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## CASE REPORT

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# Congenital Arteriovenous Malformations of The Uterus

Vorapong Phupong MD,  
Anek Aribarg MBBS, FRCOG, DCH.

*Department of Obstetrics and Gynecology, Faculty of Medicine, Chulalongkorn University, Rama IV Road, Pathumwan, Bangkok 10330.*

Arteriovenous malformation (AVM) of the uterus is a rare gynaecological disorder.<sup>(1-5)</sup> There have been less than 100 cases reported in the literatures. It may be either congenital or acquired disorder.<sup>(1,2,6,7)</sup> The most common presentation is vaginal bleeding.<sup>(1,2,4-6,8)</sup> Common ages were between 20-40 years old,<sup>(4)</sup> but the youngest case was a stillborn with 34 weeks of gestation.<sup>(9)</sup> Herein we report a congenital arteriovenous malformation in a seven month old infant presenting with severe vaginal bleeding. The case was seen and treated in the year 1989, when there was less advanced equipment comparing to current medical practice.

### Case report

A seven month old female infant was admitted to King Chulalongkorn Memorial Hospital in 1989 because of severe vaginal bleeding which was aggravated by crying. Her past history and family history were unremarkable. She was delivered vaginally without complication. The child was pale and anemic with severe active bleeding from her vagina. After parenteral fluid and blood replacement were given, her vagina was examined under general anesthesia via a small nasal speculum. Unfortunately cervix could not be viewed clearly because of severe bleeding from uterus. On auscultation vascular murmur was heard suprapubically. Consequently angiography was performed by introducing a catheter through the right

femoral artery, and a large arteriovenous malformation at posterior aspect of lower uterine segment was detected. This area was supplied by anastomosis of multiple small vessels from both uterine arteries. After continuing bleeding, exploratory laparotomy was subsequently performed. Intraoperative findings were a normal small size uterus, normal fallopian tubes and ovaries. External appearance of the uterus was normal, but vascular pulsation at posterior aspect of lower uterine segment could be felt. Bilateral hypogastric arteries, right uterine artery and uterine branch of right ovarian artery ligations were undertaken. The uterine bleeding ceased postoperative and the course of recovery was uneventful. She was discharged on the 7th postoperative day.

One month later, she was readmitted to the hospital with recurrent severe vaginal bleeding. After the patient's resuscitation, exploratory laparotomy was performed and followed by total hysterectomy with conservation of ovaries to control bleeding. She recovered gradually without any complication. The pathological findings are shown in figure 1 showing multiple large blood vessels. She left the hospital at the 7th postoperative day and was follow up periodical up to a period of two years without recurrence of bleeding. Recently in 1999, at the age of 10 years old, the patient was asked to have a pelvic ultrasonography taken and was found to have normal ovaries, normal secondary sexual development and no

recurrence of her previous gynecological disorder.

## Discussion

Arteriovenous malformation (AVM) of the uterus is a rare gynaecological disorder particularly in a young child.<sup>(1-5)</sup> The first case was reported by Dubreuil and Loubat in 1926.<sup>(10)</sup> It consists of a proliferation of arterial and venous channels with fistula formation and an admixture of small capillary-like channels.<sup>(2)</sup> These malformations may occur as a result of either congenital or acquired disorders.<sup>(1,2,4,6,7)</sup> Congenital AVM may result from a remnant of embryonic vascular connections.<sup>(11)</sup> Acquired AVM usually results from pelvic trauma, surgery or curettage, inflammation, diethylstilbestrol exposure, gestational trophoblastic disease, previous pregnancy, endometrial or cervical cancer.<sup>(2,3,5,12-14)</sup> Our case, from age and history, was most likely to be congenital in origin.

In most reported cases, the women's ages were between 20 and 40 years old.<sup>(4)</sup> The youngest patient was a stillborn at 34 weeks of gestation.<sup>(9)</sup> The oldest patient was a 72 year-old woman.<sup>(15)</sup> We reported additional case of 7 month old child who was finally treated by total hysterectomy after failed conservative bilateral hypogastric ligations.

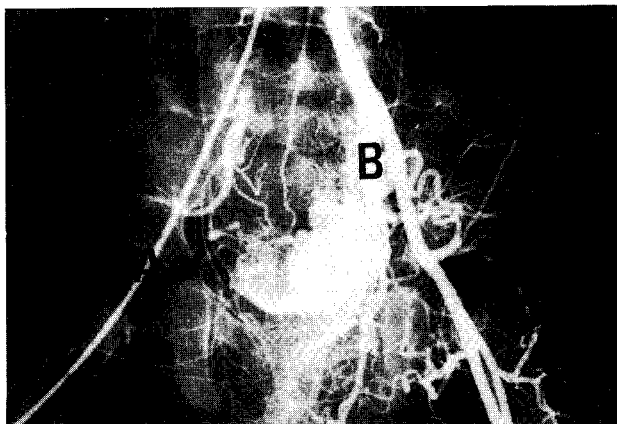
The clinical presentation of AVM of the uterus is variable. The most common presenting symptom is vaginal bleeding or menorrhagia.<sup>(1,2,4-8,11)</sup> Other presenting symptoms are postmenopausal bleeding, congestive heart failure secondary to blood shunting, or abdominal pain.<sup>(11,14)</sup> Our case was presented with vaginal bleeding similar to several previous reports.<sup>(1,2,4-8,11)</sup> However asymptomatic cases of AVM of the uterus have also been found accidentally at the time of hysterectomy.<sup>(5)</sup> A clinical sign which is suggestive of AVM of the uterus is detection of an enlarged uterus with a vascular pulsation on bimanual examination.<sup>(3)</sup>

