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Massive maternal haemorrhage due to a mandibular arteriovenous malformation in a term pregnancy: A case report

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ABSTRACT

Mandibular arteriovenous malformations (AVMs) are vascular lesions that are rare in pregnancy but that can be life threatening due to the potential for massive haemorrhage when ruptured.

This report describes a 30-year-old woman who presented at 39 weeks of gestation with hypovolemic shock secondary to a massive haemorrhage from her oral cavity. After resuscitation, she underwent an emergency caesarean section indicated for fetal distress. A live infant was born who required neonatal intensive care support. An angiogram was performed post-operatively which demonstrated a mandibular AVM that was selectively embolised. The patient and her newborn ultimately recovered with no further bleeding or complications.

This report describes the fourth case of a mandibular AVM in pregnancy published in the literature, that was managed with selective embolisation. More research is required to determine the effect of pregnancy on AVM manifestation and rupture. Antenatal management should be on a case-by-case basis with multi-disciplinary team involvement.

1. Introduction

Vascular malformations occur due to the abnormal development of blood vessels and encompass a range of classifications according to the blood vessels affected and their location. Arteriovenous malformations (AVMs) are a type of benign vascular malformation that connect the arterial and venous blood supply of an area, disrupting normal blood flow. They arise from errors in embryogenesis which are present at birth and manifest with age. [1] Mandibular AVMs are rare and potentially life threatening due to the potential for rupture and massive haemorrhage. They are often misdiagnosed and can present as innocuous episodes of gingival bleeding, slow-growing expansile masses or severe haemorrhage. [2] There are some reports of mandibular AVMs occurring in pregnancy; however, it is not well understood if the physiological changes of pregnancy have an association with AVM manifestation or rupture. Here, a rare case of life-threatening haemorrhage secondary to a mandibular AVM rupture is presented and the relevant literature is reviewed.

2. Case Presentation

A South Asian woman of child-bearing age, gravida 1, para 0, presented to a tertiary centre at 39 weeks of gestation with hypovolaemic shock, secondary to massive haemorrhage from a gingival mass.

Her relevant history included three months of right-sided lower periodontal swelling and bleeding, which was initially suspected to be due to trauma from the upper molar. This was managed by a dentist who initially trimmed tooth 18, which was later extracted. The patient, however, continued to have intermittent episodes of bleeding that were controlled by compression of the area with tranexamic acid soaked gauze. On the lower molar, tooth 47 became progressively more mobile and the bleeding was suspected to be caused by a granuloma that would eventually improve; however, no imaging was conducted to investigate this. At 38 weeks of gestation, one week prior to her presentation at the tertiary centre, she was reviewed in the antenatal clinic for iron deficiency anaemia and booked to have an induction for South Asian ethnicity, according to local policy. The patient's antenatal course was otherwise unremarkable, and she had no significant medical or surgical history.

On arrival at the emergency department she had persistent pulsatile bleeding originating from the oral cavity and was estimated to have lost more than 1.7 L of blood. She was hypotensive (blood pressure 57/30 mmHg) and tachycardic (heart rate 130 bpm), with a patent airway and

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normal neurological function. Physical examination of the patient was limited due to the amount of bleeding; however, documentation by the emergency treating team suggest she presented with soft-tissue swelling and pain in the right lower jaw. The patient was promptly resuscitated with intravenous crystalloid fluid and blood products and pressure was applied to the gingival bleeding with gauze. A cardiotocograph monitor demonstrated a fetal bradycardia with reduced variability, which persisted despite stabilisation of the maternal haemodynamics.

A multi-disciplinary decision was made to transfer the patient to the operating theatre for a category 1 caesarean section delivery and management of the gingival bleeding. The patient's airway was secured with an endotracheal tube and an uncomplicated caesarean section was performed under general anaesthesia. At this time, the gingival bleeding had subsided following direct digital pressure to the area, and upon visual inspection, the ear, nose and throat (ENT) surgical team identified a diffuse non-circumscribed submucosal cluster of dilated blood vessels centred in the right retromolar trigone and extending to the gum and buccal mucosa surrounding tooth 47. There was no further active bleeding or palpable pulsality or thrill.

