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Research Article

Temporary Uterine Artery Embolization Followed by Hysteroscopy to Treat a Case of Acquired Uterine Arteriovenous Malformation (UAVM): Case Report with Systematic Literature Review

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Abstract

Objective: This paper provides information about conservative surgery to correct uterine arteriovenous malformations (UAVMs). The main objectives are to review the current literature on this subject and to share our very interesting and challenging case of UAVM. Our innovative surgical approach consisted in a combined method with temporary uterine artery embolization (UAE) plus hysteroscopic treatment.

Materials and Methods: A search of MEDLINE, EMBASE, Web of Sciences, Scopus, ClinicalTrial.gov, OVID and Cochrane Library was performed. Any type of study has been considered including case reports. We identify articles published from the inception to July 2019. Three hundred sixty-three articles were identified: 286 articles were excluded for any reason. Overall, 77 articles were incorporated for further assessment.

Results: Four surgical techniques have been employed to treat 125 cases of UAVMs: uterine artery embolization (UAE) (69.9%),dilatation and curettage (D&C) (10.4%), operative hysteroscopy (14.4%), uterine artery ligation (4%). Two patients (1.6%) underwent combined approach. Failed procedures which required a new treatment were twenty-one (16.8%). Forty-five pregnancies after surgical conservative management of UAVMs were reported.

Conclusion: UAVM is a rare condition. In literature, there is no gold standard of treatment. For young women who wish to preserve fertility, a conservative surgical management should be recommended. As in our case, using a resorbable agent to perform UAE permitted a normal restoration of the flow in the uterine artery after the hysteroscopic resection.

Keywords: Fertility; Hysteroscopy; Uterine arteriovenous malformation; Uterine artery embolization

1. Introduction

A uterine arteriovenous malformation (UAVM) is a vascular abnormal connection between arteries and veins of the uterus, which creates enlarged, tangled vessels with a fast and turbulent flow [1]. This abnormal structure results in a direct connection between arteries and veins, while they are normally linked by capillaries. UAVMs are considered as rare, and the real incidence is unknown: less than 1000 cases are described in literature [2-4]. UAVM can be congenital or acquired. The acquired form often follows a spontaneous abortion with subsequent dilation and curettage, an interruption of pregnancy, an endometrial carcinoma, or a gestational trophoblastic disease [5-6]. Generally, women present with severe and life-threatening vaginal bleeding despite the onset of medical therapy [7]. Explorative laparotomy or total hysterectomy were once the only possible way to diagnose UAVM, upon pathological examination. After that, angiography became the gold standard for diagnosis. Lately, ultrasonography with color Doppler has been widely used to obtain a reliable diagnosis [8-10]. Treatments for these conditions are far from being standardized and range between expectation management mainly based on medical therapy, to invasive approaches, such as conservative surgery (uterine arterial embolization, dilatation and curettage, operative hysteroscopy, uterine arteries ligation) or demolitive surgery (hysterectomy) [11]. The treatment of choice depends on various factors such as pregnancy desire and patients' haemodynamic parameters [12]. There have been several reports of pregnancy after conservative management of UAVMs, that seems to be feasible and reassuring on selected patients [13, 14]. In this paper, we report a successful case of a UAVM conservative management with combined temporary UAE plus hysteroscopic resection and we perform a retrospective systematic review of the available literature with regard to the conservative surgical treatment of acquired UAVMs and the fertility outcomes.

2. Methods

2.1 Search strategy

The data research was conducted using the following databases MEDLINE, EMBASE, Web of Sciences, Scopus, ClinicalTrial.gov, OVID and Cochrane Library querying for all articles related to temporary uterine artery embolization, hysteroscopy and UAVM. We used a combination of the following Medical Subjects Heading (MeSH): ((uterine) OR pelvic) AND arteriovenous AND (malformation OR fistula OR

cirsoid aneurysm) AND (curettage OR D and C OR surgical treatment OR hysteroscopy OR uterine artery embolization) AND (bleeding OR hemorrhage). A systematic review was therefore performed following the Preferred Reporting Items for Systematic reviews and Meta-Analyses "PRISMA" guidelines [15]. We have selected articles published from inception to June 2019. We screened those record titles and abstracts retrieved from our searches. All full-text manuscript reference lists were analyzed in order to find additional eligible studies. This process was performed collaboratively by 2 authors (IR, SM).

2.2 Eligibility criteria

Studies with patients with acquired UAVMs were considered for the inclusion. Case reports were included in the selection. No language restrictions were initially applied. To avoid duplication, studies describing the same study population were included only once. The inclusion criteria were as follows: papers referring to acquired UAVMs treated by conservative surgical management (UAE, D & C, hysteroscopy, uterine ligature). Papers with congenital non acquired AVMs were excluded from the review as well as article not in English language, conference papers.reviews, and studies with information overlapping another publication. Also, we excluded studies reporting medical treatments and/or demolitive surgery (hysterectomy) as first line treatment.

