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The Importance of Ultrasound in the Differential Diagnosis of Acquired Arterio-Venous Malformation and of Retained Products of Conception: Two Similar Cases with Distinct Etiology and Management

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Abstract

Retained Products of Conception (RPOC) and Arteriovenous Malformations (AVM) are relevant causes of Postpartum Hemorrhage (PPH) which can be potentially life-threatening when treatment is deferred or when surgically mistreated due to wrong differential diagnosis.

Ultrasound (US) has a key role in the diagnosis of both entities, and can be useful when planning the therapeutic approach and assess the risk of severe bleeding. The presented case-reports illustrate both entities and the difficulties faced in its differential diagnosis, as well as the utility of US on their detection and management. On the first case, patient presented the typical clinical features and risk factors of AVM. The US findings included the classical highly-specific signs: Multiple hypoechoic myometrial spaces with multidirectional turbulent high velocity flow seen on color Doppler US, and prominent parametrial vessels. On the second case, the clinical presentation and the background of curettages prior to the current pregnancy led the medical team to initially suspect AVM, even when the patient did present the three sonographic signs with higher PPV for RPOC diagnosis: A well-defined endometrial mass, a thickened endometrium, and endometrial vascularization. Key point on US differential diagnosis between the two entities is the presence of endometrial vascularization *vs.* exclusively myometrial vascularization.

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Introduction

Early or late PPH are common complications in the obstetric wards worldwide. The differential diagnosis for early PPH –which happens within the first 24 h after delivery– includes uterine atony, lacerations, coagulopathies and RPOC; while for late or secondary PPH –which happens between 1 day and 12 weeks after delivery– RPOC, endometritis, subinvolution of the placental implantation site, coagulopathies and AVM should be taken into consideration [1,2].

RPOC are intrauterine tissues developed after conception which persist after birth, miscarriage or pregnancy termination. The anatomopathological diagnosis is based on the presence of chorionic villi. Incidence of RPOC is remarkable (about 17% during the 1st term, 40% during the 2nd term and 3% during 3rd term deliveries, with an estimated incidence of 3% to 5% for on-term vaginal births) [3]. The most prevalent risk factors are previous retained placenta, maternal age, previous dilation and curettage, caesarean section or uterine surgery, labor induction, pre-term birth, uterine malformation and placental features such as velamentous cord insertion or placental implantation site [4]. Complications include PPH, endometritis, and infertility related to the formation of intrauterine adherences and scar tissue [1,5]. There is a lack of a formal consensus on diagnostic sonographic or imaging criteria regarding RPOC, although US has a key role on its detection and management when there is a clinical suspicion. Treatment options range from expectant management to medical-induced expulsion or surgical resection of RPOC (*via* curettage or hysteroscopy) depending on the presence and intensity of hemorrhage, the presence of concomitant infections and the sonographic characteristics of the placental remnants [5,6].

Acquired AVM is an infrequent cause of severe vaginal bleeding, in which uterine tissue damage –mainly caused by uterine instrumentation, or less likely in context of trophoblastic

disease, diethylstilbestrol exposure, neoplasms or infections– cause an abnormal arterio-venous communication without a capillary bed in between [7,8]. Patients present with intermittent or severe vaginal bleeding, sometimes debuting days after birth or uterine instrumentation [9]. The gold standard for AVM diagnosis is angiography, showing early venous filling, with color Doppler US also being a reliable and available diagnostic tool. Active treatment for AVM is Uterine Artery Embolization (UAE) [1,7-9]. Early differential diagnosis between RPOC and AVM may be challenging but it is key since the primary treatment for RPOC presenting with active severe bleeding (surgical resection of the remnants) may fatally worsen the hemorrhage in case of AVM.

Materials and Methods

On April 2020, a 37-year-old nullipara was admitted with Preterm Premature Rupture of Membranes (PPROM) and clinical chorioamnionitis at 17+5w. Antibiotic coverage was initiated.