A live male infant was delivered and immediately intubated due to poor tone and perfusion, then transferred to the neonatal intensive care unit (NICU). Apgar scores were 3 at 1 min and 8 at 3 min of life. Cord blood gases demonstrated a significant metabolic acidosis (cord arterial pH of 6.89, lactate 13.6, base excess > -20; and venous pH of 6.95, lactate 13.9 and base excess > -20), and the infant was therapeutically cooled for management of moderate hypoxic ischaemic encephalopathy. Pathology tests at birth showed an elevated serum troponin (69 ng/L) with normal liver function, creatinine and electrolytes. Serial investigations showed no evidence of end-organ dysfunction and the infant was rewarmed on day 4 of life. For treatment of respiratory distress syndrome, the infant was mechanically ventilated until four hours of life, then weaned onto non-invasive respiratory support for a total of two days.

Following conclusion of the caesarean section, the patient was immediately transferred to the radiology department whilst remaining under a general anaesthetic. She underwent a computed tomographic (CT) carotid angiogram, which demonstrated a high-flow diffuse arteriovenous malformation (AVM) in the right masticator space and lateral pharyngeal wall with arterial supply arising from the maxillary, lingual and facial arteries (Fig. 1). The right common facial vein was depicted as prominent and communicating directly with these vessels at the level of the right alveolar ridge. Selective arterial embolisation of the internal maxillary artery branches supplying the AVM was undertaken by the interventional radiology team via injection of a trans-arterial particulate in a contrast + saline suspension, under CT imaging guidance. A post-embolisation CT angiogram showed reduced flow through the AVM with some ongoing filling from small lingual branches, which were left due to risk of ischemia of the area supplied.

The patient had a protracted recovery in the intensive care unit, where she remained intubated and sedated until day 6 postpartum due to significant airway oedema and risk of re-bleeding. Following extubation, she was weaned off dexamethasone and had no airway concerns or episodes of bleeding from the mandibular AVM. The patient was transferred to the postnatal ward on day 10 for supportive care. She underwent repeat right common and external carotid angiography on day 20 of admission, which showed that the AVM nidus in the socket of tooth 47 had thrombosed after the previous embolization, with no further filling of the nidus or draining veins.

The neonate's follow-up investigations, including magnetic resonance imaging (MRI) of the brain, showed no significant abnormality. The infant remained in the NICU and special care nursery for supportive care until day 12, then was transferred to the postnatal unit to be with the patient and establish breastfeeding. Follow-up with the neonatologist was arranged a month following discharge.

The patient recovered well from an obstetric perspective and was discharged home with her newborn on day 22 post -partum. Follow-up was arranged with the ENT specialist with a plan to undergo a repeat angiogram 6 months post-partum.

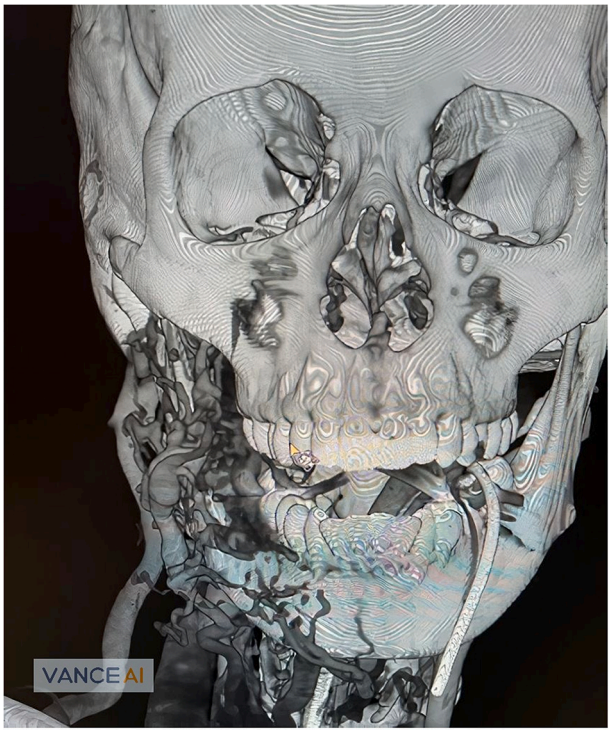

Fig. 1. Computed tomographic (CT) carotid angiogram demonstrating an arterio-venous malformation (AVM).

