Sufia Athar¹ and Badreldeen Ahmad²*

¹Specialist Obstetrics and Gynecology, Al Wakra Hospital, Qatar
²Professor of Clinical Obstetrics and Gynecology, Weill Cornell Medicine, Qatar

ABSTRACT

We present a case of acquired arteriovenous malformation with scar pregnancy in a patient with a previous Caesarean for placenta previa. Incidence of scar pregnancy is reported as 1 in 2,000 pregnancies. The incidence of arteriovenous malformation with scar pregnancy is rarer. Due to paucity of literature regarding arteriovenous malformation with scar pregnancy, these cases may present as a diagnostic dilemma for the treating physician and usually lead to delay in the management of these cases. This is case is unique in sense that she presented with massive vaginal bleeding after 4 month of diagnosis when the pregnancy test was negative.

CASE REPORT

A 33 years, G6P3 with 9 weeks pregnancy was referred with the diagnosis of missed miscarriage. She was clinically asymptomatic. She had conceived after a short pregnancy interval of 8 months following her previous Cesarean for placenta previa. Her previous Cesarean was uneventful.

The patient was offered a pelvic scan for confirmation, which revealed a low implanted gestational sac with missed abortion (Figure 1). However, the level of beta HCG was 162,288 mIU/ml so patient was requested to repeat beta HCG after 48 hours. However, she reported back after 9 days without any complaints. Beta HCG was 76,807 mIU/ml. Though HCG was showing a declining trend, but HCG value was high, so she had another pelvic scan which showed placental tissues invading the previous scar regions and likely associated with AVM (Figure 2). She was admitted in the hospital, and MRI was requested. She was offered diagnostic laparoscopy plus hysteroscopy to confirm the diagnosis, but the patient refused as she is worried about her future ability to conceive. She was kept in the hospital under observation and MRI was done. MRI was suggestive of miscarriage with possibility of molar pregnancy, scar pregnancy was excluded (Figure 3,4). Beta HCG dropped to 52,982 mIU/ml. So was had medical management of miscarriage with Misoprostol. She did not passed tissues, so she was offered second cycle of medical management and option of surgical evacuation was discussed, but she refused and went against medical advice. Later she had multiple visits to the emergency department and on every visit, she refused surgical management. Even though, she was counselled on every visit regarding the potential complications, she refused any further management. She was followed up till the beta-HCG level was less than 1 mIU/ml. The ultrasound appearance of the uterus at this point was shown in figure 3.

Figure 1: First scan of the patient (8 weeks POG), revealing low implanted gestational sac with yolk sac and fetal pole. No cardiac activity. Note the hematoma in front of the gestational sac with increased vascularity.

Figure 2: Second scan of the patient (10 weeks). Extensive per gestational abnormal irregular bizarre vascular spaces, with very low resistance suggestive of AVM.

Figure 3 – This scan picture after negative pregnancy test, revealing low implanted gestational sac with yolk sac and fetal pole. No cardiac activity. Extensive per gestational abnormal irregular bizarre vascular spaces suggestive of AVM. Image A, without color Image B, with color Doppler

Figure 4: MRI images: Areas of abnormal signals in the posterior lower uterine segment were noted, highly suspicious of AVM.
After 4 months, she reported to ER with severe bleeding with passage of clots. She was clinically unstable with evident tachycardia (Pulse-116/m), pallor and hypotension (BP 98/56 mm Hg -dropped further to 80/62 mm Hg). Estimated blood loss in the Emergency room was 1500 ml. She was resuscitated, and she received 4 units of the packed blood cells. Bed side scan revealed heterogenous, rounded lesion in the lower uterine cavity measuring 5.8x4.6cm with cystic vascular spaces with near total loss of myometrium anteriorly with thin uterus bladder interface. No evidence of intraperitoneal blood/collection was noted. Beta HCG was <1. She was taken to theatre for an ultrasound guided evacuation of products of conception. Product of conception was seen at the level of internal cervical Os which was removed easily with a sponge forceps. This is was followed by dilation and curettage, small remnant of tissue less than 2 cm in diameter could not be removed because it was firmly adherent to the site of the scar. The estimated blood loss during her surgery was 1000 ml. The postoperative bed side ultrasound showed empty cavity and no free fluid. Histopathology showed necrotic product of conception.

She was monitored in the high dependency unit for 48 hours. She had uneventful post-operative period. Post-operative
follow up scan after two weeks revealed absence of AVM (Figure 5). Figure 6 showed the trend in her Beta HCG level (Figure 6).

