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ABSTRACT: Patients with arteriovenous malformations (AVMs) are at risk of significant hemorrhage and AVMs are especially difficult to manage in those desiring future pregnancy. Few cases of successful deliveries have been previously reported.

We report an unusual case of AVM of the tongue in a pregnant woman who presented with massive pulsatile bleeding from a ruptured artery in the tongue in late gestation, this was thought to be caused by the changes in hormonal balance and the increase in cardiac output. The bleeding was controlled with transarterial embolization and transmucosal absolute ethanol sclerotherapy. Furthermore, her second and third deliveries were successfully managed.

We managed symptomatic tongue AVM by combining transarterial embolization and transmucosal sclerotherapy, which was followed by successful deliveries. This case supports the utility of transmucosal absolute ethanol sclerotherapy for tongue AVM and multidisciplinary medical care for a successful delivery.

Key Words: Arteriovenous malformation, tongue, embolization, sclerotherapy, delivery

Introduction

Arteriovenous malformations (AVM) are rare in the oral and maxillofacial regions, but patients are at lifetime risk of significant oral hemorrhage resulting to death.[1-3] Few cases of successful management by transmucosal sclerotherapy have been previously reported and AVMs are especially difficult to manage in patients desiring future pregnancy. We report a case of AVM of the tongue with massive bleeding from a ruptured artery in the tongue in a pregnant woman. After first cesarean section, our patient was managed by transarterial embolization and transmucosal sclerotherapy, which led to the other two successful deliveries.

BRIEF CLINICAL REPORT

A 30-year-old female patient was referred to our emergency unit of Hokkaido University Hospital (Sapporo, Japan) for management of an uncontrollable hemorrhage from her tongue that had begun 1 day previously. Even four years before, she was rushed to the emergency room of another hospital in shock because of massive hemorrhage from her tongue, and had emergency tracheostomy and embolization, then had been diagnosed with AVM of the tongue. After that, in the late stage of pregnancy with her first child, she had been managed because of pregnancy hypertension in another Department of Obstetrics and Gynecology. At 38 weeks' gestation there was massive hemorrhage from the tongue, so she was admitted to our hospital soon after delivery of her first child during an emergency cesarean section at the hospital. Her baby was born healthy; weight 3,095 grams, value of Apgar score 8/8.

Her hemoglobin concentration was 9.8 g/dL and 30.2% of hematocrit after immediate blood transfusions during emergency cesarean delivery. She presented with recurrent hemorrhage from a ruptured artery in the tongue, and had a significant risk of airflow obstruction. So, to keep the upper airways patent and confirm the bleeding point, tracheostomy was performed by otolaryngologists followed by elective angiography by radiologists to control the bleeding. Heavy bleeding from a ruptured artery and multiple coils in the proximal lesion of the right lingual artery that was treated 4 years ago were found (Fig.

1). Computerized tomography (CT) angiography revealed an AVM with its feeding arteries (the main was the right lingual artery and the others were the left lingual artery and the right facial artery) (Fig. 2). The bleeding from a ruptured artery in the tongue was managed by transarterial embolization using n-butyl cyanoacrylate (NBCA) three times but it was not enough to control the bleeding from the tongue AVM because multiple coils previously treated in the proximal lesion of the right lingual artery prevented access to the nidus.

Next, we performed absolute ethanol sclerotherapy under ultrasound (US) guidance while the blood flow was controlled by the balloon catheter. Exerting traction on the tongue by silk threads enabled application of a small sector US probe with ease (Fig. 3A). After the needle reached the nidus of the tongue, a sclerosing solution mixture of absolute ethanol with contrast material at a 4:1 ratio was slowly injected into the lesion under fluoroscopic guidance (Fig. 3B). The total volume of sclerosing solution used per treatment session was 41 mL. No complications occurred during and after treatment. After the therapy, the bleeding was controlled and the mass gradually regressed. Postoperative magnetic resonance imaging showed reduction in the size of the lesion (Fig. 4A and B).

Sixteen months later, she had an elective caesarean section at 37 weeks gestation and a second healthy infant was delivered, without bleeding of the tongue. Although there was dynamic volume change of the tongue before and after the second delivery (Fig. 5A and B) and a small hemorrhage occurred 3 months after the second delivery, it was managed by

embolization alone. Fifteen months later, she had second elective caesarean section at 37 weeks gestation and her third delivery was managed without embolization and no bleeding occurred.