The gold standard for diagnosis of AVM of the uterus is the angiography.<sup>(3-5)</sup> Despite invasive procedure, the advantage is allowing differentiation of an AVM from a vascular tumor;<sup>(3)</sup> allowing delineation of the draining veins and feeding arteries and their

relationship to the normal circulation of the pelvis;<sup>(3,11)</sup> and guiding for embolization therapy.<sup>(8,11)</sup> Other non invasive diagnostic methods include sonography and color Doppler ultrasonography<sup>(2-8,11,14,15)</sup> which may be able to detect hypoechoic areas in the myometrium showing vascular flow. In addition contrast-enhanced computed tomography (CT) and magnetic resonance imaging (MRI) have been shown to be valuable method of non invasive diagnostic procedure.<sup>(17,18)</sup>

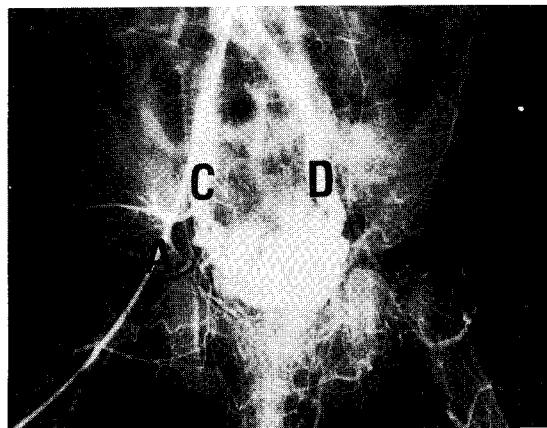
The choice of therapy is depending on the desire for fertility and the symptomatology.<sup>(3,4,11)</sup> Hysterectomy is the treatment of choice for symptomatic patients with no desire for future fertility, and refractory bleeding or severe uterine bleeding.<sup>(2,3,11)</sup> Recently, an alternative conservative therapy has been offered to patients who desire for future pregnancy. This consists of selective embolization of the defective arteries using various materials such as gelatin sponge, coils, isobutyl-2-cyanoacrylate, detachable balloons, thrombin, and polyvinyl alcohol sponge<sup>(3-5,12,13,19,20)</sup> Another successfully conservative therapy using methylergonovine maleate had also been reported.<sup>(6)</sup> In our reported case the young child was finally hysterectomised because of persistent severe refractory uterine bleeding, possibly due to rupture of a blood vessel at the endometrial surface of the uterus.

In conclusion, the possibility of an AVM of the uterus should be aware in cases of vaginal bleeding especially refractory bleeding or bleeding in young girls. Because an inappropriate procedure such as curettage in these patients may cause massive hemorrhage. Conservation of uterus and fertility may be possible today by using modern technique of embolization.



**Fig 1.**

A: Catheter in right femoral artery  
B: arterial phase showing A/V malformation



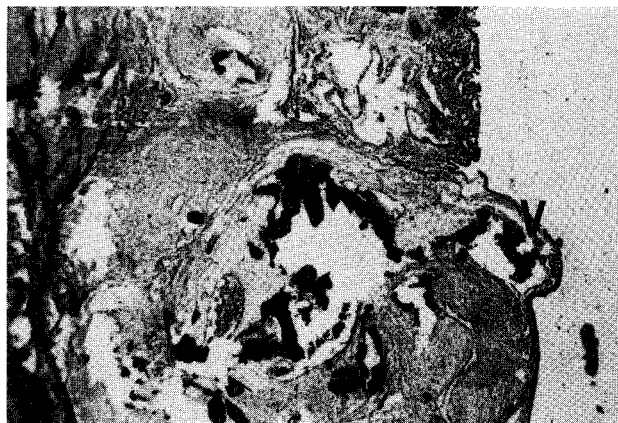
**Fig 2.**

A: Catheter  
C: right femoral vein (venous phase)  
D: left femoral vein & A/V malformation



**Fig 3.**

Histology showing multiple arterial blood vessels with thick muscle wall



**Fig 4.**

Histology showing rupture of blood vessel at endometrial surface

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