Arteriovenous Malformation of the Mandible in a Pediatric Patient: A Case Report

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Abstract

Arteriovenous Malformations (AVMs) are rare vascular anomalies characterized by abnormal arteriovenous shunts, typically lacking a connecting capillary bed. These lesions often result from embryogenic vascular developmental abnormalities. AVMs can manifest with various clinical symptoms, including hemorrhage, seizures, and neurological deficits. Here, we present a case of a 10-year-old male with an AVM involving the right mandible, presenting with intraoral bleeding, facial asymmetry, and engorged veins. The patient underwent excision of the lesion with right hemimandibulectomy, and no postoperative complications were observed.

AVMs of the face are infrequent, but when they occur in the mandibular region, hemorrhage is a common clinical presentation. Treatment modalities include microsurgery, embolotherapy, and radiosurgery. However, recurrence remains a concern, particularly in pediatric cases.

This case underscores the importance of recognizing AVMs in pediatric patients to facilitate early diagnosis and appropriate management, which can alleviate symptoms, prevent life-threatening complications and reduce mortality. Despite their rarity, AVMs should be considered in the differential diagnosis of facial asymmetry and intraoral bleeding, especially in children. Timely intervention, as demonstrated in our case, can improve outcomes and enhance the quality of life for affected individuals.

Key words: Arteriovenous malformation; Intraoral bleeding; Mandible.

Introduction

Arteriovenous malformations may be defined as vascular malformations characterised by arteriovenous shunt through a collection of tortuous vessels without a connecting capillary bed.[1] The pathogenesis of such lesions are not clear but Martin and Vinters have given a suggestive explanation.[2] Most of the lesions occur due to the abnormality in the embryogenic development of the vascular system.[3]

The prevalence of AV malformations is estimated to be around 140 to 500 per 100,000 people , which affects around 0.14- 0.15% of the population.[4,5] Half of the patients with AV malformations present with haemorrhage.[6] It has been reported that children are more susceptible to haemorrhagethan adults.[7]

Other clinical manifestations include seizures (30%) and gradual neurological deficit (12%) and some patients also reported to have headaches.[6] The main aim of treatment is complete angiographic obliteration of the arteriovenous malformation.[1] There are three main treatment modalities- microsurgery, endovascular embolization and radiosurgery, of which microsurgery is regarded as the gold standard of treatment and endovascular embolization to be the least successful when done alone.[1] However even after treatment, there is a risk of recurrence, especially in children.[7,8]

Case Presentation

Our patient, a 10-year-old male, student, hailing from Dhaka city, got admitted in 05th June 2020 at Dhaka Dental College and Hospital with the complaints of swelling in the right lower jaw for 3 months along with frequent intraoral bleeding from the site for the same duration. He also reported occasional pain in the swelling for 15 days. He was unable to take any hard and chewy foods for last couple of days due to pain and bleeding.He was on liquid diet.The patient noted that the swelling was initially small and increased gradually in size. He got himself admitted in the hospital for management of bleeding from the swelling. He has no other significant history.

On physical examination, he appeared ill-looking and anxious with an average body built and weighed around 39 kgs. Moderate anaemia was also present. cyanosis, jaundice, clubbing, dehydration, oedema, skin pigmentation were

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absent. Further examination revealed pulse 74 beats/min, respiratory rate 14 breaths/min, blood pressure 100/70 mm of Hg and temperature 37.2 C. There was an engorged vein visible on the right side of his cheek. He has no palpable lymph nodes and examination of all other systemic examinations revealed no abnormalities.

Upon inspection of the extra oral region, there was facial asymmetry present in the right side along with visible pulsation and engorged vein in right mid cheek. On palpation, local temperature was raised, pulsation present with no tenderness. On inspection of the oral cavity, there was a well-defined mass in the right side of the lower gingivoalveolar mucosa, approximately 3cm X 2cm which extended from the second premolar to RMT antero-posteriorly and involved the lower buccal vestibule to mid inner cheek inferio-superiorly. Upon palpation, the surface of the mass was smooth, had a well-defined border, was soft to firm in consistency, non-tender, compressible and did not bleed on touch. Tongue movement was normal in all direction, oral hygiene was good, there was no missing teeth and a 3rd degree mobile tooth was present, namely lower right second premolar. Based on the history and physical examination, a provisional diagnosis of vascular tumour of right side of the jaw was made. The differential diagnoses were haemangioma, vascular malformation, pyogenic granuloma and central giant cell tumour.