2.3 Data extraction

From each eligible paper, we extracted and tabulated the following clinical data: author's name and year; age; gravidity and parity; symptoms at the diagnosis, previous uterine surgical treatments; surgical management, time of follow-up, rate of failed procedures and pregnancy outcomes. Since most of the papers were case reports, we did not conduct a meta-analysis of these studies. The flow chart of the search strategy and the selection process is shown in Figure 1.

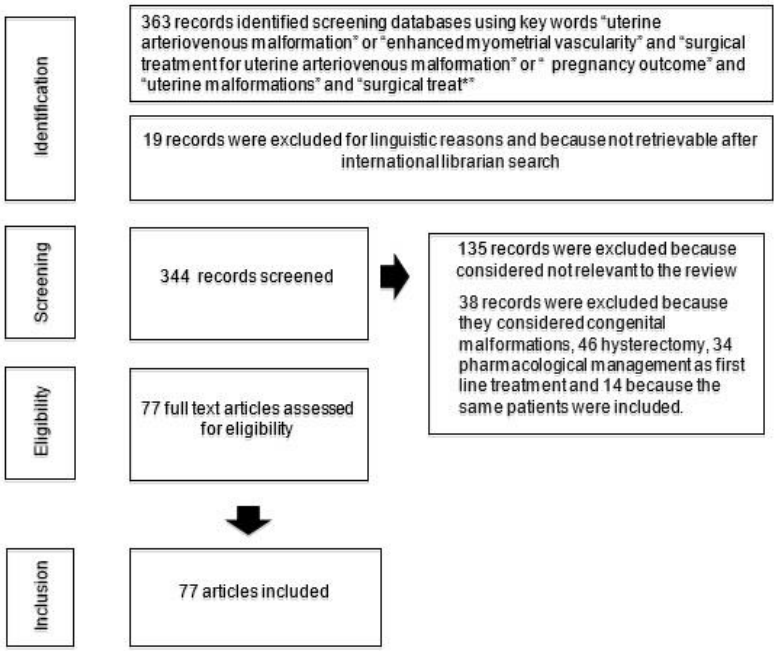

Figure 1: Study selection process.

3. Results

Our electronic database search revealed 363 articles of which 135 were excluded because they were after all, not relevant for the review, 19 records for linguistic reasons, 38 records because they included congenital AVMs, 46 records because they included demolitive surgery, 34 records because they included pharmacological management as first line treatment and 14 records because the same patients were included in latest records. Overall, 77 articles were incorporated for further assessment reporting 125 patients diagnosed with UAVM. The summary of all the papers is outlined in Table 1.

3.1 Patients characteristics

All patients were symptomatic and presented with acute abnormal vaginal bleeding, which was defined as severe. Mean age at diagnosis was 29.38 years, standard deviation (SD) was +/- 6.81 years (range 16-51). Among considered population, mean gravity and parity at the moment of the AVM diagnosis was respectively 2.34 years, SD+/- 1.68 years (range 0-8) and 1.02 years, SD +/- 1.10 years (range 0-5). Ninety-two patients (73.6%) presented with vaginal bleeding post interruption of pregnancy or spontaneous miscarriage. Twelve patients were (9.6%) diagnosed shortly after vaginal delivery and 4 (3.2%) after cesarean delivery; 1 patient (0.8%) presented after a diagnosis of chorioamnionitis. For 16 (12.8%) patients the cause of the UAVM remained unknown. Among all patients ninety-four patients (74%) underwent D&C before the occurrence of the UMAV.

3.2 Management and failed procedures

In terms of management, our search of current literature identified 85 patients that primarily underwent uterine artery embolization (69,6%) and this was the most common treatment option. Eighteen (14.4%) patients were treated with operative hysteroscopy. Five (4%) patients primarily had laparoscopic uterine artery ligation. Thirteen (10.4%) patients underwent D&C. Combined management was the treatment of choice for 2 (1.6%) patients. Of them, one (0.8 %) underwent UAE plus D&C, one (0.8%) had UAE plus operative hysteroscopy.

3.3 Failed procedures

Overall, failed procedures were 21 (16.8%): UAE was insufficient in 17 cases (19.5%). Among these patients fifteen (12%) required a second procedure and six (4.8%) had total abdominal hysterectomy performed due to uncontrolled bleeding (all among the UAE group). Of the patients treated by D&C and uterine ligation, 3 (2.4%) and 1 (0.8%), respectively, had a recurrence and required a second procedure. Two patients treated with combined method had no complications, and the procedures were successful.

3.4 Fertility outcomes

Follow-up data were available for 89 (71.2%) patients, with a mean follow-up of 22 months with SD +/- 11 (range 5-36). Follow-up data is described in table 2. Forty-five (50.6%) conceptions were observed, considering a positive blood beta-hcg. Delivery rate was 82.2% (37 patients). Four (10%) patients had a miscarriage. Four patients were lost to follow-up after positive b-hcg. Twenty-six patients had a term vaginal delivery. Six patients had a term caesarean section. Six patients reported obstetric complications such as pre-eclampsia, IUGR and pPROM. Twenty-eight patients with positive blood beta-hcg were reported after UAE.