During the expulsion of the fetus, she presented massive acute vaginal bleeding (1.4 L in 2 min). Patient underwent an emergent echo-guided suction curettage to evacuate the placenta and a Bakri Balloon was placed. Intra-surgery, she required transfusion with 6 blood units and administration of vasoactive drugs to preserve hemodynamic stability. Patient was discharged asymptomatic 3 days after the surgery. She was readmitted 5 days later presenting endometritis: endocervical culture was positive for *E. coli* BLEA. Treatment with Imipenem was started. US showed a thickened endometrium of 69 mm with a solid mass of 28 mm × 27 mm with moderate vascularization on Doppler-US (color score 3), compatible with RPOC (Figure 1).

A second echo-guided curettage was performed. Due to severe intra-surgical bleeding, patient required another Bakry Balloon placement and transfusion with 2 more blood units. After removing the Bakry Balloon, patient persisted asymptomatic and was discharged. Two weeks later she was readmitted in hypovolemic shock, needing 2 more blood units and vasoactive drugs. US showed a thickened avascular endometrium of 34 mm and a heterogeneous and intensely vascularized myometrium, with important dilatation of the arcuate and radial arteries, and a nodular image of 28 mm ×13 mm in its posterior wall, color score 4 with turbulent multidirectional high-velocity flow, suggestive of AVM (Figure 2).

Arteriography was performed, objectivizing hypertrophic uterine arteries with 4 points of arterio-venous fistula. Patient needed 2 consecutive embolization's on the left and on the right uterine arteries within 20 days, both with conical-shape pushable coils, to successfully solve the AVMs. Bleeding receded and patient is currently asymptomatic.

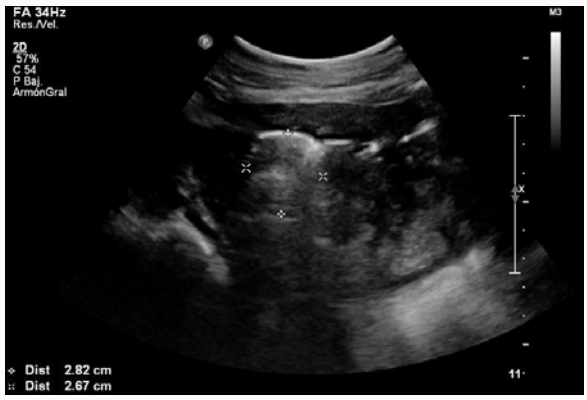

Figure 1: Ultrasound showed a thickened endometrium of 69 mm with a solid mass of 28 mm × 27 mm, color score 3, compatible with RPOC.

