Rupture of an intracranial arteriovenous malformation (AVM) in pregnancy: case report

R. RISPOLI1, L. DONATI2, G. GIOVANNINI3, G. ZOFREA1, A. DI CHIRICO1, N. BARTOLINI4, G.P. PASSALACQUA2, S. CARLETTI1


The rupture of an intracranial arteriovenous malformation (AVM) in pregnancy is a rare occurrence, but may have fatal consequences. A link between AVM rupture and pregnancy has been proposed; it may be caused by the increased cardiac output or circulatory effects of the elevated estrogen levels. Here, we report the case of a 27-week pregnant woman who presented with a symptomatic ruptured cerebral AVM, treated by cesarean section followed surgical resection.

We achieved good maternal and fetal outcomes in our case. Surgical intervention for ruptured AVM during pregnancy could prevent re-bleeding, and allow for determination of the delivery method based on the obstetrical indications. Cooperation between neurosurgeons, obstetricians, and anesthesiologists and sufficient information about the treatment strategy given to the patients are essential.

KEY WORDS: Arteriovenous malformation - Pregnancy - Cerebral hemorrhage - Cerebral angiography.

Malformazione arterovenosa - Gravidanza - Emorragia cerebrale - Angiografia cerebrale.

Introduction

The rupture of an intracranial arteriovenous malformation (AVM) in pregnancy is a rare occurrence, but may have fatal consequences (1). A link between AVM rupture and pregnancy has been proposed; it may be caused by the increased cardiac output or circulatory effects of the elevated estrogen levels (2). However, in some series the reported hemorrhage rate from AVMs in pregnancy is around 0.6% to 3.5%, which is similar to the 2% to 4% rate in non-pregnant women. Due to this low risk of hemorrhage, most Authors recommend conservative management of unruptured AVMs during pregnancy (2, 3). An aggressive approach is warranted, however, when pregnant patients present with a ruptured AVM.
The risk of re-bleed during the same pregnancy (27% to 30%) is greater than the risk of re-bleed in non-gravid women within one year of their initial bleed (6%). These bleeds in pregnant women are associated with high maternal and fetal mortality (10% to 40%) (2, 4). Thus, despite the potentially deleterious effects of radiation on the fetus, surgical management is generally indicated when pregnant women present with a ruptured AVM. Historically, ruptured AVMs in pregnant women have been managed surgically with great success (2, 3, 5). In patients presenting with AVM rupture, resection has been shown to be associated with lower rates of maternal and fetal mortality when compared to conservative management (4, 6). Here, we report the case of a 27-week-pregnant woman who presented with a symptomatic ruptured cerebral AVM, treated by cesarean section followed surgical resection.

Case report

A 23-year-old woman, 27 weeks into her first pregnancy, presented to an outside hospital following the sudden onset of severe headache associated with nausea and vomiting. She denied weakness or numbness, but reported occasional double vision. Her past medical history was significant for a cerebral arteriovenous malformation partially embolized. Our patient’s only physical exam finding was a left hemiparesis. Non-contrasted computed tomography of her head revealed an acute right fronto-temporal hemorrhage with mild mass effect (Figure 1). Magnetic resonance imaging (MRI) revealed a 5 Â x 2.6 cm hematoma in the right fronto-temporal region abutting the ventricle with flow voids, characteristic of an AVM; mass effect was noted with midline shift. Digital subtraction cerebral angiography demonstrated that the nidus was being supplied by the posterior branches of her right middle cerebral, right anterior and posterior cerebral arteries (Figure 2, A and B). The majority of the venous drainage was via her superior sagittal sinus. No intra-nidal or flow-related aneurysms were noted. The weight of the fetus was estimated to be 1756g, and we were expected premature. Discussions with obstetricians, neurosurgeons and neonatologists, encouraged us to schedule cesarean section followed by craniotomy under general anesthesia. Obstetricians and gynecologists performed Cesarean section immediately followed by the removal of an intra cranial hematoma under general anesthesia. A 1.9kg girl was delivered after 10 minutes of induction. Baby was intubated and shifted to Neonatal Intensive Care Unit. Apgar scores was 2 at 1 minute and 9 at 5 minutes. The craniotomy was centered over the right fronto-temporal AVM and using the standard microsurgical technique we proceeded to safely and effectively resect the AVM, the wound was closed in the standard fashion and our patient was transferred to the neurosurgical intensive care unit.

