Case Report:

Catastrophic Post-cesarean bleeding due to arteriovenous malformation: Case report

[1]Dr. Data Shilpa, [2]Dr.Mule Vidya , [3]Dr. Dinde Meenakshi , [4]Dr. Patil Shrenik

[1,2,3]Department of OBGY ,GMC , Miraj , Maharashtra , India

[4]Vedh Diagnostic, Sangli , Maharashtra , India

Corresponding author: Dr. Date shilpa

Abstract

Uterine arteriovenous malformation (AVM) is a rare but life threatening condition presents usually with torrential bleeding common in reproductive age group. We are presenting a case of 23 year primipara reported with profuse bleeding two months after Cesarean section. Ultrasonography (USG) with Doppler showed presence of heterogeneous area near right side of cervix with feeding vessel having high velocity, low resistance more likely to be AVM. Because of poor economy and lack of facilities, pelvic angiography and uterine artery embolization was not possible. We have performed bilateral Hypogastric artery ligation with ovarian branch ligation. Follow up USG described significant reduction of previously described lesion with no internal and peripheral vascularity. Patient didn't have any episode of profuse bleeding since then (six months from surgery). Conservative management has been proved successful.

Key Words : Arteriovenous malformation, Ultrasonography, Color Doppler, pelvic angiography, Hypogastric artery ligation.

Introduction:

Uterine arteriovenous malformation (UAVM) is a rare condition with fewer than 100 cases reported in literature [1]. It results from formation of multiple arterio-venous fistulous communications within the uterus without an intervening capillary network. Presentation can vary. UAVMs can cause life-threatening massive genital bleeding in young women. Bleeding is the major presenting symptom in AVM constituting almost 1-2% of cases of genital bleeding.

Case Report:

23 years old P_1L_1 was referred from private hospital for profuse per vaginal (P/V) bleeding. She had two episodes of sudden onset profuse bleeding and was found unconscious at home. Six units of whole blood were transfused and for further management she was referred to our institute. She had undergone emergency Cesarean Section two months back for deep transverse arrest with history of extension of incision on Right side uterine artery which was ligated during the procedure.

On admission her vitals were stable with hemoglobin level of 9.5g/dl. Mild pallor was noticed. On per speculum examination minimal bleeding was present through os and on P/V examination uterus was of normal size, anteverted, smooth, firm, mobile with no forniceal mass or tenderness. Ultrasonography with color Doppler was done which showed evidence of subendometrial or intramural heterogenous lesion with few cystic areas in it on right side of cervix of size 19x25 mm (Fig 1). A feeding vessel was noted supplying this lesion with high velocity low resistsnce flow (Fig 2) -possibility of AVM, other less likely possibility is fibroid. For confirmation

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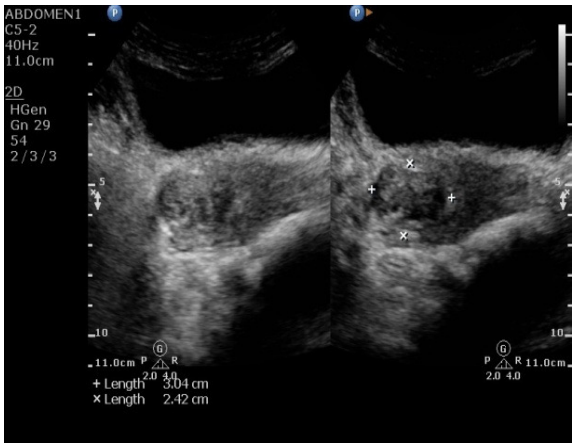
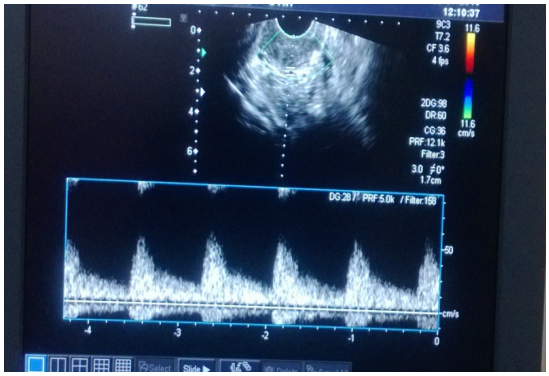
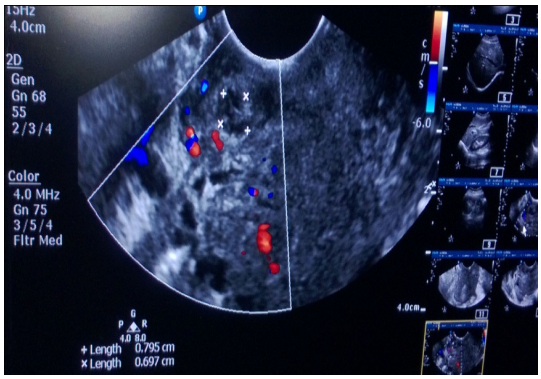
pelvic angiography or Magnetic Resonance Imaging (MRI) was advised. Option of uterine artery embilization was also given to relatives. As those facilities were not available in our institute and because of patient's economic constraints we decided to go for surgical uterine devascularization. Patient was posted for exploratory laparotomy. There was no visible or palpable uterine anatomical as well as vascular abnormality found during surgery. Decision of uterine devascularization was finalized and conveyed to relatives and consent obtained for same. Bilateral anterior division of internal iliac artery was ligated followed by ligation of ovarian branches supplying uterus with O'Leary stitch. Postoperative course was uneventful. Patient was discharged on 7th postoperative day. Ultrasonography was repeated after a month which described reduction in size of previous lesion to 7x8mm with no peripheral or internal vascularity (Fig 3). Patient didn't have any abnormal P/V bleeding on six months follow up till date.







