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Spontaneous Regression of a Spinal Extradural Arteriovenous Fistula After Delivery by Cesarean Section

—Case Report—

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Abstract

A 23-year-old Japanese woman presented with a newly developed spinal extradural arteriovenous fistula (AVF) during pregnancy. She had been followed up for a suspected spinal cavernous angioma and became unable to walk during the 29th week of her pregnancy. Magnetic resonance (MR) imaging showed a spinal extradural AVF at the T3 to T4 levels compressing the spinal cord. After delivery by cesarean section, her neurological symptoms gradually began to resolve, and she was able to resume walking without assistance. MR imaging confirmed spontaneous regression of the AVF. This case suggests that exacerbated neurological symptoms and AVF growth triggered by pregnancy can improve after delivery without interventional treatment. Careful follow up of neurological findings is required to prevent unnecessary interventional procedures in pregnant women with spinal AVF.

Key words: spinal extradural arteriovenous fistula, pregnancy, cesarean section, contrast-enhanced magnetic resonance angiography, cavernous angioma

Introduction

Complications caused by vascular anomalies of the central nervous system (CNS) are not uncommon during pregnancy, because the circulating blood volume and central venous pressure are increased and the blood coagulation system is activated.[4–6] Therefore, the progression of CNS vascular anomalies may be fatal in pregnant women. Various strategies to treat pregnant women with CNS vascular anomalies have been reported.[1,6,7] However, no correlation between pregnancy and intracranial arteriovenous malformation (AVM) rupture has been documented,[4] so pregnant women with unruptured intracranial AVMs can be expected to carry to term normally. Two women with spinal AVMs presented with exacerbated neurological symptoms of leg weakness during pregnancy.[8] Subarachnoid hemorrhage (SAH) occurred during follow up in both patients, and surgical intervention was required in one. The management of AVMs or unruptured intracranial aneurysms during pregnancy has been documented,[2,4,9] but the optimal management of spinal vascular anomalies complicating pregnancy remains to be determined.

We treated a pregnant woman who presented with a symptomatic spinal extradural arteriovenous fistula (AVF) extending from the spinal canal to the extracanal paravertebral space, which regressed spontaneously without residual neurological deficits after delivery by cesarean section.

Case Report

A 23-year-old Japanese woman with an intramedullary spinal tumor, suspected to be a cavernous angioma, had been followed at our institution since the age of 10 years. The tumor was located at the T2 to

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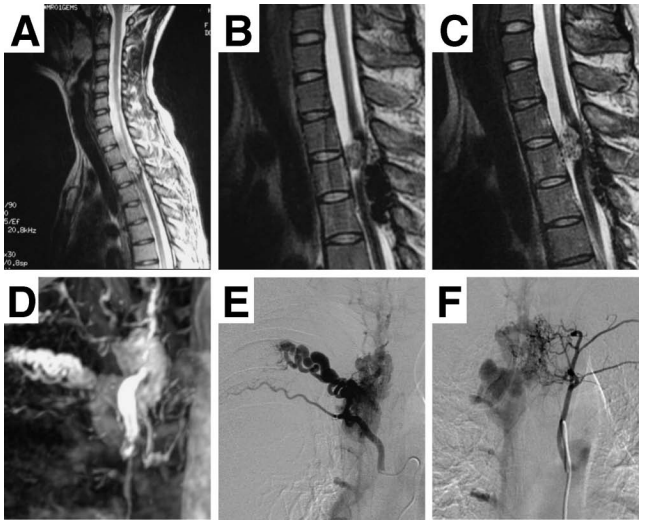

Fig. 1 A–C: Sagittal T_2-weighted magnetic resonance (MR) images showing a spinal tumor 4 years before pregnancy (A), and a hypointense mass lesion which emerged to the right and caudal to the spinal tumor during pregnancy (B), which had spontaneously disappeared by the 6th post-partum month (C). D: Coronal MR angiogram showing a varix-like dilated vascular system 2 days post-partum. E, F: Right (E) and left (F) spinal angiograms (anteroposterior view) revealing an arteriovenous fistula fed by the bilateral 6th intercostal arteries 1 week post-partum.

T3 levels and appeared as heterogeneous intensity on both T_1- and T_2-weighted magnetic resonance (MR) imaging (Fig. 1A). She had been presenting only deep sensory disturbance with no motor deficits. She became unable to walk during the 29th week of pregnancy. She presented with incomplete superficial and deep sensory disturbance below the T2 dermatome. She also had severe motor weakness in both legs. She became unable to stand during the 33rd week of pregnancy, so was admitted to our institution for high-risk delivery. T_1- and T_2-weighted MR imaging of the spinal cord showed a hypointense mass lesion at the dorsal surface of the spinal cord just caudal to the spinal tumor, which was not detected 4 years earlier, suggesting that the newly developed neurological symptoms were attributable to this lesion (Fig. 1B). Her neurological symptoms continued to worsen, so cesarean section was successfully performed at 34 weeks of pregnancy.

MR angiography with contrast medium was then performed to differentiate between CNS vascular anomaly and hemorrhagic lesion, which showed pooling of the contrast medium at the level of the mass lesion, and several abnormally dilated vascular systems both within and outside the spinal canal (Fig. 1D). These findings were indicative of extradural AVF. Spinal angiography identified a large extradural AVF within and outside the spinal canal, and three-dimensional computed tomography revealed that the 2 components of the AVF were connected through the T6 and T7 intervertebral foramina. The AVF was mainly fed by the right 6th intercostal artery (Fig. 1E), as well as the left 6th intercostal artery (Fig. 1F). An anonymous dilated vein within the thoracic wall formed a varix connecting to the extracanal portion of the main AVF. Detailed microcatheter study was not performed, so we were unable to identify the primary shunting point.

Endovascular embolization of the spinal AVF was planned. However, her neurological symptoms gradually improved after delivery, so she was placed under conservative observation. Concurrent with the improvement of her neurological symptoms, MR imaging showed that the main AVF spontaneously regressed. Follow-up MR imaging showed release of the spinal cord compression (Fig. 1C). Six months after delivery, her neurological symptoms had abated and she was able to walk without assistance.

Discussion

Our present patient with a pre-existing intramedullary spinal lesion, suspected to be a cavernous angioma, developed a symptomatic spinal extradural AVF during pregnancy. The anatomical proximity suggests some connection, but any relationship between the spinal angioma and AVF remains unclear. The possible involvement of pregnancy in the development and progression of AVF is also unclear. MR imaging performed 4 years earlier showed no evidence of the AVF, and the lesion regressed spontaneously after delivery, suggesting that pregnancy was important in the development and subsequent regression of her spinal extradural AVF. As her neurological symptoms started to improve within the 1st week after delivery, sudden post-partum changes in the circulatory system such as decreases in central venous pressure and total blood volume may be contributory factors. A relationship between pregnancy or menstruation and the symptoms of spinal AVM has been reported,[3,5,8] suggesting that changes in the concentration of circulating estrogen and progesterone may be key factors.

The present case suggests that, in the absence of hemorrhagic insult to the spinal cord, patients with exacerbated neurological symptoms attributable to expansion of spinal AVM or AVF can be treated conservatively with observation, so avoiding un-

necessary interventional treatment during pregnancy. However, we recommend detailed post-partum neurological studies because some patients may have high risk for AVM or AVF rupture and SAH, as suggested by the previous two cases.[8]

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