

Arteriovenous Malformation of the Mandible: A Case Report

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ABSTRACT

An arteriovenous malformation (AVM) is an abnormal shunt between arteries and veins that bypass the capillary bed. A 38-year-old female presented with a carious, non-restorable maxillary molar requiring extraction. The radiographic evaluation revealed the incidental finding of an asymptomatic radiolucent lesion of the right mandibular ramus. Based on clinical and radiographic findings in association with the location of the lesion, the patient was treated in the hospital setting. Further evaluation of the area revealed the lesion to be of vascular origin, requiring embolization. The incidental discovery of this radiographic finding underscores the need for extreme caution when considering surgical intervention in the presence of unexplained bony changes, as an AVM of the head and neck poses a risk of acute hemorrhage. Comprehensive imaging, thorough diagnostic workups, and proper biopsy protocols with these associated lesions play a critical role in identifying life-threatening vascular pathologies.

INTRODUCTION

An arteriovenous malformation (AVM) is an abnormal shunt between arteries and veins that bypasses the capillary bed. An AVM of the mandible is regarded as a rare entity that poses the risk of life-threatening hemorrhage. The International Society for the Study of Vascular Anomalies (ISSVA) classifies AVMs of the mandible as high flow vascular malformations that can be either congenital or acquired, with most being congenital. The distinction between different anatomic subtypes of AVMs within the mandible has also been adopted by further clinical literature and includes intraosseous AVMs, extraosseous AVMs, and combined lesions affecting both the bone and soft tissue [1]. The complications from these direct communications between arteries and veins depend on their location and developmental stage. These AVM complications can include stroke, hypoxemia, and life-threatening hemorrhage. Current treatments such as surgery, embolization, or radiosurgery aim to mitigate these risks. Regardless, AVMs can recur in about 25% of patients within the first year after treatment due to reperfusion from previously unrecognized vessels [2]. The need for non-surgical medical treatments for those malformations with multiple or diffuse AVMs exists in scenarios in which surgery is not safe or feasible. The limited availability of FDA-approved drugs for AVMs reflects the poor understanding of the development and progression of these malformations.

Emerging evidence indicates that a germline defect is insufficient for AVM development, suggesting additional somatic mutations and pro-angiogenic stimuli are also required. Insights from animal models show that the endothelial cell (EC) is the key cell type in AVM formation. For possible new therapies to succeed, the pathways that alter EC behavior must be identified. Research has shown that a single mutation, whether somatic or germline, can lead to a complex interplay between multiple cellular pathways.

The natural history of AVMs suggests they progress over time, causing complications like bleeding, pain, and tissue destruction. Studies of cutaneous AVMs in patients with hereditary hemorrhagic telangiectasia (HHT) have outlined a three-stage developmental sequence. First, a dilation of postcapillary venules surrounded by inflammatory cells develops. Next, connections form between arterioles and these venules. Once these connections form, the capillary bed disappears and is replaced by the direct arteriovenous communication. This three-stage development has also been described in pulmonary AVMs using CT scans.

The three-hit hypothesis attempts to explain the presence of familial AVMs as these malformations appear in seemingly random locations despite the presence of a causative mutation in every cell. Primarily from animal studies, this three-hit hypothesis suggests that the first hit is the loss of one functional gene allele. In animal studies, mice heterozygous for HHT genes rarely developed AVMs, suggesting a single mutation is not sufficient. The second event is a local loss of protein via loss of heterozygosity through an additional somatic mutation or protein shedding. The third event is an angiogenic stimulus consistent with inflammation or a dermal wound. Animal studies show that even a complete loss of protein was not enough to trigger AVM formation, suggesting an angiogenic stimulus was also required. This hypothesis suggests AVM development in mice needs a combination of a germline mutation, a local loss of protein, and an angiogenic stimulus [3].

While many AVMs are isolated, these malformations are a cardinal sign of several syndromes. HHT presents with AVMs commonly found in the brain, lungs, and liver. HHT is caused by loss-of-function mutations in genes like ENG or ACVRL1, which are crucial for the TGF-beta signaling pathway and proper vascular development. Another syndrome, Capillary Malformation-Arteriovenous Malformation (CM-AVM) syndrome, is caused by mutations in RASA1 and EPHB4. The observation of a somatic mutation in addition to a germline variant in a patient with CM-AVM syndrome further supports components of the three-hit hypothesis. PTEN hamartoma tumor syndrome is another genetic condition associated with AVMs, often present in the extremities [3].