Routine investigations such as random blood sugar, serum creatinine, HBsAg, anti HCV, Chest X-ray P/A view and ECG revealed no abnormalities. Results of complete blood count revealed haemoglobin is 7.5 gm/dl and moderate decrease in RBC count. Specific investigations to confirm the diagnosis were ordered which included Orthopantomogram X-ray, CT angiogram and MRA of maxillofacial region with right carotid area. The films are shown below.

CT angiogram of neck vessels revealed AVM involving ramus of right hemi-mandible with the mass feeded by right inferior alveolar artery and drained into jugular venous system. CT scan of the maxillofacial region revealed radicular cyst like lesion involving the root of the right lower premolar and molar teeth.

So this case is diagnosed as AVM on right mandible (Part of body and ramus).

Excision of lesion with right hemi mandibulectomy under GA was done with primary closure. 3 units of fresh human blood were transfused per-operatively. No post-operative complications were observed and patient was discharged with advice for monthly follow up. Broad spectrum antibiotics and a mouthwash was prescribed on discharge. The author have taken appropriate consent from the patient's guardian that his images and other clinical information to be reported in the journal and necessary permission was obtained from the proper authorities before start the study.

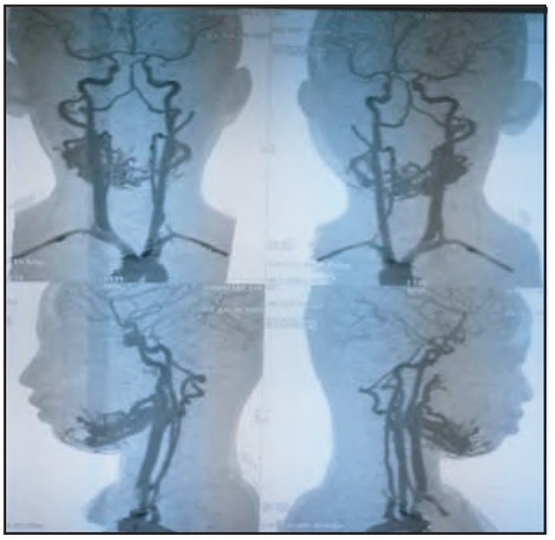

Figure 1 Large expansile lytic lesion in the ramus of the right hemi-mandible with increased inferior cortical destruction

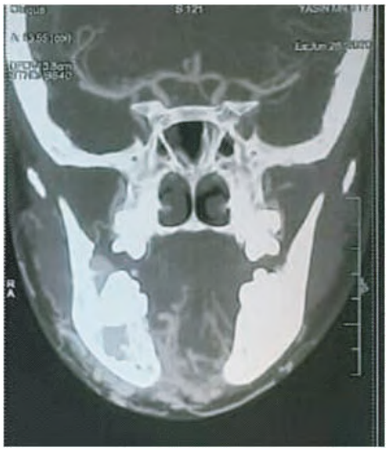

Figure 2 A well-defined radiolucent lesion measuring about 47mm X 16 mm adjacent to the roots of the right lower molar and premolar teeth. Post-contrast scan shows mild inhomogeneous enhancement

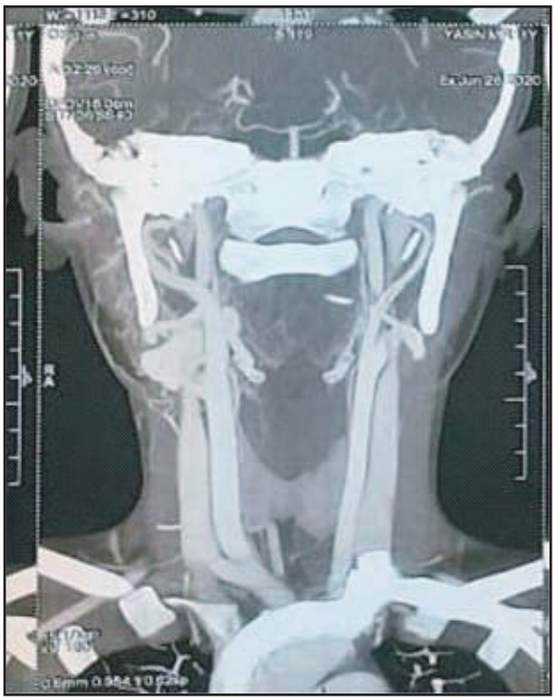

Figure 3 Involved right inferior alveolarartery,a branch of maxillary artery of ECA is visible here

