of new pathways of flow. When embolization is used, surgery must be done within 24–48 hours to prevent the development of collateral blood supply. Sclerotherapy has a promising but limited role in the management of vascular lesions and is successful in the treatment of macrocystic lesions.

Surgery has been used effectively to eradicate or minimize the lesion. The only trouble that may arise during excision of the lesion is profuse haemorrhage. The technique recommended by Nair.
et al. in his review of 115 cases gave its imprimatur to this simple procedure of external carotid artery control.\textsuperscript{10}

We used this simple technique and found it extremely effective during the osteotomy. Mandibular defects following resections impact both form and function and requiring a definite approach to optimize functional and cosmetic outcomes. Mandibular defects in the past have been traditionally reconstructed using non-vascularized bone grafts such as rib and iliac, or vascularised grafts such as fibula depending on the size and location of the defect. Non-vascularized bone grafts continue to be well suited for small (less than 5 cm) mandibular defects with adequate soft tissue coverage in non-irradiated bone.

The concept of immediate reconstruction was introduced by Weaver et al. who used the patient’s own prefrozen mandibular bone in 1973. Behnia and Motamedi were the first to perform an immediate replantation without freezing or autoclaving of the resected AV mandible in a 11 year old patient with three year follow up and concluded that this technique is safe, convenient and effective alternative to treat vascular malformations and 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.

In this case, we adopted two simple procedures, external carotid artery control along with re-implantation of the resected bone to effectively manage a patient with vascular malformation of the mandible manifested as a life threatening post extraction complication. The case will continue to be under strict follow up as the child continues to grow into adolescence and adulthood to rule out any late recurrence and the fate of the re-implanted mandible.

References