In non-syndromic, sporadic AVMs, somatic mutations are hypothesized to be the cause. Furthermore, determinants like blood flow and shear stress have been identified as contributors to AVM formation. ECs that have lost endoglin, a protein mutated in HHT, migrate with the direction of blood flow, supporting the idea that AVMs may originate on the venous side of the capillary bed [4].

AVMs of the jaw are uncommon but require careful detection, including auscultation, to prevent adverse complications. Additionally, performing an aspiration of a cyst-like lesion of the jaw is standard biopsy protocol. The aspiration assists in recognizing the presence of AVMs and helps mitigate risk prior to surgical access and biopsy of these lesions. A positive aspiration of bright red heme suggests the presence of a vascular lesion, whereas the lack of heme is an imperative negative for proceeding with surgical intervention. In the event of a positive heme aspiration,

further vascular studies including a computed tomography angiogram is indicated to determine the appropriate treatment [4].

CASE PRESENTATION

A 38-year-old female presented with complaints of mild, intermittent pain in the left maxillary region associated with a carious, non-restorable maxillary molar tooth indicated for extraction. The patient reported no prior symptoms, history of trauma, radiation, infection, nor systemic disease. Despite the patient reporting no other orofacial symptoms, radiographic examination revealed the incidental finding of a cyst-like expansion of the right mandibular ramus. Clinical examination revealed no signs of infection, trauma, nor bony expansion in the region associated with the lesion of the right mandibular ramus. The patient denied neural changes and tenderness upon palpation of the associated area intra- and extra-orally.

Investigations

A panoramic image was captured to initially evaluate carious dentition, incidentally, revealing the well-demarcated, partially corticated radiolucency of the right ramus. A cone beam computed tomography (CBCT) scan of the affected region demonstrated a radiolucent lesion with cyst-like expansion of the right ramus extending from the inferior alveolar canal towards the posterior mandible and sigmoid notch. Buccal and lingual expansion in addition to buccal and lingual cortical breakthrough of the partially corticated 1.7 x 2.6 x 3.0 cm lesion were appreciated on the CBCT.



Figure 1: Panoramic image of right mandibular ramus lesion pre-operatively

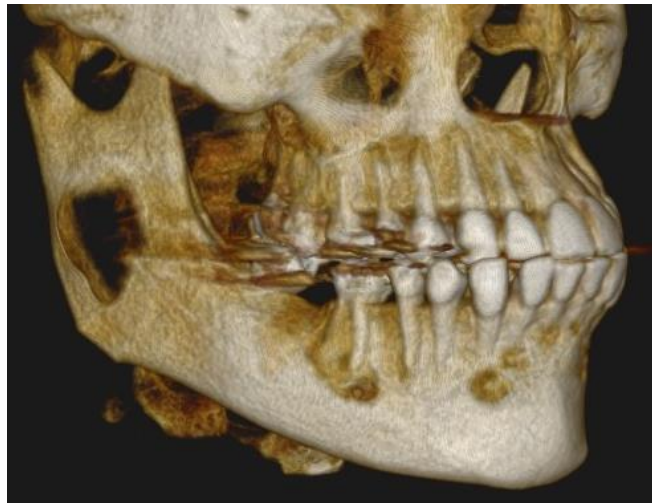


Figure 2: Three-dimensional rendering of right mandibular ramus lesion pre-operatively

Differential Diagnosis

Based on initial clinical and radiographic findings, the differential diagnosis included ameloblastoma, AVM, central giant cell granuloma (CGCG), odontogenic keratocyst (OKC), and odontogenic myxoma.

Treatment

The patient was taken to the operating room for further evaluation and possible biopsy of the right ramus lesion. An aspiration of the lesion was performed per standard biopsy protocol of cyst-like lesions. The aspirate from the lesion revealed minimal heme without keratinaceous debris or cystic components. The lesion was carefully exposed in the anterior and medial aspects of the ramus for further evaluation due to insignificant findings with aspiration. Brisk bleeding and tortuous vessels were encountered during the exposure. The hemorrhage was controlled using local measures. However, these clinical findings suggested the presence of an AVM. The procedure was aborted and neurointerventional radiology (NIR) was consulted for diagnostic work-up and management.