	Year	Author	Article Type	N°	Pt N°	Age	G	P	A	Cause
1	1982	Forssman[1]	case report	1		20	2	0	1	D&C
2	1986	Brown[2]	case report	1		21	2	0	1	D&C
3	1986	Markoff[3]	case report	1		27	5	1	1	D&C
4	1987	Poppe[4]	case report	1		25	0	0	0	NS
5	1994	Ginsberg[5]	case report	1		42	2	1	1	D&C
6	1994	Manolitsas[6]	case report	1		21	3	1	1	D&C
7	1995	Chow[7]	case report	1		20	2	0	1	D&C
8	1996	Gaens[8]	case report	1		27	6	1	1	D&C
9	1997	Palmeri[9]	case report	1		16	1	0	1	D&C
10	1997	Hoffman[10]	case series	2						
					1	24	2	0	1	D&C
					2	21	1	0	1	D&C
11	1997	Arredondo-Soberon[11]	case report	1		33	3	0	1	NS
12	2001	Elia[12]	case report	1		29	6	3	1	D&C
13	2002	Kwon[13]	case report	1		26	2	1	1	D&C
14	2003	Gopal[14]	case report	1		42	6	0	1	D&C
15	2003	Takeuchi[15]	case report	1		29	3	0	1	D&C
16	2003	Chia[16]	case report	1		37	3	2	1	D&C
17	2003	Clarke[17]	case report	1		29	4	1	1	D&C
18	2003	Kelly[18]	case report	1		36	1	1	0	D&C
19	2003	Pope[19]	case report	1		19	2	1	1	D&C
20	2003	Chan[20]	case report	1		33	3	2	1	D&C
21	2003	Chen[21]	case report	1		51	3	2	1	D&C
22	2004	Winsor[22]	case report	1		30	1	0	1	D&C
23	2004	Batch[23]	case report	1		28	1	1	0	VD
24	2004	Irikoma[24]	case report	1		28	1	0	1	D&C
25	2004	Neto[25]	case report	1		21	1	0	1	D&C
26	2004	Aziz[26]	case report	1		30	1	0	1	D&C
27	2004	Amagada[27]	case report	1		17	1	0	1	D&C
28	2004	Nikolopoulous[28]	case report	1		39	1	0	1	D&C
29	2005	Ahn[29]	case report	1		37	3	1	1	D&C
30	2005	Ghi[30]	case report	1		29	1	1	0	VD
31	2006	Mccormick[31]	case report	1		21	0	0	0	NS

32	2006	Yahi-Mounta-sser[32]	case report	1		31	7	3	1	D&C
33	2006	Morikawa[33]	case report	1		39	8	3	1	D&C
34	2006	Phoon[34]	case report	1		31	1	0	1	D&C
35	2006	Delotte[35]	case report	1		33	2	1	1	D&C
36	2007	Lin[36]	case report	1		28	4	3	1	D&C
37	2007	Halperin[37]	case series	3						
					1	27	2	1	1	D&C
					2	24	3	0	1	D&C
					3	28	4	1	1	D&C
38	2007	Rangarajan[38]	case report	1		22	2	0	1	D&C
39	2008	Bae[39]	case report	1		35	NS	NS	NS	NS
40	2008	Bagga[40]	case report	1		24	2	1	1	D&C
41	2008	Dar[41]	case report	1		28	1	1	0	
42	2008	Bamberg[42]	case report	1		31	0	0	0	
43	2008	Singh[43]	case report	1		21	2	1	1	D&C
44	2008	Maldonado[44]	case report	1		21			1	D&C
45	2009	Griffin[45]	case report	1		25	1	1	0	
46	2009	Taylor[46]	case report	1		37	3	1	1	D&C
47	2009	Vilos[47]	case report	1		19	1	0	1	D&C
48	2009	Wijesekera[48]	case report	1		27	1	0	1	D&C
49	2010	Guo[49]	case report	1		18	0	0	0	
50	2010	Kim[50]	case series	2						
						35	4	2	1	D&C
						20	1	0	1	D&C
51	2010	Wani[51]	case report	1		37				
52	2010	Przybojews-ki[52]	case report	1		21				
53	2011	Shaba[53]	case report	2						
					1	30		1	1	D&C
					2	45	4	0	1	D&C
54	2011	Vaknin[54]	observational	16						
					1	27	2	1	1	D&C
					2	24	3	0	1	D&C
					3	27	1	1	0	CS
					4	28	4	1	1	D&C