3. Discussion

To our knowledge there are only three previously reported cases of mandibular AVM in pregnancy. [2–4] All three cases presented with uncontrollable oral bleeding. Two cases, which presented at 23 and 35 weeks of gestation, were managed with angiography and embolisation [2,4]. In both of these cases the women had a normal vaginal delivery at 36 weeks and underwent surgical resection of the AVM post-partum. The final case was conservatively managed with local compression and transfusion, then delivery via caesarean section at 36 weeks. Post-partum, the AVM spontaneously regressed, then recurred in the subsequent pregnancy necessitating delivery via caesarean section at 34 weeks. [3]

Vascular lesions of the head and neck are extremely rare, but most frequently occur in the mandible and affect women more than men. [2] The clinical presentation of mandibular AVMs can be highly variable, leading to difficult and delayed diagnosis. Symptoms may include pain, bleeding, swelling, discolouration of the affected area or displacement of teeth [3,5] Predisposing factors for the proliferation and subsequent rupture of AVMs include local trauma and infection [2], of particular relevance in lesions of the oral cavity as in this case.

The mechanism of AVM proliferation in pregnancy is not well understood. It has been postulated that the physiological changes in the cardiovascular and endocrinological systems of pregnant women such as increased cardiac output and circulating blood volume may significantly increase the risk of vascular malformations; however, evidence is lacking. [2,3,6] Moreover, surges in endogenous estrogen, as in pregnancy, also have an effect on endothelial proliferation and function. This poses a question surrounding the patient's risk of further vascular malformation and haemorrhage in subsequent pregnancies. In the absence of no established guidelines, the risk of subsequent rupture triggered by the physiological changes of pregnancy remain unknown, but likely

warrants active surveillance and consideration to timing of delivery. Maternal hypovolemia caused by AVM rupture in the antenatal period may lead to hypoxia and subsequent acidosis of the developing foetus secondary to decreased uteroplacental blood flow, as demonstrated in this case. This is associated with potential short- and long-term morbidity for the infant, which is variable and largely unpredictable at the time of birth. [7]

Treatment of vascular malformations is dependent on the location and hemodynamic features of the lesion. Options include embolization, sclerotherapy, surgical resection or observation, each with varying risk profiles. [1] CT angiography remains the gold standard imaging modality for vascular malformations in any location and allows classification of contributory vessels and direction of flow to aid diagnosis and treatment. [5] In this case, CT angiography and subsequent selective arterial embolisation were successfully utilised. The likelihood of spontaneous regression after a period of observation in the post-partum period remains unclear, but spontaneous regression has reported in similar cases. [2,6]

This seems to be the first published case of a mandibular AVM in pregnancy presenting at a term gestation with severe haemorrhage. The initial management goal was maternal resuscitation and haemodynamic stabilisation and thereafter urgent delivery in the setting of fetal distress. In this case the decision to deliver was simplified as the pregnancy was at term and otherwise uncomplicated. In the setting of a premature gestation or maternal medical co-morbidities, an expedited delivery via caesarean section could have been more complicated. The engagement of multi-disciplinary input from the obstetric, ENT, radiology and intensive care teams ultimately led to the successful management of a high-risk obstetric presentation.

Contributors

All of the authors were involved in the work-up and immediate management of the patient presented and were responsible for the ongoing care of the patient. All authors contributed to the literature review that made the basis for the discussion, and were involved in writing the discussion and editing.

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Patient consent

Written consent obtained.

Provenance and peer review

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Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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