Acquired Uterine Vascular Anomaly

Experience from a tertiary care center in Pakistan

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Abstract

Objective: To retrospectively review imaging findings and the outcomes of uterine artery embolization (UAE) in symptomatic uterine vascular anomalies. Methods: We identified 15 cases of acquired uterine vascular anomaly from 2010 to 2020 who were evaluated with ultrasound, computed tomography, and magnetic resonance imaging, either alone or in combination. All patients had history of dilatation and curettage or uterine instrumentation. They underwent angiography and embolization of the uterine arteries. Primary outcome post embolization was assessed clinically and/or in combination with ultrasound. Post procedure pregnancies were also recorded. Results: Non-invasive imaging was abnormal in all patients, however this pre intervention imaging was unable to accurately classify the type of vascular anomaly except in the case of pseudoaneurysm. Conventional angiography showed uterine artery hyperemia in 6, arteriovenous malformation in 7 and pseudoaneurysm in 2 patients. The technical success rate was 100% with no repeat embolization needed. Follow up ultrasound in 12 patients revealed resolution of abnormal findings, remaining three were normal on clinical follow up. Seven patients (46.7%) had a normal pregnancy, 15.7 months after the procedure.
(range 4-28 months). **Conclusion:** UAE is a safe and effective management option for intractable severe bleeding in patients with uterine vascular anomaly post instrumentation and is seen not to impair future pregnancy.

**Keywords:** Uterine artery embolization; Pseudoaneurysm; Arteriovenous malformation; Ultrasound; Computed tomography; Magnetic resonance imaging.

**Advances in Knowledge**
- All hypervascular lesions in the uterus on ultrasound are not true arteriovenous malformations. Placental subinvolution theory should be kept in mind when assessing patients for suspected uterine vascular anomaly post-abortion on imaging.

**Application to Patient Care**
- Pelvic artery embolization prevents hysterectomy in cases of severe vaginal bleeding.
  - Embolization of the uterine artery does not preclude successful future pregnancy

**Introduction**
Uterine vascular anomalies (UVA) are classified into two main types, a) vascular malformations, and b) vascular neoplasms, according to the International Society for the Study of Vascular Anomalies classification system.\(^1\) The first category includes several entities such as venous malformations, arteriovenous fistulas, pseudoaneurysms, arteriovenous malformations (AVMs), and rarely a combination of pseudoaneurysm and AVM.\(^1-3\) AVMs are the most reported uterine vascular anomaly although their true incidence is unknown.\(^4,5\) These may be either congenital or acquired, the latter being far more common.\(^4,6\) Acquired uterine AVMs are most commonly secondary to uterine trauma, such as curettage or uterine surgery, which results in abnormal communication of uterine artery branches with the myometrial venous plexus and lack a true nidus.\(^4,7,9\)

Rare causes of uterine AVM include endometrial or cervical carcinoma, leiomyoma, uterine infection, gestational trophoblastic diseases or endometriosis.\(^9-11\) Direct communication between artery and vein results in arteriovenous fistulas.\(^1,12\) Pseudoaneurysms constitute another rare acquired vascular anomaly. These are focal areas of confined defects communicating with the
vessel lumen through a traumatic defect, frequently post pelvic/uterine surgery or a curettage procedure, and lack a true wall.\textsuperscript{2} Patients with acquired uterine vascular anomaly present with acute heavy bleeding, which may be intermittent or continuous.\textsuperscript{5,8} Other symptoms such as lower abdominal pain, urinary frequency or incontinence, dyspareunia, and hypotension or hypovolemia secondary to blood loss have also been described.\textsuperscript{11} All suspected cases of uterine AVM initially undergo ultrasound evaluation for diagnosis, supplemented by computed tomography (CT) or magnetic resonance imaging (MRI) in cases where ultrasound is inconclusive.\textsuperscript{8,12} Although conventional angiography is the gold standard for diagnosis, it is reserved for cases that are unresponsive to conservative management and in whom a therapeutic embolization is planned.\textsuperscript{13-16}

The purpose of our study was to review the diagnostic accuracy of imaging in identifying uterine vascular anomalies in symptomatic patients and to assess the technical success of percutaneous uterine artery embolization (UAE) in the management of this patient cohort.