One day after treatment we performed a cerebral CT that revealed the good evacuation of the hemorrhage.

Two days after surgery we performed a cerebral angiogram (Figure 3, A and B). The angiogram revealed complete resection of the AVM.

Our patient recovered well with and was discharged home on postoperative day two.

Discussion

The prevalence of cerebral AVMs is estimated at 0.01-0.50% of the population. AVM is generally present in patients aged between 20 and 40 years, and is more common in those over 30 years, the childbearing age for women.

The natural history of AVMs is poorly understood, and even less understood in pregnant patients, because the frequency is rare and changes in the maternal body are complicated during pregnancy. No definitive guidelines for the treatment of AVMs during pregnancy exist and the management of cerebrovascular dis-
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Figure 2 A, B - Digital subtraction cerebral angiography demonstrated that the nidus was being supplied by the posterior branches of her right middle cerebral, right anterior and posterior cerebral arteries.

Figure 3 A, B - Two days after surgery we performed a cerebral angiogram (the angiogram revealed complete resection of the AVM).
ease in pregnancy is under discussion.

Cerebral arteriovenous malformations (AVMs) may affect the prognosis for both mother and fetus because they may result in fatal intracranial bleeding during pregnancy.

Given the increased risk of re-hemorrhage in pregnant mothers that present with AVM bleed and the high associated maternal and fetal mortality (2, 6, 7), aggressive management of these lesions during pregnancy is warranted. These patients should be treated similarly to their non-gravid counterparts. There are also several additional risk factors that increase the likelihood of AVM hemorrhage and may be considered when indications for surgery are not clear. These include hypertension, increased age, coagulopathy, disseminated intravascular coagulation (DIC) and recent use of vasoactive substances. Of these risk factors, our patient had untreated gestational hypertension, which is the single factor most closely linked with AVM rupture. Maternal management of patients with ruptured AVMs should be based mainly on neurosurgical indications rather than on obstetrical indications. When neurological deterioration occurs due to AVM rupture, emergency surgery is necessary. If the fetus is sufficiently mature, simultaneous cesarean section is possible. When there is no indication for emergency surgery for AVM, blood pressure management is important. However, this is not necessarily effective for the prevention of rebleeding because patients with ruptured AVM do not always have a history of hypertension. Although radical treatment tended to be performed after delivery in many case reports and case series, some Authors suggested that early surgical intervention for AVM before delivery led to improved maternal and fetal prognosis.

Surgery for AVM is determined primarily by the Spetzler-Martin grading Scale (8). A potential complication of surgery for AVM during pregnancy is the risk of intraoperative bleeding leading to deterioration of the uterine and placental circulation. Although preoperative embolization is possible for cases with a high risk of intraoperative bleeding, such as deep-seated AVMs, the endovascular treatment itself carries the risk of ischemic and hemorrhagic complications. In addition, there is not enough evidence to presume the safety of iodinated contrast agents which cross the human placenta and enter the fetus. The potential radiation risk and the potential added risks of contrast medium should be considered in the preoperative study. Previous reports of endovascular treatment for AVM during pregnancy are limited (9, 10). There would be wider surgical indications by discussing the efficacy and risk more about endovascular treatment for AVMs during pregnancy.

Radical treatment for ruptured AVMs in patients with a mature fetus tends to be performed in the early postpartum period. It is desirable for patients with unruptured AVMs to undergo radical treatment before pregnancy due to the increasing risk of AVM rupture during pregnancy.

Prior to pregnancy, multimodal therapies such as direct surgery, endovascular embolization, and radiosurgery can be performed. In patients with unruptured AVMs diagnosed during pregnancy, conservative treatment is performed based on the risk of surgical treatment.

Conclusions

We achieved good maternal and fetal outcomes in our case. Surgical intervention for ruptured AVM during pregnancy could prevent rebleeding, and allow for determination of the delivery method based on the obstetrical indications. Cooperation between neurosurgeons, obstetricians, and anesthesiologists and sufficient information about the treatment strategy given to the patients are essential.

References

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