					5	35	6	5	1	D&C
					6	20	3	1	0	CS
					7	36	5	4	0	VD
					8	36	5	2	1	D&C
					9	20	2	0	1	D&C
					10	29	6	3	1	D&C
					11	32	1	1	0	CS
					12	31	4	3	1	D&C
					13	29	3	2	0	CS
					14	41	4	3	1	D&C
					15	39	8	3	1	D&C
					16	31	1	0	1	D&C
55	2012	Scioscia[55]	case report	1	/	36	1	0	1	D&C
56	2012	Clarke[56]	case report	1		23	1	0	1	D&C
57	2012	Levy-Zauber-mann[57]	case report	1		29	3	2	0	D&C
58	2013	Chittawar[58]	case report	1		36	3	2	0	D&C
59	2013	Sharma[59]	case report	1		25	2	1	1	VD
60	2014	Woodhams[60]	case series	2						
					1	33			1	D&C
					2	30	1	1	0	Chorioamniotitis
61	2015	Aslan[61]	case report	1		27	2	1	1	D&C
62	2015	Vilos[62]	case series	5						
					1	19	1	0	1	D&C
					2	32	3	1	1	D&C
					3	30	1	0	1	D&C
					4	18	1	1	0	VD
					5	32	2	2		
63	2015	Tullius[63]	case report	1		37				D&C
64	2016	Calzolari[64]	retrospective	11						
					1	18	1	0	1	D&C
					2	28	2	0	1	D&C
					3	37	2	1	1	D&C
					4	23	2	1	1	D&C
					5	44	2	1	1	D&C
					6	26	2	1	1	D&C

					7	30	1	0	1	D&C
					8	34	2	1	1	D&C
					9	21	1	0	1	D&C
					10	32	2	1	1	D&C
					11	31	1	1	0	VD
65	2017	Bandura[65]	case series	14						
					1	25	1	1	0	VD
					2	40	3	2	1	D&C
					3	39	1	1	0	VD
					4	25	2	1	0	VD/D&C
					5	31	1	1	0	VD
					6	30	2	2	0	VD/D&C
					7	28	5	4	1	D&C
					8	34	3	2	1	D&C
					9	35	2	1	1	D&C
					10	36	3	0	1	D&C
					11	34	5	3	1	D&C
					12	25	2	0	1	D&C
					13	31	1	0	1	D&C
					14	27	1	0	1	D&C
66	2017	Lehrman[66]	case report	1		23	3	0	1	D&C
67	2017	Evans[67]	case report	1		33				
68	2017	Ho[68]	case report	3						
					1	22	3	3	1	
					2	38	0	3		
					3	34	1	0	1	
69	2018	Wee[69]		1		22	1	1	0	VD
70	2018	El agwany[70]	case report	1		38	3	3	0	CS
71	2018	Bettaiah[71]	case report	1		22	2	1	1	D&C
72	2018	Schwartz[72]	case report	1		37				
73	2018	Kaouther[73]	case report	1		28	4	2	1	RPOC (retained product of conception)
74	2019	Guida[74]	case report	1		28	2	0	1	
75	2019	Da joung[75]	case report	1		37			1	D&C
76	2019	Kim[76]	case report	1		20	1	0	1	D&C

77	2019	Khan[77]	case report	1		30	1	0	1	D&C

Table 1: Demographics.

N°	Diagnostic Method	Treatment	Successful	Complications	Follow Up	Conservative	Pregnancy	Outcome
1	Angiography	UAE	YES	NO	22	YES	YES	Elective CS at term
2	US/Angiography	UAE	YES	Rec (1m)	12	YES	NS	
3	Angiography	UAE	YES	Rec (1m)	12	YES	NS	
4	Angiography	UAE	YES	Rec (1m)	6	YES	YES	VD at 35W
5	US/Angiography	UAE	YES	NO	6	YES	NS	
6	US/Angiography/ Hysteroscopy	UAE	YES	NO	10	YES	NS	
7	Angiography	UAE	YES	NO	6	YES	YES	TVD
8	Angiography	UAE	YES	NO	3	YES	YES	NS
9	US/Angiography	UAE	PARTIAL	Rec (12m), TV excision.	12	YES		
10	US/Angiography	UAE	NO	HYSTERECTOMY	NS	NO		
	US/Angiography	UAE	YES	NO	NS	YES	NS	
11	US/Angiography	UAE/metroplasty	YES	NO	12	YES	YES	TVD
12	US/Angiography/ MRI	UAE	YES	NO	5	YES	NS	
13	US/MRI	UAE	YES	NO	NS	YES	YES	TVD
14	US/Angiography/ MRI	UAE	YES	NO	8	YES	YES	IVF/TWIN pregnancy/TCS
15	US	UAE/danazol	PARTIAL	NO	16	YES	NS	
16	US/Angiography	UAE	YES	NO	14	YES	NO	
17	US	UAE	YES	NO	NO	YES		
18	US/Angiography	UAE	YES	NO	1W	YES	NS	