Discussion :

AVM consists of a proliferation of vascular channels with fistula formation and an admixture of small, capillary-like channels. The size of these vessels can vary considerably. They are classified as congenital or acquired. The latter are more common and are often described as a uterine arteriovenous fistula. Congenital UAVMs tend to have multiple feeding arteries, a central nidus (a tangle of vessels with histologic characteristics of both arteries and veins), and numerous large draining veins .Acquired or traumatic UAVMs represent multiple small arteri-ovenous fistulas between intramural arterial branches and the myometrial venous plexus. They typically represent a single artery joining a simple vein [2-4]. Acquired UAVM's disease is associated with obstetric disorders and procedures such as multiple pregnancies, spontaneous abortion, previous dilation

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and curettage, therapeutic abortion, and / or Caesarean section. It is also associated with infection, retained POC, gestational trophoblastic disease, gynecological malignancies and exposure to Diethylstilboestrol [3-5].

Acquired UAVM is a rare but potentially life threatening condition and as such must be considered in the differential diagnosis of cases of abrupt, profuse vaginal bleeding following uterine curettage. The condition can easily be confused with retained products of conception and gestational trophoblastic disease[6]. UAVMs result in sudden and massive vaginal bleeding that maybe life-threatening. They may occur as late postpartum hemorrhage or post abortion hemorrhage, and the bleeding results from a spontaneous vessel rupture or vessel rupture triggered by a D&C [7].

AVM was first described by G Dubreil and E Loubat in 1926 [8]. Traditionally it has been diagnosed at the time of Laparotomy or during examination of hysterectomy specimen [5] but with advent of ultrasonography and color Doppler we can diagnose this rare condition by non invasive way. Nevertheless digital subtraction pelvic angiography remains the gold standard for diagnosis [2].

Gray-scale sonographic appearances can be non-specific and can have a range of manifestations including areas of subtle myometrial inhomogeneity, tubular spaces within the myometrium, a intramural uterine, endometrial or cervical mass or like cystic lesion or sometimes as prominent parametrial vessels [3, 4].Colour Doppler typically shows serpingineous or tubular anechoic structures within the myometrium with a low resistance (RI ~ 0.2 - 0.5), high velocity flow pattern [7, 9]. Pelvic MRI allows one to confirm the diagnosis of UAVM noninvasively. Multiple serpentine flow-related signal voids are typically seen in the uterine wall, endometrial cavity, and parametrium on T1 and T2 weighted images. Contrast-enhanced dynamic MR angiography can depict complex serpentine abnormal vessels that enhance as intensely as normal vessels and show early venous return [10].

Management of UAVM depends upon hemodynamic stability, age, amount of blood loss and desire of future fertility. Traditionally Hysterectomy was treatment of choice. However in postmenopausal women or in life threatening conditions still Hysterectomy remains treatment of choice. In stable patients, who have ability for regular follow up, expectant or medical treatment can be planned as there are documented cases of spontaneous regression of AVM as well as regression with medicines [5, 11]. Embolisation of uterine AVM has been documented as definitive therapeutic modality to preserve reproductive capability [2, 3] as these cases are common in child bearing age group. Successful pregnancy outcome after embolisation has been documented in literature [12-13]. In our case due to poor economy of patient transcatheter embolisation was not possible but successful pregnancy after bilateral hypogastic artery ligation has been reported in literature [14-15].

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Date of submission: 20 January 2014 Date of Provisional acceptance: 08 February 2014
Date of Final acceptance: 27 February 2014 Date of Publication: 04 March 2014
Source of support: Nil; Conflict of Interest: Nil