

# Arteriovenous malformation in uterine cervix during pregnancy

Sung-Mee Kim, Won Kyu Jang, Joon-Cheol Park, Jeong-Ho Rhee, Jong-In Kim, Jin-Gon Bae

Department of Obstetrics and Gynecology, Keimyung University School of Medicine, Daegu, Korea

As the development of Doppler ultrasonography, many cases of uterine arteriovenous malformation (AVM) have been diagnosed. But there is no case of cervical AVM in pregnant uterus. We present a 33-year-old pregnant woman who was diagnosed with AVM of the uterine cervix during the midtrimester. Color Doppler sonography and magnetic resonance image were used for diagnosis. We performed Cesarean section because of the risk of massive bleeding from the cervical AVM at 34 weeks' gestation. This is the first case of cervical AVM during pregnancy with a successful outcome and an uneventful postpartum course.

**Keywords:** Arteriovenous malformation; Pregnancy; Uterine cervix

## Introduction

Arteriovenous malformation (AVM) is an abnormal connection between an artery and a vein that bypasses the capillary system. This malformation is presented as an asymptomatic condition in many cases, but it can also cause massive bleeding and/or severe pain. Compared with AVM, varix is a venous engorgement without arterial pulsation on Doppler ultrasonography. Six cases of uterine cervical varix during pregnancy have been reported [1], but there was no report in Korea. Furthermore, AVM formed within the uterine cervix is extremely rare, to our knowledge, there is no case about cervical AVM arising in a pregnant woman, which can lead to life-threatening obstetrical hemorrhage.

We describe herein a case of AVM of the uterine cervix diagnosed during pregnancy, which was detected after vaginal spotting at 32th gestational week.

## Case report

A 33-year-old pregnant woman, gravida 1 para 0, presented with vaginal spotting on her first visit to our hospital at the 32th gestational week. Cervical AVM had not been previously identified. Her past medical history was unremarkable, except that she was a hepatitis B carrier. She has no pathological history on the uterine cervix. Obstetric sonog-

raphy revealed a single active fetus in the cephalic position. The structural survey remained unremarkable, except an upper-normal amount of amniotic fluid (amniotic fluid index was 23 cm). The fetus was average in size (estimated body weight was 2,000 g). Ultrasonography revealed a 4.4 × 2.28 cm sized hypoechoic mass like lesion in the cervix and a suspected severe engorged vessel (Fig. 1A). Placenta previa was not suspected. For the differential diagnosis of cervical varix and cervical AVM, Doppler velocimetry was performed. Color Doppler analysis demonstrated expanding of numerous vessels and rapid and turbulent blood flow in the cervix with a mosaic pattern. Pulsed Doppler evaluation revealed pulsed wave with high-velocity flow during both systole and diastole with the maximum systolic and diastolic velocity of 39 cm/sec

Received: 2013.5.28. Revised: 2013.8.18. Accepted: 2013.9.10.