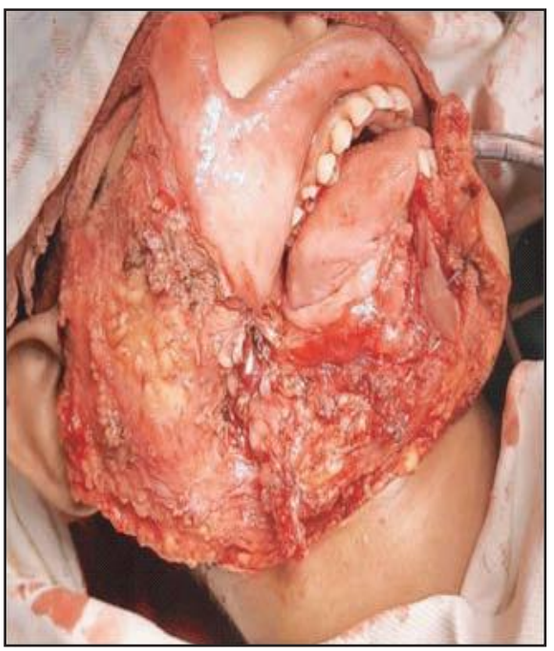

Figure 4 Per-operative picture is showing the excised area after right sided hemi-mandibulectomy with disarticulation

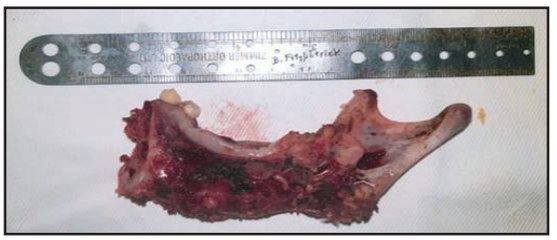

Figure 5 The excised mass with right hemi-mandible including condyle.

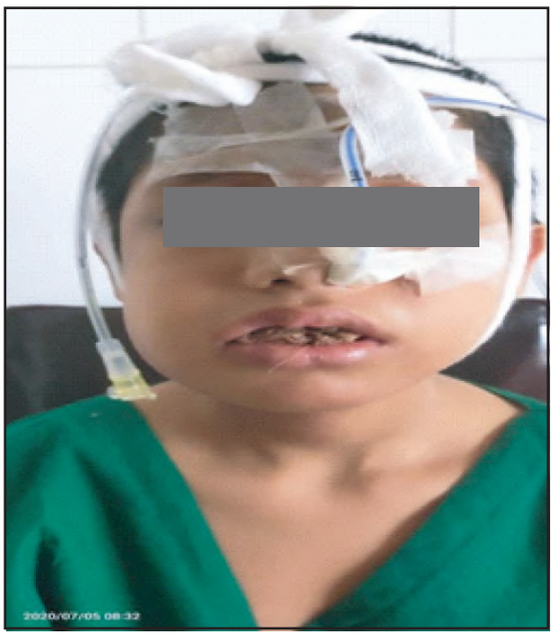

Figure 6 Pre-operative image

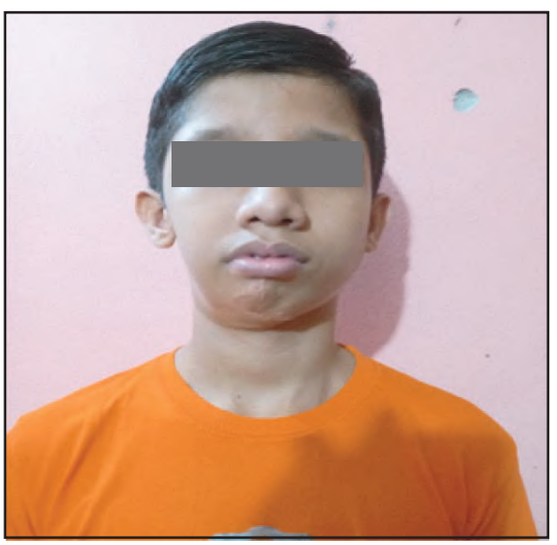

Figure 7 Follow up after 3 years of surgery. His parents are interested to reconstruct the area by vascularised free fibula flap due to aesthesis and functional issues

Discussion

Arteriovenous malformations are quite rare lesions.[9,10] Even though arteriovenous malformations of the face occur infrequently, most of the bony lesions occur in the mandibular region.[11] Hemorrhage is the most common clinical feature of this lesion which was presented by our case. The main treatment options are surgery, embolotherapy and radiosurgery. Recurrence is a common side effect even after surgery, even though our patient did not report any difficulty during the follow up period.

Limitation

Only one case in a single centered.

Conclusion

Even though AVM are rare lesions presented in childhood, appropriate diagnosis and management can relieve the patient from presenting difficulties, life threatening complications and widely reduce mortality.

Recommendation

Further research is warranted to better understand the pathogenesis and recurrence mechanisms associated with AVMs, particularly in the pediatric population.

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Contribution of author

Whole study was performed by the author himself.

Disclosure

The author declared no competing interest.

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