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Case report

A Rare Presentation of Suspected Temporal Bone Arteriovenous Malformation in Pregnancy

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Abstract

Arteriovenous malformations (AVMs) are vascular anomalies that may lead to severe complications due to increased pressure and flow in venous structures. While AVMs can manifest in diverse anatomical regions, a temporal bone AVM in pregnancy is rare. This report presents the case of a 28-year-old woman diagnosed with a temporal bone AVM during her third trimester. She was conservatively managed and experienced an uneventful delivery. This case underscores the complexities in managing AVMs in pregnancy, particularly the delicate balance of weighing maternal and foetal risk. Given the potentially catastrophic sequelae of AVM rupture during pregnancy, management must be carefully tailored, considering factors such as AVM location, size, the potential for haemorrhage, and patient comorbidities. In our case, a collaborative approach with a tertiary neurosurgical team, alongside meticulous imaging analysis, was vital in ensuring favourable maternal and fetal outcomes. This case highlights the importance of multidisciplinary collaboration, appropriate imaging utilisation, and patient involvement in decision-making.

Introduction

An arteriovenous malformation (AVM) is an anomalous vascular communication between arterial and venous systems that circumvents the normal interposing capillary bed and may lead to shunting of highpressure arterial blood towards low-pressure efferent vessels. The abnormal flow may damage the veins causing complications such as varicosity, pain, swelling and hemorrhage (1) AVMs' dimensions, clinical manifestation, and complexity vary considerably depending on the location, calibre, and complexity of the vessels involved. Head and neck AVMs represent approximately 37.3% of all AVMs, with cerebral AVMs having a prevalence of about 0.01%–0.5% in the population. (2,3)

Approach to management depends on symptoms, such as local tissue damage or dysfunction, aesthetic concern or the risk of potentially life-threatening haemorrhage if rupture occurs. Where therapeutic intervention is required, choices may include resection, endovascular embolisation, or stereotactic radiosurgery (gamma knife) (3,4).

The cardiovascular adaptations of pregnancy, such as increased blood volume, reduced peripheral resistance, and potential hypertension, present a considerable risk for individuals with pre-existing or newly diagnosed AVMs. These may lead to a heightened risk of rupture and potentially life-threatening haemorrhage (5). Although this occurrence is a rare pregnancy complication, the repercussions for both the mother and fetus can be catastrophic. In most instances, a risk–benefit analysis of intervention does not support active AVM treatment during pregnancy (6). Instead, the preferred approach is generally conservative, emphasising ongoing observation and risk management strategies

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antenatally and at birth.

This article reports the case of a 28 year old multiparous woman diagnosed with a previously unknown arteriovenous malformation in the right temporal-frontal bone with intraosseous cavitation during pregnancy. She was managed expectantly with diagnostic surveillance and went on to have an uneventful delivery.

Case

In this report, we discuss the case of a 28-year-old woman, gravidity 2, parity 2 (G2P2), who received a first diagnosis of arteriovenous malformation (AVM) in the temporal bone at 29 weeks and three days of gestation. The patient had noted discomfort with chewing in the preceding two weeks. The symptoms were right-sided, and with palpation, she discovered a small non-tender swelling on the right side of her head above the jaw. She could not recall having felt it before. She was otherwise well. Her current pregnancy was complicated by recently diagnosed gestational diabetes, which was being managed by dietary and lifestyle interventions. She was also noted to be iron deficient despite oral supplementation. There were no other antenatal concerns. She had had two successful pregnancies previously, both delivered by Caesarean section. The first was an emergency during labour for failure to progress, and the second was a booked elective procedure.

Upon examination, a non-fluctuant, non-translucent 2 cm mass was identified. There was no pulsatility or associated lymphadenopathy in the neck or axilla. An ultrasound examination revealed an ovoid lesion of 18.5mm with mixed echogenicity suggesting solid-cystic components. This lesion displayed prominent hypervascularity, consistent with vascular malformation or hemangioma. The lesion extended to the deep cutaneous tissues and was closely intimated to the bone. The possibility of an intracerebral extension or communication could not be ruled out. A non-contrast magnetic resonance (MR) imaging of the brain confirmed the presence of an arteriovenous malformation (AVM) with secondary extension into the adjacent temporalis muscle and right temporal-frontal bone. Magnetic resonance angiography (MRA) revealed afferent feeders from the external carotid artery. There was no discernible communication with cerebral vessels. There was no suggestion of a primary tumour, vascularised malignancy, or other vascular anomaly.