Corresponding author: Jin-Gon Bae

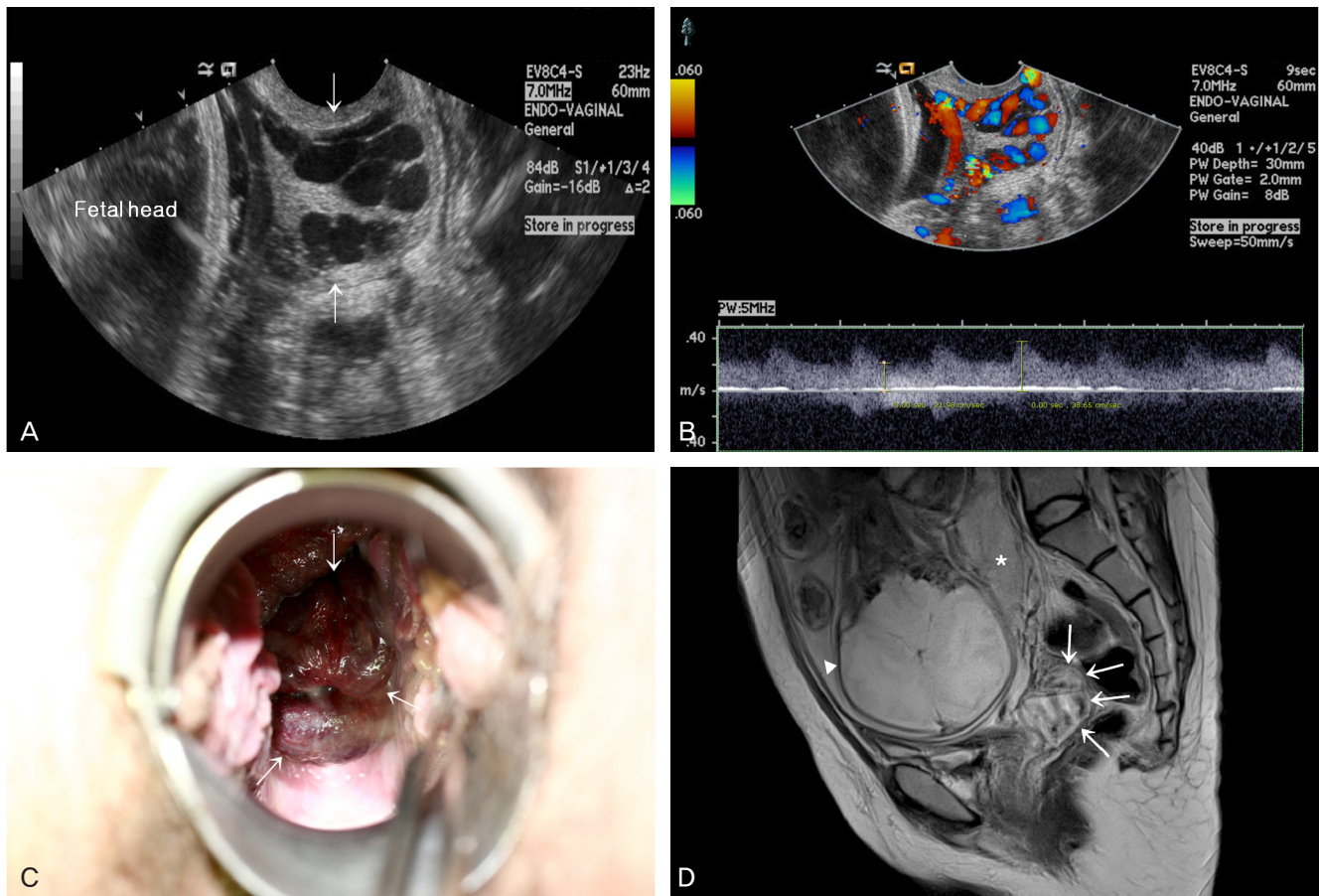
Department of Obstetrics and Gynecology, Keimyung University Dongsan Medical Center, 56 Dalseong-ro, Jung-gu, Daegu 700-712, Korea

Tel: +82-53-250-7599 Fax: +82-53-250-7599

E-mail: gonmd@dsmc.or.kr

Articles published in *Obstet Gynecol Sci* are open-access, distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0/>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

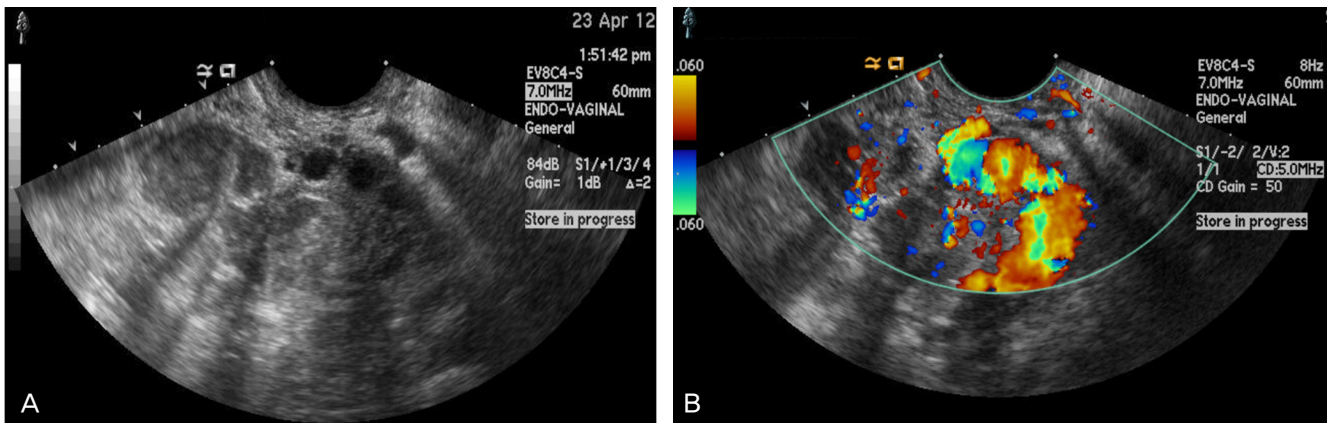
Copyright © 2014 Korean Society of Obstetrics and Gynecology