DISCUSSION

Although an AVM may be present in any body tissue and over half of them occur in the head and neck,[4] they are rare in the oral and tongue regions.[5] Richter et al[6] suggested that tongue AVM can occur within a disease spectrum with different clinical presentations, radiographic findings, and histology among patients with focal versus advanced lesions. Focal lingual AVM are more often unilateral firm lesions with discrete borders within the substance of the tongue. Advanced lingual AVM involving the tongue, floor of mouth, and upper neck are diffuse and spongy with rebound and indefinable boundaries. Inadequate treatment is thought to contribute to collateral flow and disease progression in advanced AVM, making further management difficult. Advanced AVM of the tongue can arise after previous interventions that incompletely obliterated the AVM because of recalcitrant and progressive disease. In this case, we classified the case as an advanced tongue AVM because the lesion involving the tongue and floor of mouth had the abovementioned features of advanced AVM. Furthermore, prior embolization had been performed, and more importantly, bilateral vascular contributions including lingual and facial arteries were found on CT angiography and conventional angiography with diffuse lesions, as opposed to unilateral and solitary lingual artery contribution in the focal tongue AVM.

Treatment is rarely indicated for an asymptomatic AVM. Once the diagnosis is made, the patient should be closely followed up at 6-monthly or yearly intervals. Intervention should

be performed when there are signs and symptoms of pain, bleeding, ulceration, infection, or concern about endangering vital structures.[7] Ligation or proximal embolization of feeding vessels should be avoided as it will lead to rapid recruitment of flow from nearby arteries and access to embolization will be blocked.[8] Nevertheless, she was treated with proximal embolization of the lingual artery 4 years ago.

Few data are currently available regarding AVMs in pregnancy. According to Robinson and Sabiston,[9] women with AVMs in pregnancy that were most likely to bleed tended to be younger (20–25 years) and were usually primiparous, with bleeding being most common between 15 and 20 weeks of gestation. However, bleeding could occur at any stage, including during labor and in the puerperium.[9] Our patient presented with pregnancy hypertension in late gestation, and showed massive hemorrhage from AVM of the tongue. Firstly, this was thought to be caused by the 30%–40% increase in cardiac output during normal pregnancy, which is mediated by increases in both the stroke volume and heart rate.[10-12] It has been estimated that the coexistence of pregnancy and AVM may result in a 150% increase in cardiac output above normal levels.[8] Secondly, the changes in hormonal balance during pregnancy are thought to cause venodilation and progression.[3, 13] Progesterones have been related to increased venous distensibility during pregnancy and during the menstrual cycle and, this may be a reasonable hypothesis for the relationship of AVM and pregnancy.[10, 14, 15]

In addition to the maternal treatment priorities, consideration of the fetus is also necessary and cooperation between obstetricians and anesthesiologists is essential during delivery. In our case, obstetricians prepared for elective cesarean section in case of fetal distress and anesthesiologists performed combined spinal and epidural anesthesia during second and third deliveries.

There are several ways of treating AVM of the head and neck, including embolization, sclerotherapy, resection, and combination treatment.[16-19] These treatments allow clinicians to form appropriate treatment plans using a team approach. However, the evolution of the AVM becomes unpredictable during pregnancy. In fact, the effect of progesterone can lead to AVM dilatation in a short period of time, and subsequent rupture which can result in severe complications and risk the lives of the woman and her fetus.[7]

In this case, after otolaryngologists established a surgical airway, the radiologists performed arterial embolization, and the plastic surgeons performed absolute ethanol sclerotherapy under US guidance. Cooperation between otolaryngologists, radiologists, and plastic surgeons, and provision of sufficient information about the treatment strategy to the patient are essential.

As seen in our case, if the radiologists cannot perform adequate transarterial embolization because of difficulties in approaching the nidus due to past ligation and coil embolization, transmucosal sclerotherapy may make it easy to reach the distal nidus. In the

second delivery, the effectiveness of this treatment might have prevented heavy bleeding from the arteries because we could manage a small hemorrhage 3 months after the second delivery by embolization alone. No treatment was needed during the third delivery.

Focal lesions of the tongue would be a good indication for our technique. In cases where the AVM is mainly located in the floor of the mouth, a transmucosal approach by US is more difficult. In focal AVM of the tongue, compressibility and rebound are rarely present. In contrast, recurrence and complications are more often found in patients with advanced tongue lesions involving the floor of mouth.[6]

In conclusion, we achieved good maternal and fetal outcomes in our case by transarterial embolization and transmucosal sclerotherapy. This case supports the utility of this treatment option in patients desiring future pregnancy who have an AVM of the tongue, and provides additional evidence that a successful pregnancy is indeed possible with interdisciplinary medical care.

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FIGURE LEGENDS

FIGURE 1. Angiography of the right lingual artery. Note the heavy bleeding from the right lingual artery and the multiple coils used in the previous treatment.

FIGURE 2. CT angiography showing AVM in the tongue. The main feeder was the right lingual artery and the minor feeders were the left lingual artery and the right facial artery.

FIGURE 3. Intraoperative findings.

A. Traction on the tongue was exerted by silk threads.

B. The US image shows absolute ethanol sclerotherapy given through a needle using the transmucosal approach.

FIGURE 4. Sagittal T1-weighted MR image obtained before and after transmucosal sclerotherapy showing a reduction of the lesion with no flow in the posterior part of the tongue.