The patient underwent a diagnostic cerebral angiogram with NIR, and the diagnosis of AVM was confirmed. Following definitive diagnosis, informed consent was obtained, and a therapeutic cerebral angiogram was performed via transfemoral approach. The AVM was supplied by the internal maxillary artery, with a prominent contribution from the inferior alveolar branch. A large aneurysmal dilation, corresponding to the cyst-like expansion of the right ramus seen on imaging, was identified proximal to the fistulous point. Venous drainage was into the pterygoid venous plexus and right internal jugular vein, with reflux into the right superficial temporal vein, inferior petrosal sinus, and right cavernous sinus.

Endovascular treatment was performed via microcatheter navigation distal to the aneurysmal formation and fistulous site. Coil embolization was achieved with sequential deployment of Penumbra PAC400 and packing coils, resulting in complete occlusion of the aneurysm and shunt. Post-embolization angiography confirmed preservation of normal arterial branches and absence of residual fistula.



Figure 3: Angiogram intraoperatively.

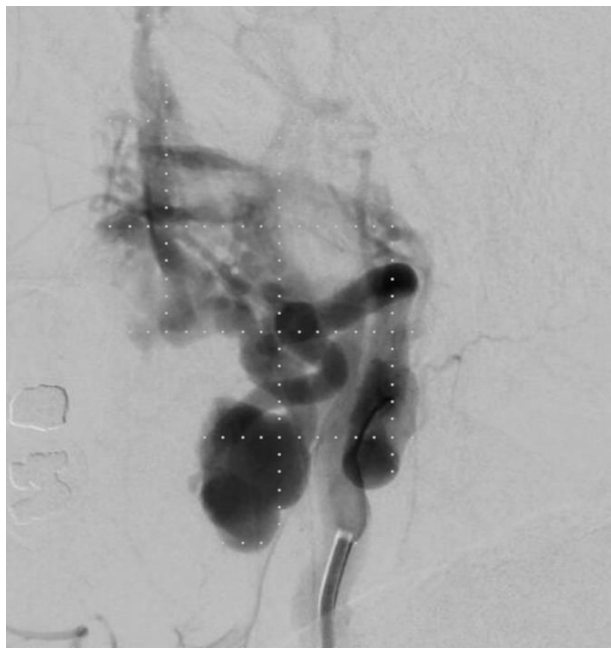


Figure 4: Angiogram status post embolization.

Outcome and Follow-up

A new panoramic image was exposed on post-operative day four revealing extensive coil placement overlying the right mandibular ramus and coronoid process.

The patient has ongoing follow-up with NIR and was subsequently referred to the head and neck surgery team for resection. The patient is also undergoing genetic testing.



Figure 5: Panoramic image of right mandibular ramus lesion post-operatively.

DISCUSSION

AVMs of the mandible are rare, high-flow vascular anomalies that present significant diagnostic and therapeutic challenges due to their risk of life-threatening hemorrhage. Standard biopsy protocols associated with cyst-like lesions assist in preventing these life-threatening events. Aspiration of these lesions is crucial to rule out an AVM prior to surgical access and biopsy. The risks associated with these lesions underscore the importance of completing a comprehensive diagnostic work-up, as AVMs may mimic odontogenic cysts or tumors and can present with non-specific dental symptoms. Additionally, even in the setting of a negative aspiration, it is important to proceed with extreme caution when an AVM is suspected. The case presented in this report demonstrates this principle well.

The appropriate timely management of these identified vascular lesions is of utmost importance. The presence of a suspected AVM often initiates multidisciplinary management for diagnosis and treatment. Upon discovery of an AVM via aspiration, the surgical team aborts surgery and refers to NIR for further evaluation via vascular studies. Definitive treatment of the lesion is to be completed following embolization if indicated. Recent literature emphasizes that endovascular techniques offer a safer and more effective treatment modality than traditional surgical resection alone, particularly in high-flow mandibular AVMs [2].

Long-term follow-up remains essential as recurrence rates are reported in up to 25% of cases within the first year due to reperfusion from collateral vessels. A multimodal treatment strategy, integrating surgical, endovascular, and medical management, provides the best chance for long-term success. The necessity of vigilance when encountering atypical radiographic findings in the mandible is emphasized, as timely recognition and multidisciplinary coordination are critical to reducing morbidity and improving prognosis. Ultimately, early recognition, adherence to aspiration protocols, and timely referral for endovascular management are essential in preventing catastrophic outcomes in mandibular AVMs [2].

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