19	US/Angiography	UAE	YES	NO	2	YES	NO	
20	US/Angiography	UAE	YES	NO	NS	YES	NS	
21	US/Angiography/ MRI	Ligation of UA	YES	NO	14	YES	NO	
22	US/Angiography/ MRI	UAE	PARTIAL	Re-embolization	YES	YES	YES	NS
23	US/Angiography/ MRI	UAE	YES	NO	3	YES	NO	
24	US/Angiography/ MRI	UAE	YES	NO	NS	YES	NS	
25	US/Angiography	UAE	YES	NO	NS	YES	NS	
26	US/Angiography	UAE	YES	NO	6W	YES	NO	
27	US/Angiography	UAE	YES	NO	24	YES	YES	TVD
28	US/Angiography/ Hysteroscopy/LPS	Laparoscopic excision	YES	NO	14	YES	YES	TCS
29	US/Angiography/ MRI	UAE	NO	HYSTERECTOMY	12	NO		
30	US/Angiography	UAE	YES	NO	4	YES	NO	
31	US/MRI	UAE	YES	NO	NS	YES	YES	TVD
32	US/Angiography/ MRI	UAE	YES	NO	4	YES	NO	
33	US/Angiography	UAE	YES	NO	12	YES	NO	
34	US/Angiography	UAE	YES	NO	12	YES	YES	Miscarriage+HR (bleeding)
35	US/Angiography	UAE	YES	NO	20	YES	YES	TVD
36	US/Angiography	UAE	YES	NO	8	YES	NS	
37	US/Angiography	Ligation of UA	YES	NO	NS	YES	NS	
	US/Angiography	UAE	YES	NO	NS	YES	YES	NS
	US/Angiography	UAE	YES	NO	NS	YES	NS	
38	US/Angiography/ MRI	UAE	YES	NO	NS	YES	NS	
39	US/Angiography	UAE	YES	NO	29	YES	NO	
40	US/Angiography	UAE	NO	HYSTERECTOMY	6	NO		

41	US/Angiography	suction/curettage	YES	NO	42	YES	NO	
42	US/Angiography	UAE	YES	NO	24	yes	NO	
43	US/MRI	UAE	YES	NO	1	YES	NO	
44	US/Angiography/ MRI	UAE	YES	NO	NS	YES	NS	
45	US/Angiography/ Hysteroscopy	UAE	NO	HYSTERECTOMY	NS	NO		
46	US/Hysteroscopy	UAE	YES	NO	NS	YES	NS	
47	US/Angiography	UAE	YES	NO	11	YES	YES	TVD
48	US/Angiography	UAE	YES	NO	NS	YES	NS	
49	US/Angiography	UAE	NO	HYSTERECTOMY	NS	NO		
50	US/Angiography	UAE	YES	NO	36	YES	YES	TCS
	US/Angiography	UAE	YES	NO	12	YES	NO	
51	CT scan	UAE	YES	NO	NS	YES	NO	
52	US/MRI	UAE	PARTIAL	Rec (1Y), Surg excision(2Y)	36	YES	NS	
53	US/Angiography	UAE/Progesterone	YES	NO	30	YES		
	US/RM	UAE	YES	MODERATE PELVIC PAIN	12	YES	YES	NORMAL PREGNANCY
54	US	Artery ligation	PARTIAL	NO		YES	NO	
	US	UAE	YES	NO		YES	NO	
	US	UAE	YES	NO		YES	YES	TVD
	US	UAE	YES	NO		YES	NO	
	US	UAE/Hysteroscopy	YES	NO		YES	YES	TVD
	US	Observation	YES	NO		YES	YES	TVD
	US	D&C	YES	NO		YES	NO	
	US	UAE	YES	NO		YES	NO	
	US	Observation/MTX	YES	NO		YES	YES	TVD

	US	Hysteroscopy/Guided D&C	YES	NO		YES	NO	
	US	UAE	YES	NO		YES	NO	
	US	UAE	YES	NO		YES	NO	
	US	D&C	YES	NO		YES	NO	
	US	UAE	YES	NO		YES	NO	
	US	UAE	YES	NO		YES	NO	
	US	D&C/MTX	YES	NO		YES	YES	TVD
55	US	Hysteroscopy	YES	NO		YES	NS	
56	US	UAE	PARTIAL	NO	NO		NS	
57	Angiography	UAE	PARTIAL	LPS iliac artery occlusion	1	YES		
58	Hysteroscopy/Angiography	UAE	YES	NO		YES	NS	
59	US/Angiography	UAE	YES	NO	NO		NO	
60	US/CT	UAE	YES	NO	6	YES	NS	
	US/MRI	UAE	YES	NO	6	YES	NS	
61	US	UAE	YES	NO	ns		NO	
62	US	UAE	YES	NO	12	YES	YES	TVD, no complications
	US	UAE	YES	NO	24	YES	YES	Cerclage, PPROM at 34w
	US	UAE	PARTIAL	re-embolization	14	YES	YES	pre-eclampsia, VD at 34w, recurrence of AVM
	US	UAE	PARTIAL	re-embolization	36	YES	YES	IUGR, VD at 36w
	US	UAE	PARTIAL	re-embolization	9	YES	YES	TVD, no complications