A. Before transmucosal sclerotherapy.

B. After transmucosal sclerotherapy.

FIGURE 5. Figures showing the dynamic volume change of the tongue AVM before and after the second delivery. Note that the mass of the tongue markedly regressed after the second successful delivery.

A. The day before the second caesarean section.

B. One month after the second delivery.



FIGURE 1.



FIGURE 2.

A



B

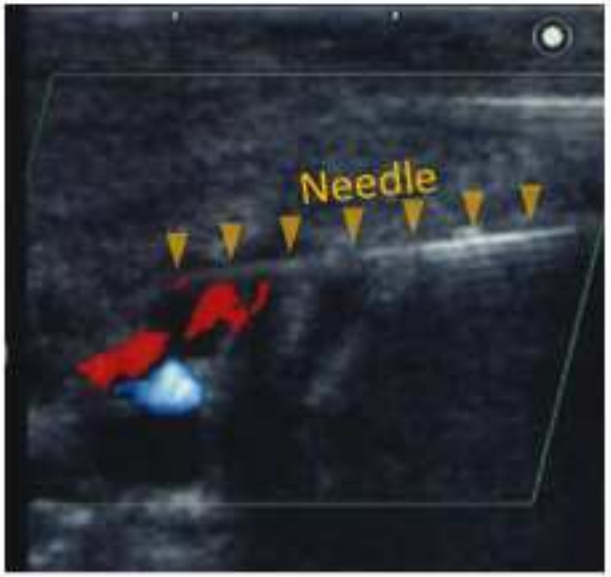


FIGURE 3.

A

B

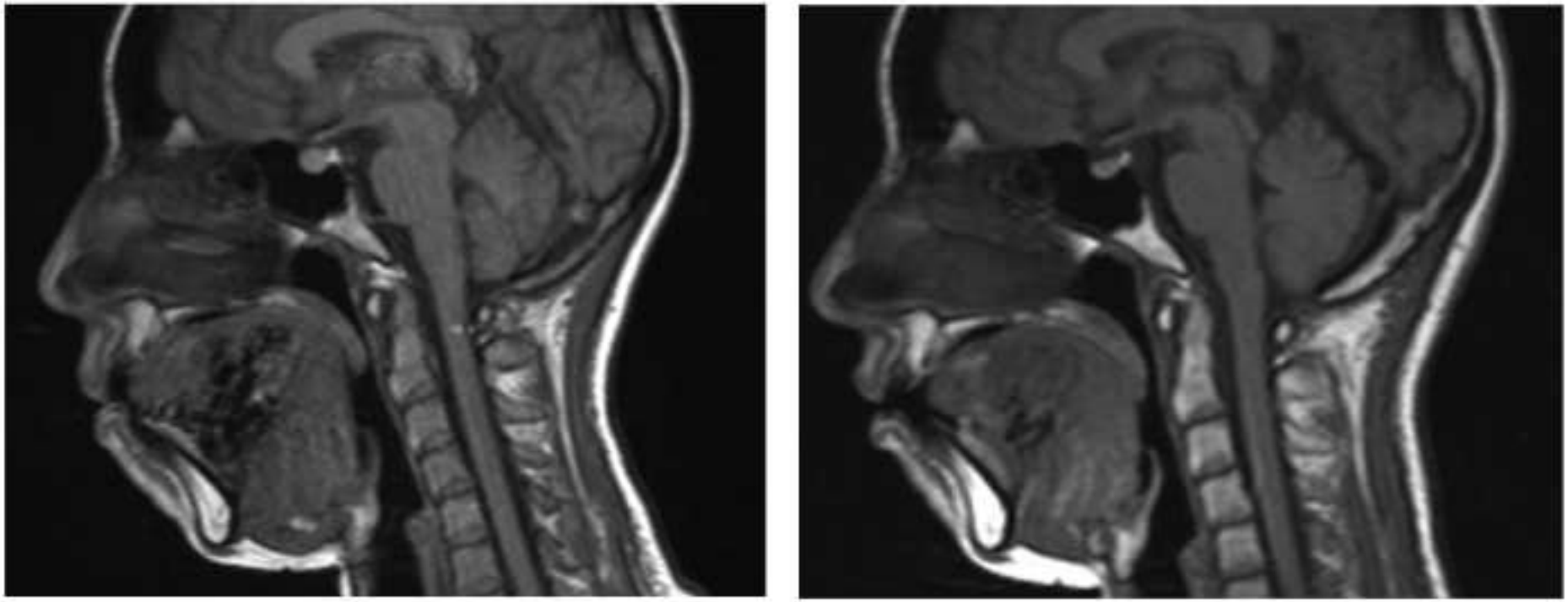


FIGURE 4.

A



B



FIGURE 5.