**Fig. 1.** (A) Transvaginal ultrasonography revealed multiple sized hypoechoic lesions in the whole cervix suspicious for a dilated vessel (arrows). (B) Color Doppler analysis demonstrated expansion of numerous vessels and rapid and turbulent blood flow in the cervix with a mosaic pattern. Pulsed Doppler revealed pulsed wave with high-velocity flow during both systole and diastole with the maximum systolic and diastolic velocity of 39 and 22 cm/sec. (C) Pelvic examination revealed a severe engorged vessel (arrows) on the surface of cervix, but no bleeding was noted. (D) Pelvic magnetic resonance imaging showed a 20 × 25 mm sized T2 high signal intensity lesion in the endocervix, indicating an engorged vessel on the cervix (arrows). Placenta (\*). Fetal head (arrow head).

and 22 cm/sec. Both the arterial and venous waveforms were mixed (Fig. 1B). A presumptive diagnosis of uterine cervix AVM was made. The non-stress test showed a reactive pattern of fetal heart tone, but regular uterine contraction was noted. Subsequent speculum pelvic examination revealed an engorged vessel on the surface of the cervix which was suspicious for vessel disorder, but no bleeding was noted (Fig. 1C). She was hospitalized for management of preterm labor and further evaluations of the engorged vessel on the cervix. After tocolytics, pain was subside. Pelvic magnetic resonance imaging (MRI) revealed about a 20 × 25 mm sized T2 high signal intensity lesion in the endocervix. This indicated the presence of an engorged vessel on the cervix; the possibilities of cervical AVM were considered (Fig. 1D).

At 32 weeks and 6 days' gestation, active bleeding with a large amount of blood clot from AVM was detected by speculum pelvic examination. Hemoglobin measurement was dropped from 10.9 to 9.7 g/dL even after two units of packed red blood cells transfusion. Transvaginal sonography was performed, and there was no evidence of placental bleeding, for example, placenta abruption or placenta previa. To gain time for preparing emergency cesarean section, we performed clamping with ring forceps and ligation of bleeding points. Fetal cardiocotography showed reactive pattern of heart rate without any deceleration. After 20 minutes or so, there was no bleeding and we decided to maintain the pregnancy. At 34 weeks and 2 days' gestation, we performed emergent cesarean section under general anesthesia because of massive



**Fig. 2.** (A) Postpartum transvaginal ultrasonography revealed remaining multiple arteriovenous malformation in the lower cervix. (B) Postpartum color Doppler sonography showed a tortuous shape with mosaic pattern and a mixed wave form of arteries and veins.

uterine bleeding which was uncontrolled with vaginal packing tamponade. A male infant weighing 2,700 g was delivered without difficulty; Apgar scores were 4 and 6 at 1 and 5 minutes, respectively.

During the operation, we noticed a severe engorged vessel suspected as the venous sinus near to endocervix. Except that site, we could not find AVM or venous sinus on the whole uterine corpus. Massive active bleeding from the suspected uterine cervical AVM occurred. Direct suture ligation of the cervical AVM was performed inside of the uterus with ligations of both uterine arteries, and they were successful in arresting the bleeding. The operation was completed uneventfully, and the estimated blood loss during surgery was about 2,500 mL. We could maintain the hemoglobin level of 10.7 g/dL by transfusion with 7 units of packed red blood cells and 7 units of fresh frozen plasma. She was discharged in stable condition without vaginal bleeding associated uterine cervical AVM at postoperative day 7. Four weeks later, transvaginal sonography revealed remaining multiple AVM in the lower cervix (Peak systolic velocity 34 mm/sec) (Fig. 2). Eight weeks from delivery, AVM of the uterine cervix decreased dramatically and no visible vessel engorgement of the cervix was noted by speculum and transvaginal sonography.