**Methods**

This study was a retrospective analysis conducted at the Aga Khan University Hospital (AKUH), Karachi after taking approval from the institute’s Ethical Review Committee (Ethical Review Number: ERC # 2020-3690-10189). We searched our Radiology database for patients undergoing UAE from January 2010 to May 2020. After excluding patients with known uterine tumors, retained products of conception, gestational trophoblastic disease, and post-partum hemorrhage, we identified 15 cases suspected of uterine vascular anomaly clinically and on imaging.

A pre-structured proforma was used to record patient demographics including age, parity, pattern, and volume of vaginal bleeding, history of uterine surgery or dilatation and curettage (D&C), time interval since the intervention, findings on imaging and angiography, and patient outcome. The duration of hospital stays, post-procedure complication, follow-up ultrasound findings, and post-embolization fertility/pregnancy were also recorded. The patient’s imaging was reviewed on picture archiving and communication system (PACS), Rogan Delft View Pro-
X, while additional data was collected from the Health Information Management Services (HIMS).

The pre angiography imaging modality was chosen at the discretion of the referring physician, which included ultrasonography with color Doppler Imaging, pelvic MRI, and CT, either alone or a combination. The referring physician decided on embolization after consulting with the interventional radiologist. In the angiographic suite of Aga Khan University Hospital, interventional radiologists performed embolization procedures. Consent was taken in every case to explain the benefits and risks. Under local anesthesia, the procedure was performed on a flat panel monoplane digital subtraction angiography machine Axiom-Artis, Siemens. The femoral artery was punctured and a 4F vascular access sheath was inserted. A 4Fr Simmons (SIM 1) catheter (Cordis), or a Cobra (C1) angiographic catheter (Cordis) was advanced over a 0.035-inch guidewire. An angiographic run was performed after selective catheterization of the uterine artery, followed by super-selective cannulation using a microcatheter (Progreat Terumo) which was placed coaxially as near as possible to the feeder vessel. The embolization materials used were polyvinyl alcohol particles (PVA), size 355-500 μm, gel foam, glue, and coil, either in combination or isolation. In a few cases, the ovarian artery was also embolized. Clinical success was defined as resolution of vaginal bleeding and/or abnormal imaging findings on post embolization follow-up.

SPSS version 20 was used for statistical analysis. Quantitative data were expressed as mean ± standard deviation; qualitative data were expressed using frequencies (percentages). Descriptive analysis was done for all variables, including the demographic variables as well as the other categorical variables, and frequencies, proportions, and percentages were reported.

**Results:**
The mean patient age was 28.2 years (range: 20-35 years). Fourteen patients had undergone a prior uterine procedure. Twelve patients had a prior D&C, One patient had a repair of a uterine rupture and one patient had a C-section. The patient without prior D&C or surgery had a history of medical termination of pregnancy.
The clinical features are shown in Table 1. Main presenting complain was abnormal per vaginal bleeding. It was considered mild if there was only spotting, when there was continuous bleeding but no clots it was labelled as moderate and severe when there was passage of clots. The mean time interval of patient presentation after the intervention was 64.6 days (range: 1-365 days).

All patients underwent pre-embolization US except one, who underwent only MRI examination. Greyscale ultrasound identified an abnormal area in the myometrium in 12 patients (85.7%) and an abnormal area in the endometrium in 2 patients. On Doppler imaging, eight patients showed mixed arterial/venous flow. Six of the eight were confirmed as AVM on angiography (figure 1) while the other two showed only uterine hyperemia on angiography. Four patients showed focal increased vascularity within the myometrium, one turned out to be an AVM on angiography while the other three just showed uterine artery hyperemia. Two patients showed pseudoaneurysms on Doppler that were confirmed on pre-embolization CT and angiography (figure 2). Two other patients had abnormal focal vascularity on arterial phase with prominent veins on pre-embolization CT suggesting AVM. Among these, one proved AVM on angiography (figure 3) while the other showed uterine hyperemia only.