A collaborative decision was made with a tertiary neurosurgical team to recommend a computed tomography (CT) cerebral angiogram. The lesion was now 5 cm but remained essentially asymptomatic. Imaging was performed at 39 weeks' gestation and ruled out any intracranial involvement. The feeder was confirmed to originate from the right external carotid artery, with efferent venous drainage leading into the posterior cavernous sinus, anterior to the temporal lobe and the petrous right venous plexus. Post-procedure consultation with the neurosurgery team concluded a minimal risk for neurosurgical complications. The patient was, however, referred for an elective repeat Caesarean section at a tertiary centre where standby neurosurgery intervention was at hand should a vascular haemorrhage occur. The Caesar was performed without complications. Both mother and baby enjoyed an uncomplicated post-natal recovery. Six months later, the mother reported no change of symptoms. The lesion was still palpable but had not grown in size. Neurovascular surveillance remained expectant with ongoing imaging arranged at a further 6-month interval.

Discussion

Arteriovenous malformations (AVMs) are most frequently observed in craniofacial bones, with a predilection for teeth-bearing bones (7). They are rarely affecting the temporal bones (8). They may occasionally arise following trauma but more generally develop as congenital aberrations of localised vasculogenesis during the early embryonic period. They usually remain dormant and unnoticed during childhood and only become symptomatic during rapid growth phases elicited by puberty or pregnancy hormonal changes (9). As AVMs are benign, treatment revolves around symptom management and mitigating potential complications such as unabated growth, disruption of adjacent tissue structure, and haemorrhage secondary to vascular failure. Factors that may affect management decisions include the size and location of the AVM, particularly if involving the face or neck, and the presence of co-existing medical or congenital conditions that may predispose the patient to multiple lesions or an elevated risk of complications such as haemorrhage (10, 11).

Physiological adaptations in pregnancy, particularly those of the cardiovascular system, which include expansion of blood volume to support the increased cardiac output necessary for foetal growth, are particularly significant provocateurs (12). While instances of intracranial haemorrhage secondary to AVMs are uncommon, when they do occur, they often do so in the third trimester or at the time of delivery when changes to cardiovascular health are maximal. During labour, the risk of haemorrhage is heightened by hypertensive episodes associated with pain and the work of pushing and the episodic tidal floods of venous filling that occur with uterine contractions. Patients with underlying vascular disease, either pre-existent or acquired as comorbidities during pregnancy, such as diabetes, hypertension or pre-eclampsia, may be

particularly susceptible (2,3).

There is no definitive consensus on the effective management of pregnant women with AVMs. A review reported that AVM haemorrhage was significantly associated with poor maternal outcomes, specifically neurological disability (3). Owing to the potential risk of treatment and resultant risk–benefit analysis, routine active AVM treatment during gestation is not universally advocated (2,3 & 6). Should intervention be necessary, it is typically scheduled for no later than the second trimester. Active AVM therapeutic modalities include surgical resection, endovascular embolization, or stereotactic radiosurgery (such as gamma knife). (2,4). The most commonly employed and comparatively more accessible procedures include embolization and sclerotherapy.

Embolization entails using materials such as medical glue, metal coils, or plugs to impede blood flow within an AVM, introduced via a catheter into an artery connected to the AVM. Conversely, sclerotherapy injects obliterative agents into an AVM, instigating scar formation and destructive effects on the feeder vasculature. As these techniques do not eliminate the AVM, there remains a risk of AVM re-expansion post-treatment. Treatment options for temporal bone AVMs must be selected judiciously, with due consideration given to the potential risks to the facial nerve.

In cases of unruptured AVMs during pregnancy, conservative observation may be adopted, provided there are no precipitating factors for bleeding, such as hypertension, co-existing aneurysm, or venous ectasia. There is limited evidence addressing whether the risk of cerebral AVM haemorrhage is increased with vaginal delivery or mitigated by caesarean section. Irrespective of the delivery method, meticulous blood volume and pressure control are crucial to minimise the risk of rupture (13).

Conclusion

This report presents a rare and previously unreported example of a temporal bone arteriovenous malformation (AVM) discovered in the third trimester of pregnancy. It highlights the role of open, collaborative communication, so vital in rural healthcare settings where partnerships with tertiary healthcare services are often required for effective, and comprehensive care delivery. In all such instances, the patient must remain cantered and included as an active participant in decision-making.

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