## Discussion

Uterine cervical AVM is an extremely rare condition and a potentially life-threatening disease, which can cause massive bleeding. As Doppler ultrasonography developed, AVM of uterus could be diagnosed easily than the old days. As a

result, AVM in uterine corpus is not a rare case in now. However AVM of uterine cervix is extremely rare, especially there is no case during pregnancy, to our knowledge. Uterine AVM is classified as congenital or acquired lesion [2]. Acquired uterine AVM is associated with conditions that damage uterine tissue, like cesarean section and curettage procedure. Other causes of uterine AVM include infection, remnant products after conception, trophoblastic diseases like H-mole and other gynecologic malignancies [3-6]. In our case, she had no history of abdominal or pelvic surgery including curettage. The most common presenting symptom is vaginal bleeding and/or menometrorrhagia. The bleeding pattern is usually intermittent and profuse, sometimes, the patient is hemodynamically unstable and needs massive blood transfusion. In the presenting patient, intermittent vaginal bleeding in the third trimester of pregnancy was the chief complaint. Many swollen vessels, feeding arteries and drainage veins were detected by inspection, color and Doppler sonography and MRI. The patient was diagnosed with uterine cervical AVM.

In the case of vaginal bleeding during the second to third trimester, placental disorders like placenta previa, vasa previa and placenta abruption should be considered first. Ultrasonography and non-stress test are useful diagnostic tools in the differential diagnosis of these conditions. Ultrasonography shows the condition of the placenta, amniotic fluid, the well-being of the fetus and abnormal vessel like AVM or varix. In the case of cervical varix or AVM, tortuous engorged vessels in the cervix can be noted by inspection with a speculum. It is difficult to distinguish them by inspection. In those cases, on gray scale ultrasonography, multiple anechoic tortuous lesion like masses can be found in the cervix. Color Doppler ultraso-

nography is valuable for the diagnosis of many uterine vascular diseases including AVM, because it is non-invasive, highly sensitive and accurate [7]. On color Doppler sonography, there is a mosaic pattern which represents increased vascularity with expansion of numerous vessels, rapid and turbulent blood flow in cervical AVM [7-9].

However, the condition is distinguished by pulsed wave Doppler, exactly. In cervical varix, there is no pulsating wave. In cervical AVM, on the other hand, pulsed wave is noted with increased flow velocity during systole and diastole with low resistance index (between 0.25 and 0.55). Mixed arterial and venous waveforms are also noted [8-10]. In our case, color and wave Doppler sonography showed a tortuous shape with a mosaic pattern and a mixed wave form of arteries and veins. It revealed high-velocity, but a slightly increased resistant index (0.65–0.79). In principle, definite diagnosis of cervical AVM is established by angiography. Angiography can be used as a treatment too as well as for diagnosis [11]. It confirms the status of blood supply like feeding arteries and drainage veins, and it is used to guide angioembolization for the treatment of AVM. However, pregnant women hesitate to undergo angiography due to the radiation, dye effect on the fetus and its invasiveness. For such reasons, angiography was not performed in the present case. Computed tomography and MRI are excellent diagnostic tools along with angiography in determining the size, extent, and vascularity of AVM and defining the involvement of adjacent organs [12]. Compared with angiography, MRI is non-invasive, thus it is considered a useful diagnostic method in pregnant women [13].

After the diagnosis of AVM, hemodynamic stabilization of the patient is the first consideration. Appropriate intravenous hydration, massive transfusion and vaginal tamponade for the control of acute vaginal bleeding should be considered. For the treatment of AVM, options are operation like hysterectomy and uterine artery embolization (UAE). Surgical management of AVM is the ligation of feeding arteries such a both uterine arteries to decrease the blood supply to the drainage veins. In the presenting case, we clamped with ring forceps and ligated the bleeding points on cervix to gain time for cesarean section, when vaginal bleeding was occurred at 33 weeks' gestation. In some cases, hysterectomy is performed for its advantage of definitive treatment to eliminate bleeding. However, surgical treatment of AVM contains several risks, like massive intraoperative hemorrhage, incomplete removal of the AMV nidus, surrounding organ injury, and high recurrence rate [14].