63	US	UAE	YES	NO	3	YES	NO	
64	US	Hysteroscopy	YES	NO	24	YES	NO	
	US	Hysteroscopy	YES	NO	6	YES	NO	
	US	Hysteroscopy	YES	NO	24	YES	YES	Miscarriage I trimester
	US	Hysteroscopy	YES	NO	22	YES	YES	TVD
	US	Hysteroscopy	YES	NO	20	YES	NO	
	US	Hysteroscopy	YES	NO	26	YES	NO	
	US	Hysteroscopy	YES	NO	28	YES	YES	TVD
	US	Hysteroscopy	YES	NO	30	YES	NO	
	US	Hysteroscopy	YES	NO	29	YES	YES	TVD
	US	Hysteroscopy	YES	NO	4	YES	YES	TVD
	US	Hysteroscopy	YES	NO	14	YES	NO	
65	US	Glue injection/Hysteroscopy	YES	NO	55	YES	YES	TVD
	US	Glue injection/D&C	YES	NO	55	YES	YES	TVD
	US	Glue injection/Hysteroscopy	YES	NO	55	YES	YES	TVD
	US	Glue injection/Hysteroscopy	YES	NO	55	YES	YES	TVD
	US	Glue injection/D&C	NO	Recurrence	55	YES	NO	

	US	Glue injection/Hysteroscopy	YES	NO	55	YES	YES	Miscarriage
	US	Glue injection/D&C	NO	Recurrence	55	YES	YES	Miscarriage
	US	Glue injection/D&C	YES	NO	55	YES	NO	
	US	Glue injection/D&C	YES	NO	55	YES	NO	
	US	Glue injection/D&C	YES	NO	55	YES	YES	Twin pregnancy, term CS
	US	Glue injection/D&C	YES	NO	55	YES	NO	
	US	Glue injection/D&C	YES	NO	55	YES	YES	TVD
	US	Glue injection/D&C	YES	NO	55	YES	YES	TVD
	US	Glue injection/D&C	NO	Recurrence	55	YES	NO	
66	US	UAE	PARTIAL	Rec (2m)/ transvag excision	17	YES	NO	
67	US/Angiography	UAE	YES	NO	17	YES	NO	
68	US/Angiography	UAE	YES	anovulatory cycles (3m)	36	YES (TWICE)	YES	(1st preg) TVD/ (2nd preg) IUGR+Pp Haemorrhage

	US/Angiography	UAE	YES	transitory ischemic pain	24	YES	YES	TVD
	US/Angiography	UAE	PARTIAL	Recurrence (3m)	24	YES	YES	SGA/Oligohydramnios/preterm birth
69	US/Angiography	UAE	YES	NO	2	YES	NS	
70	US/MRI	UAE	YES	NO	1	YES	NS	
71	US/Hysteroscopy	Ligation of UA		NO	6	YES	NS	
72	Hysteroscopy	UAE	NO	HYSTERECTOMY		NO	NS	
73	US/Angiography	UAE	YES	Re-embolization	8	YES	YES	NS
74	US/CT	UAE	YES	NO	5	YES	YES	normal pregnancy
75	US/Angiography	UAE/D&C	YES	NO	2	YES	YES	CS at 37w
76	US/MRI	Hysteroscopy	YES	NO	12	YES	NO	
77	US/MRI	UAE	YES	NO	6	YES	NS	

Table 2: Diagnosis, management and obstetric outcomes.

4. Case Report

A 33-year-old woman (IG 0P) with no relevant clinical history was referred in February 2019 to our gynaecology department after a miscarriage at 21 weeks of gestation, treated by evacuation with aspiration and curettage. After persistent uterine bleeding she started therapy with acetate norethisterone and tranexamic acid. In april 2019 she was admitted to our department due to secondary anemia requiring blood transfusion. The level of haemoglobin concentration decreased from 12.7 gr/dL to 7.2 gr/dL. Transvaginal ultrasonographic evaluation (2D and 3D scan) showed an anteverted uterus of 82x41x57mm and a thickened endometrium of 16mm. The uterine cavity was occupied by a hypervascular region on the posterior uterine wall of 21x16 mm, colour score 4, with a PSV > 100 cm/s (Figure 2). The b-hcg value was negative. Ovaries and cervix were normal. Based on clinical and imaging findings, a uterine arteriovenous malformation (UAVM) was considered. After multidisciplinary agreement, the patient underwent a computed tomography angiography (CTA) that confirmed the presence of a mass located between the endometrium and the myometrium with contrast enhancement (intense captation during arterial