Six out of 15 patients underwent pre-embolization MRI. MRI findings demonstrated abnormal signal intensity areas in either the myometrium or endometrium with abnormal enhancement and multiple flow voids suggesting AVM. Three of these were confirmed as AVM (figure 4) on angiography, while the other three showed enlarged, prominent uterine arteries.

The details of the angiographic findings and procedures are shown in Table 2.

The embolization procedure was technically successful in all 15 patients, and none required a repeat embolization or post embolization transfusion. None of the patients had an on-table procedure-related or puncture site complication. The mean duration of hospital stay was 2.73 days, (range: 2 - 4 days). Nine out of fifteen patients had mild bleeding at the time of discharge from the hospital which resolved by the next clinic visit. Two patients had an episode of per vaginal bleeding a month later which responded to conservative management.

Twelve patients underwent follow-up ultrasound examinations. The mean time of the follow-up ultrasound, after embolization, was 40.6 days (range- 15 to 90 days). In five patients, the follow-
up ultrasound was completely normal. Five patients showed persistent greyscale findings, however, abnormal vascularity had resolved. One patient showed a decrease in size of the abnormal area on ultrasound with persistent mild vascularity, although she was asymptomatic. In another patient, both greyscale and Doppler abnormality was demonstrated on initial follow-up ultrasound, but it resolved completely on repeat ultrasound two months later. Three patients did not have any follow-up imaging but were clinically asymptomatic.

Seven patients (46.7%) had normal pregnancies that carried to term after the procedure. The mean time interval between the procedure and the pregnancy was 15.7 months (range 4 to 28 months). The remaining eight did not conceive to our knowledge.

**Discussion**

Our retrospective study at a tertiary referral center reviewed the spectrum of imaging findings in patients with suspected acquired vascular uterine anomalies and the outcomes of super-selective UAE. We found true AVMs in seven cases including one arteriovenous fistula. Additional anomalies that we found were uterine hyperemia and pseudo-aneurysm. Timmerman et al studied 30 cases of suspected uterine vascular malformations out of which eight underwent angiography. Their study showed true AVMs in three patients while the rest had only abnormal arterial blush. Occasionally, the number of AVMs is purportedly higher on (conventional) angiography. Hugues et al did a study on 26 cases of suspected uterine AVMs, of which about a quarter showed uterine hyperemia, whilst true AVMs were observed in the remaining cases. Also, a study of iatrogenic uterine arterial injuries, which were treated by UAE, found AVMs in the majority of cases (15 out of 24), either alone or in combination with pseudoaneurysm.

All of our cases showed abnormalities on both greyscale and color Doppler ultrasound examinations. Both pseudoaneurysms were accurately identified. Pseudoaneurysms appear as cystic spaces on greyscale ultrasound which show swirling multidirectional flow on color Doppler with varying degrees of turbulence, allowing for correct identification in most cases. The greyscale appearances of AVMs, on the other hand, are non-specific, ranging from subtle myometrial inhomogeneity to linear, anechoic spaces in the myometrium which show color
filling on Doppler interrogation with a mosaic pattern.\textsuperscript{12,19} The specificity is increased by Duplex US/ spectral analysis which reveals high velocity, low resistance arterial flow.\textsuperscript{17}

Unfortunately, the spectral analysis findings were not available in all our cases as mostly greyscale and color Doppler evaluation was done. It has been seen that low resistance abnormal vascularity often persists in the myometrium at the site of trophoblast/placental implantation and takes time to resolve.\textsuperscript{1,11} This has been referred to as sub-involution of placental bed and may account for abnormal findings on ultrasound and MRI as seen in some of our cases which subsequently demonstrated only uterine hyperemia on angiography with no AVM. Nevertheless, angioembolization was justified as these patients presented with moderate to severe bleeding that had been resistant to conservative management. Symptoms completely resolved following a single session of embolization, with no discernable adverse effects.