By these reasons, endovascular intervention with various embolic and sclerosing materials like gelform is preferred rather than hysterectomy, especially in younger women who want to save her fertility. UAE has been successful in the control of pelvic hemorrhage in a range of conditions including uterine AVM [15]. Therefore, UAE by itself or UAE with surgical resection of AVM can be an appropriate therapeutic option of uterine AVM [14,15]. In the presenting case, the patient was in singleton pregnancy, at a young age and wanted more babies if possible. We performed cesarean section because of the risk of massive bleeding from cervical AVM, and direct ligation of the feeding arteries located in the uterine cervix and the both uterine arteries. After ligation of the vessels, bleeding was controlled successfully, and we could avoid the cesarean hysterectomy. At 8 weeks after delivery, AVM of the uterine cervix decreased dramatically and disappeared engorged vessel of cervix, we believed that cervical AVM might be occurred due to hyperdynamic status during pregnancy.

In summary, uterine cervical AVM is a very rare condition, and this is the first report of cervical AVM in a pregnant woman. Ultrasonography with gray-scale and color Doppler could be helpful to differentiate from other causes of vaginal bleeding during pregnancy. After excluding placental disorder such as placenta previa or abruption, cervical varix or AVM should be considered. We present a very rare case of cervical AVM with pregnancy, which had a successful outcome and an uneventful postpartum course.

## Conflict of interest

No potential conflict of interest relevant to this article was reported.

## References

1. Sukur YE, Yalcin I, Kahraman K, Soylemez F. Cervical varix complicating marginal placenta previa: a unique coexistence. *J Obstet Gynaecol Res* 2011;37:1515-7.
2. Vogelzang RL, Nemcek AA Jr, Skrtic Z, Gorrell J, Lurain JR. Uterine arteriovenous malformations: primary treatment with therapeutic embolization. *J Vasc Interv Radiol* 1991;2:517-22.
3. Delaloye JF, Maillard C, Laurini R, Hack I, De Grandi



- P. Acquired uterine arteriovenous fistulas after choriocarcinoma. A case report. *Eur J Gynaecol Oncol* 1998;19:453-4.
4. Schiller VL, Raft E, Linden R. Uterine arteriovenous malformation. *AJR Am J Roentgenol* 1998;170:219-20.
  5. Elia G, Counsell C, Singer SJ. Uterine artery malformation as a hidden cause of severe uterine bleeding. A case report. *J Reprod Med* 2001;46:398-400.
  6. Wiebe ER, Switzer P. Arteriovenous malformations of the uterus associated with medical abortion. *Int J Gynaecol Obstet* 2000;71:155-8.
  7. Polat P, Suma S, Kantarcy M, Alper F, Levent A. Color Doppler US in the evaluation of uterine vascular abnormalities. *Radiographics* 2002;22:47-53.
  8. Huang MW, Muradali D, Thurston WA, Burns PN, Wilson SR. Uterine arteriovenous malformations: gray-scale and Doppler US features with MR imaging correlation. *Radiology* 1998;206:115-23.
  9. Mungen E, Yergok YZ, Ertekin AA, Ergur AR, Ucmakli E, Aytaclar S. Color Doppler sonographic features of uterine arteriovenous malformations: report of two cases. *Ultrasound Obstet Gynecol* 1997;10:215-9.
  10. Timmerman D, Wauters J, Van Calenbergh S, Van Schoubroeck D, Maleux G, Van Den Bosch T, et al. Color Doppler imaging is a valuable tool for the diagnosis and management of uterine vascular malformations. *Ultrasound Obstet Gynecol* 2003;21:570-7.
  11. Badawy SZ, Etman A, Singh M, Murphy K, Mayelli T, Philadelphia M. Uterine artery embolization: the role in obstetrics and gynecology. *Clin Imaging* 2001;25:288-95.
  12. Gulati MS, Paul SB, Batra A, Sarma D, Dadhwal V, Nath J. Uterine arteriovenous malformations: the role of intravenous 'dual-phase' CT angiography. *Clin Imaging* 2000;24:10-4.
  13. Nicholson AA, Turnbull LW, Coady AM, Guthrie K. Diagnosis and management of uterine arterio-venous malformations. *Clin Radiol* 1999;54:265-9.
  14. Do YS, Kim YW, Park KB, Kim DI, Park HS, Cho SK, et al. Endovascular treatment combined with embolosclectomy for pelvic arteriovenous malformations. *J Vasc Surg* 2012;55:465-71.
  15. Vedantham S, Goodwin SC, McLucas B, Mohr G. Uterine artery embolization: an underused method of controlling pelvic hemorrhage. *Am J Obstet Gynecol* 1997;176:938-48.