**DISCUSSION**

Arteriovenous Malformation (AVM) also known as Enhanced Myometrial Vascularity (EMV), are anomalous channels between arteries and veins. They may present as a normal variant (primary) or may be formed after any surgical intervention or trauma (acquired) [1,2]. Uterine AVM may be acquired after Cesarean Sections, dilatation and curettage and after myomectomy [3]. Recent Studies have illustrated their occurrence even after trophoblastic diseases, cancer endometrium and cancer cervix [4,5]. The occurrence of AVM with scar pregnancy has been established by several authors [6,7,8].

Incidence of scar pregnancy is reported as 1 in 2,000 pregnancies [3]. The incidence of AVM with/after scar pregnancy is rarer. However due to paucity of literature regarding AVM with scar pregnancy, these cases may present as a diagnostic dilemma for the radiologist.

The presentation of AVM is variable. Patients may be clinically asymptomatic or may have catastrophic bleeding episodes. Many authors have revealed the cases of AVM with torrential vaginal bleeding [9,10].

In most of the cases the diagnosis is delayed as the differential diagnosis of low implantation, cervical pregnancy, gestational trophoblastic disease and scar pregnancy creates diagnostic dilemma for the radiologist and the clinicians. The diagnosis is even more arduous in cases of missed miscarriage. Downscaling titers of HCG often prompts delays in the management of these cases. However due to its rarity and diagnostic dilemma, usually cases are diagnosed very late, often confirmed only after severe hemorrhage and complications.

In cases of low implantation gestational sac with previous history of uterine trauma (previous Cesarean sections, dilatation and curettage, myomectomy etc.) the suspicion of ectopic pregnancy is high. In these cases, the radiologist may find reporting the diagnosis challenging. However, patients’ history and clinical presentation may aid in the diagnosis. In cases of suspected /confirmed scar pregnancy, vascularity should be commented in every case to exclude the possibility of AVM/EVC. AVM/EVC may be misinterpreted as products of conception if vascularity is not assessed as radiological images may be analogous.

Color Doppler scan, in cases of AVM usually reveals turbulent flow in the aberrant vasculatures with intense signal, which may be noted near the previous scar or in the myometrium. These vessels usually demonstrate high velocity within low resistance vessels. Peak Systolic Velocity (PSV) is a useful predictor of the extent of proliferation of AVM. Usually AVM with PSV greater than 60 cm/s are associated with intractable bleeding and complications. These AVMs are less likely to regress spontaneously. In AVM with PSV ≤ 40 cm/s, the chances of spontaneous regression are higher, and likelihood of adverse outcomes is less.

In case of doubt, other modalities for the confirmation of the diagnosis as MRI, hysteroscopy and laparoscopy ought to be offered to avoid delay in the management and to prevent future complications. AVM may be noted as multiple aberrant and enlarged myometrial and para-metrial blood vessels in MRI.

Different modalities for management of AVM/EVC are illustrated by authors. Main aim of management is to prevent complications of gestational sac rupture as it may provoke intractable bleeding. In these patients, medical verses surgical management should be individualized. In cases with failing pregnancy medical management with systemic/local Methotrexate may be offered. Other medical options are local injection of Potassium Chloride and oral Mifepristone. Combined use of Methotrexate and Potassium Chloride is also suggested by some authors.

Some authors have suggested uterine curettage, evacuation under ultrasound/hysteroscopic guidance while others have emphasized the benefits of laparoscopy and laparotomy.

Uterine Artery Embolization is the treatment of choice of AVM/EVC [6]. It has higher success rate and allows preservation of future fertility [11,12]. The success rate after UAE for cases with traumatic AVMs is reported 93 % by Ash et al. [13-16]. No technical issues in the procedure were encountered in UAE of their study [16,17]. In cases with no response after UAE, surgery, repeat UAE, laparoscopy, or hysterectomy may be considered [6,8]. Many authors have illustrated the success after UAE in cases with post –partum hemorrhage [3,5,7,9]. Due to rarity of association of AVM with

scar pregnancy, limited literature is available. Smith et al I their case report specified that the AVM with scar pregnancy may need selective UAE. In these cases, surgical management remains the only option in some cases [11]. Rygh et al have also reported the need for surgery after UAE in a case of scar pregnancy with AVM.

In refractory cases with severe bleeding, hysterectomy may be the sole option [6]. Comprehensive evaluation of patient, including a meticulous history and thorough examination, followed by a transvaginal scan with color Doppler may minimize missed cases. In cases with previous history of trauma to the lower uterine segment, ultrasound reporting should be performed with caution. In suspected cases of scar pregnancy with/without AVM/EVC further evaluation should be carried out. Early diagnosis and planned management is the key to avoid irreversible complications later.

REFERENCES