phase and early venous drainage) confirming the suspect of UAVM (Figure 3). The procedure was followed by a right uterine artery embolization with reabsorbable agent (Spongostan) (Figure 4) to decrease the ongoing vaginal bleeding and reduce the possible successive risk of intraoperative blood loss. Access through the right common femoral artery was performed, permitting the selective catheterization of hypogastric artery and of the right uterine artery. Spongostan was used to perform temporary UAE also in order to protect the ovary from permanent ischemic damage. There were no post-operative complications. After 24 hours from the UAE a second transvaginal ultrasound showed absence of hematic flow after the UAE . The uterine cavity was still enlarged by clots and the presence of hyperechoic lesion of mm13x14 (Figure 5). In the same day the patient underwent the hysteroscopic treatment. Hysteroscopy was performed with vaginoscopic approach, with a 5-mm diameter continuous-flow hysteroscope with oval profile, a 30° fore-oblique telescope and a 5 Fr operating channel (Office Continuous Flow Operative Hysteroscopy 'size 5'; Karl Storz, Tuttlingen, Germany). Saline solution (NaCl 0.9%) was used as distension medium, which was provided through an electronic system of irrigation and aspiration (Endomat; Karl Storz, Tuttlingen, Germany). A stable intrauterine pressure of about 40 mmHg was obtained. The hysteroscopic view showed the uterine cavity filled with haemorrhagic material as well as parenchymatous material. The surgeon decided to undergo hysteroscopic resection using a bipolar 15 Frresectoscope (bipolar 15Fr office resectoscope; Karl Storz, Tuttlingen, Germany). The procedure was safe and successful (Figure 6). No post-operative complications were reported and resulted in a complete absence of symptoms. One month after surgery a normal menstrual cycle was restored and the ultrasonography showed a restoration of right uterine artery flow (Figure 7).

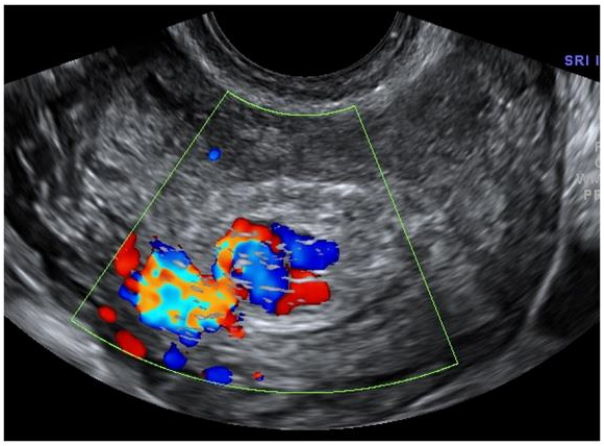

Figure 2: Hypervascular lesion into the uterine cavity.

Figure 3: Computed tomography angiography (CTA).

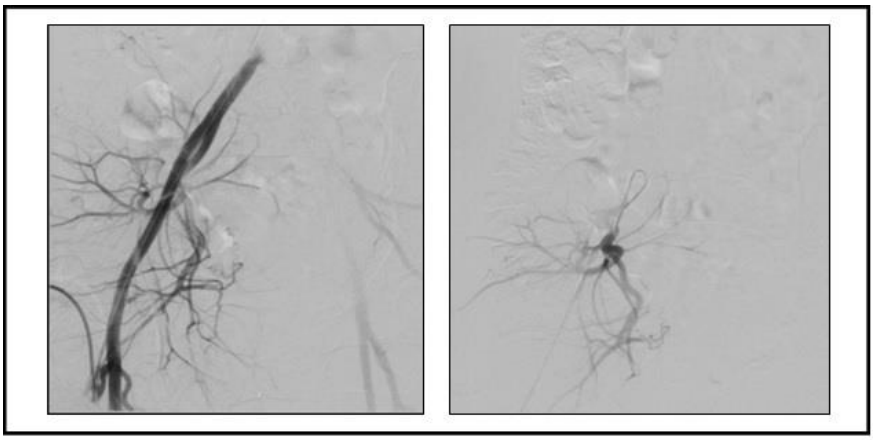

Figure 4: Selective embolization of right uterine artery with Spongostan.

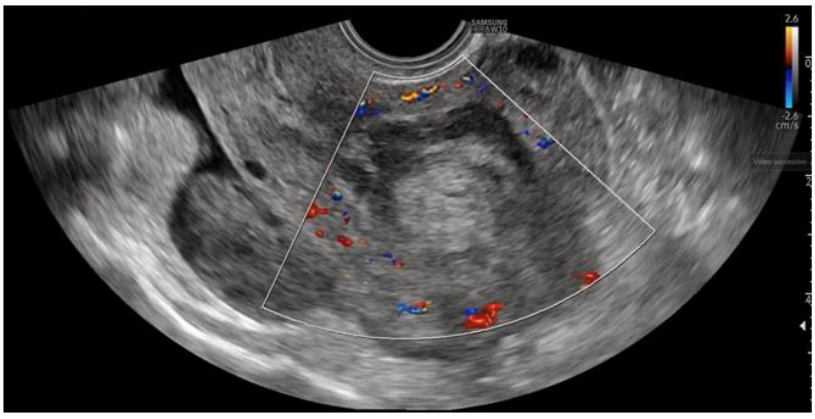

Figure 5: US view of hyperechoic lesion after temporary UAE.