Currently, digital subtraction angiography (DSA) is the gold standard for the diagnosis of uterine vascular anomalies; however, its use is not justified unless it is the precursor to an embolization. The management of uterine vascular anomalies depends on the clinical presentation as well as the severity of the anomaly. Patients who have minimal symptoms and/or are hemodynamically stable may be followed clinically and by ultrasound. Bleeding usually resolves spontaneously within weeks to months in milder cases.\textsuperscript{20} UAE is offered to patients with severe intractable or recurrent bleeding. It avoids hysterectomy preserving chances of future fertility. UAE may theoretically result in reduced vascular supply to the uterus; however, the presence of rich collaterals prevents uterine infarction.\textsuperscript{13} Many case reports and studies have shown successful pregnancy outcomes post angioembolization.\textsuperscript{13,20,21} Nearly half of our cases had pregnancies that carried to term, post embolization.

Several embolic agents have been used for the treatment of UVAs. These include gel foam, PVA particles, glue, coil, or a combination.\textsuperscript{13,15,16,18,22,23} In our study, PVA particles were the most used embolization material, used in thirteen out of fifteen cases. It was the sole embolic agent in ten cases and used in combination with gel foam in one case and combination with histoacryl glue and coil in two patients. Gel foam was used as the sole embolic agent in one case. One
patient with a right uterine artery pseudoaneurysm was embolized with cyanoacrylate glue. The reported complication rate of pelvic artery embolization is low.\textsuperscript{23}

Minor complications such as puncture site pain or hematoma, fever, and transient lower limb and buttock claudication are more frequent than rare severe complications such as iatrogenic rupture of a pelvic artery, sloughing of perineal skin, vesicovaginal fistula, or major distal ischemia.\textsuperscript{13,22,23} We did not encounter any major post-procedural complications. Only three patients experienced mild abdominal pain not requiring any treatment while fever was observed in two patients which resolved before discharge.

**Conclusion**

Ultrasound is the first-line imaging modality employed for the diagnosis of uterine vascular anomaly. It has high specificity for pseudoaneurysms but lacks specificity for AVMs. It is important to consider placental bed sub-involution whilst diagnosing AVMs on non-invasive imaging in cases with a history of a recent miscarriage. In summary, UAE is a safe and effective management option for intractable severe bleeding in patients with uterine vascular anomaly post instrumentation and does not limit future pregnancy outcomes.

**Conflict of Interest**

The authors declare no conflicts of interest.

**Funding**

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**Author Contributions**

KF, RS and MA conceived the idea, KF and MZ collected the data, KF, MZ and RS analysed and interpreted the data, KF, RS and MZ drafted the article, MA critically reviewed the manuscript and supervised the study. All authors approved the final draft.

**References:**


Table 1. Baseline characteristics of the study population

<table>
<thead>
<tr>
<th>Age</th>
<th>Years (mean)</th>
<th>28.2, range (20-35)</th>
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<tbody>
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<td></td>
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<tr>
<td>1-2</td>
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<td></td>
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<tr>
<td>&gt;3</td>
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<td></td>
</tr>
<tr>
<td>History of D&amp;C</td>
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<td></td>
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<tr>
<td>Time since D&amp;C, days (mean)</td>
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<td></td>
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<tr>
<td>History of uterine surgery</td>
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<td></td>
</tr>
<tr>
<td>History of MTP</td>
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</tr>
<tr>
<td>Amount of bleeding</td>
<td></td>
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</tr>
<tr>
<td>Moderate</td>
<td>6 (40%)</td>
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</tr>
<tr>
<td>Severe</td>
<td>9 (60%)</td>
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<td>Pattern of bleeding</td>
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<tr>
<td>Intermittent</td>
<td>9 (60%)</td>
<td></td>
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<tr>
<td>Continuous</td>
<td>6 (40%)</td>
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</table>