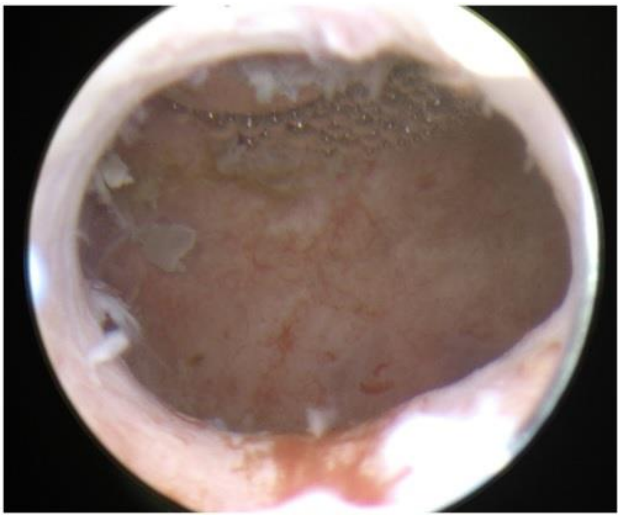

Figure 6: Uterine cavity after procedure.

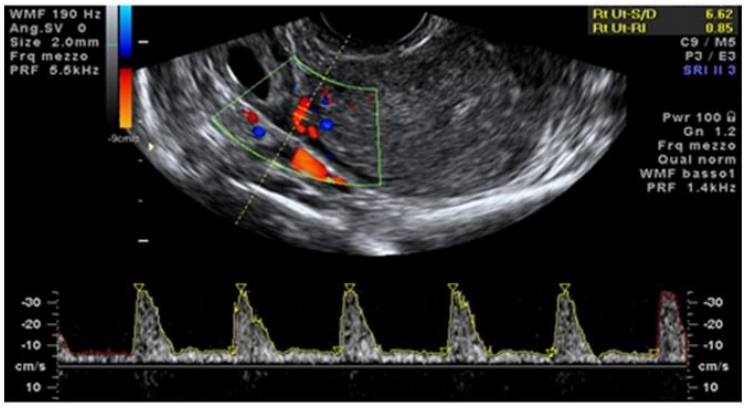

Figure 7: Restoration of right uterine artery flow.

5. Discussion

UAVM is a very rare condition but potentially life-threating. This paper described a case of UAVM treated by temporary UAE plus hysteroscopy and provided a systematic review of current available literature on UAVM conservative surgical management. UAVMs often occur in woman who previously underwent D&C, probably because the intrauterine trauma is involved in the pathogenesis of the disease [16]. This review confirmed this trend, as we found 74.4% patients with a acquired UMAV diagnosis after IVG o D&C for spontaneous miscarriage. Until not so long ago, the gold standard treatment was hysterectomy [17]. However, such a demolitive management is no longer acceptable, especially with stable hemodynamic conditions in young women. Among the conservative management, UAE was the most used but literature showed that UAE treatment alone is often not successful [18]. In fact, our series presented 19.5% failed UAE that required either further procedure. Among these patients, 6.9% had heavy recurrence of bleeding that lead to hysterectomy. Only 2 cases of combined surgical strategy were

reported in available literature (UAE plus D&C and UAE plus hysteroscopy) in cases of UAVMs [54,75]. To our knowledge, this case report is the first to describe a combined method with temporary UAE and hysteroscopy. In fact, in our case, the UAE was performed with Spongostan that has an absorption time of only 15 days. This permitted a safe hysteroscopic treatment avoiding in the same time the subsequent ischemic damage. The use of UAE with no resorbable agent reduce definitively the blood flow to the uterus. Despite the presence of collateral vessels the reduction of the flow could create problems in subsequent pregnancies [20]. Moreover, it is still reported an increased incidence of miscarriage and stillbirth in pregnancy after UAVM embolization [20], which could be explained by a reduction of the uterine flow. Using of resorbable agent for the UAE could avoid this consequence. In any case there have been several case studies of pregnancy after conservative management of UAVMs. In fact, our review shows 45 cases of pregnancies, most of them after UAE treatment. Although we performed a systematic review our paper suffers of the lack of RCTs. Most papers are case reports and this does not allow a methodological analysis of the results. After all, we share a rare UAVM management with a combined method (UAE plus hysteroscopy) and for the first time the UAE is performed with a resorbable agent. In the same fashion the systematic review is the first one that report data only about conservative surgery.

6. Conclusions

In conclusion, UAVMs are very rare life-threatening lesions. There is not a gold standard of treatment. Conservative surgery management should be proposed to young women with pregnancy desire in stable hemodynamical conditions. Using a resorbable agent to perform UAE permitted a normal restoration of the flow in the uterine artery after the hysteroscopic resection but it needs RCT to have solid results about surgical and fertility outcomes.

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Nothing to declare

Disclosure

Nothing to declare

Conflict of Interest

The authors declare that they have no conflict of interest.

Ethical Approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent

Informed consent was obtained from all individual participants included in the study.

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