D&C: Dilatation and curettage, MTP: Medical Termination of Pregnancy

Table 2. Details of the Embolization procedure

<table>
<thead>
<tr>
<th>Case</th>
<th>Angiographic Finding</th>
<th>Vessels embolized</th>
<th>Embolization material used</th>
<th>Complications</th>
<th>Duration of hospital stay</th>
<th>Post embolization pregnancy</th>
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<td>PVA</td>
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<td>Hyperemia</td>
<td>B/L Uterine Artery</td>
<td>PVA</td>
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<td>3</td>
<td>Hyperemia</td>
<td>B/L Uterine Artery</td>
<td>PVA + gel foam</td>
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<td>U/L Uterine Artery</td>
<td>PVA</td>
<td>Fever</td>
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<td>5</td>
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<td>PVA</td>
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<tr>
<td>6</td>
<td>Hyperemia</td>
<td>B/L Uterine Artery</td>
<td>PVA</td>
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<td>7</td>
<td>AVM</td>
<td>B/L Uterine Artery</td>
<td>PVA</td>
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<td>8</td>
<td>AVM</td>
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<td>PVA</td>
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<td>9</td>
<td>AVM</td>
<td>B/L uterine artery</td>
<td>PVA</td>
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<td>Gel foam</td>
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<tr>
<td>11</td>
<td>AVM</td>
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<td>Fever</td>
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<tr>
<td>12</td>
<td>AVM</td>
<td>B/L uterine artery</td>
<td>PVA</td>
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<tr>
<td>13</td>
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<td>B/L uterine artery + ovarian</td>
<td>PVA + coil + glue</td>
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<tr>
<td>14</td>
<td>Pseudoaneurysm</td>
<td>U/L uterine artery</td>
<td>Glue</td>
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<td>PVA + coil + glue</td>
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</table>

AVM: arteriovenous malformation, B/L: bilateral, U/L: unilateral, PVA: polyvinyl alcohol

Figure 1: A 25-year-old with moderate vaginal bleeding. (A) Doppler ultrasound shows a mosaic color pattern in the myometrium on the right side extending into endometrium indicating both arterial and venous flow (arrow), (B) digital subtraction angiography (DSA) image shows the AVM supplied by right uterine artery, (C) DSA image shows the early draining vein (black arrow), (D) post embolization shows resolution of the AVM.
Figure 2: A 29-year-old with moderate vaginal bleeding post uterine surgery. (A) Greyscale transvaginal ultrasound image shows an irregular anechoic area in the myometrium (arrow) with the turbulent arterial flow on spectral analysis, (B) Doppler image shows heterogeneous color filling in the pseudoaneurysm, (C) and (D) digital subtraction angiographic images, left uterine artery (long white arrow) and left ovarian artery (black arrow) supplying the pseudo-aneurysm.
Figure 3: A 25-year-old with continuous vaginal bleeding. Transvaginal ultrasound Doppler images (A) show abnormal vascularity in the uterine myometrium, (B) Coronal contrast-enhanced CT image confirms abnormal myometrial vascularity and dilated draining gonadal vein (white arrow), (C) DSA image shows an abnormal bunch of vessels supplied by right uterine artery, (D) post embolization image showing complete resolution of the AVM.
Figure 4: A 31-year-old with severe vaginal bleeding. (A) T2 weighted sagittal MRI shows a heterogeneous bulging mass with serpentine signal voids involving the lower uterine cavity and anterior myometrium (white arrow). No myometrium is seen between the urinary bladder and this mass. (B) Coronal T2 weighted image shows multiple serpentine signal voids (white arrow). (C) Digital subtraction angiographic image shows dilated tortuous right uterine artery supplying AVM (D) post embolization image showing complete resolution of the AVM.