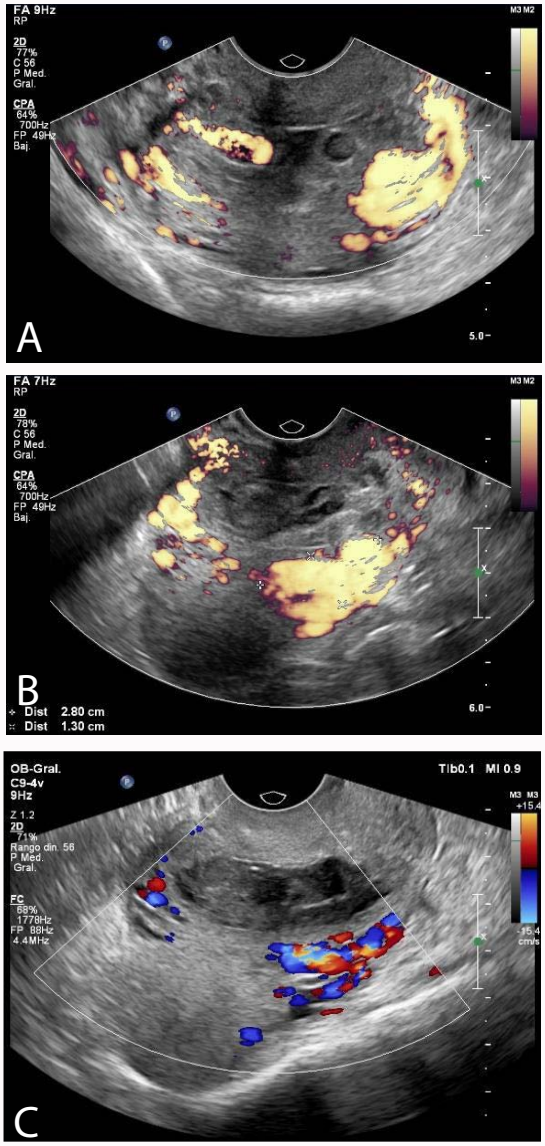

Figure 2: US showed a thickened avascular endometrium of 34 mm and a heterogeneous and intensely vascularized myometrium, with important dilatation of the arcuate and radial arteries, predominantly on the left side (A – transverse plane), and a nodular image of 28 mm × 13 mm in its posterior wall (B – sagittal plane), color score 4, with turbulent systolic and diastolic multidirectional and high-velocity flow (C – sagittal plane), suggestive of AVM.

On the second case, on December 2021, a 35-year-old nulliparous woman with an antecedent of two curettages in context of a therapeutic abortion due to fetal acrania and a posterior transfer of a cryopreserved embryo, presented early and late postpartum hemorrhage after a breech delivery instrumented with forceps at 37+5w, with hemodynamic instability.

First transvaginal US performed in the emergency ward 16 h after delivery showed a thickened avascular endometrium of 40 mm, congruent with clots. An emergency transabdominal echo-guided suction curettage was performed 48 h after birth. During the procedure,

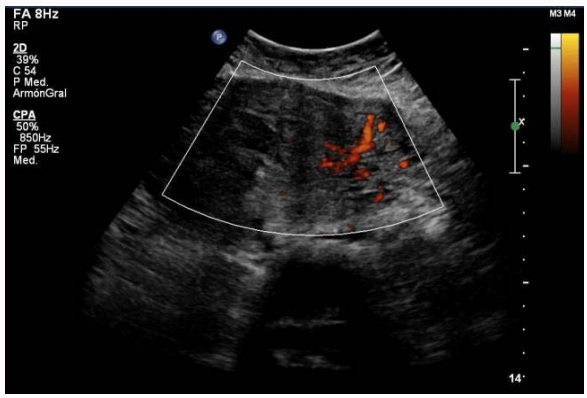

Figure 3: During the echo-guided emergency curettage, a highly vascularized myometrial zone was seen, with pulsatile vascularization and dubious continuity with the endometrial cavity, initially suspected to be an AVM.

a highly vascularized myometrial zone was sonographically detected on the upper third of the uterus, with pulsatile vascularization and dubious continuity with the endometrial cavity, initially suspected to be an AVM (Figure 3).

Due to this diagnosis suspicion and with the patient in progressive anemization (Hb 53 g/L), angio-TC and UAE with resorbable material were performed. Vaginal bleeding receded. During the admission she was transfused with 8 blood units. A week later, a sonohysterography was performed, observing a 26 mm heterogeneous endometrial cavity with a solid heterogeneous formation of 53 mm × 22 mm × 41 mm which appeared to depend on the left posterior-lateral uterine wall, with moderate vascularization on Doppler-US (color score 3), suggestive of RPOC (Figure 4).

A consecutive attempt to evacuate the RPOC was made with a second echo-guided curettage, which resulted unsuccessful due to the intense vascularization of the remnants and the danger of massive bleeding, as well as the fact that they were intensely adhered to the myometrium. As the patient was asymptomatic at the time, she was discharged with a sonographic control scheduled 3 weeks later.

However, a week later, she restarted profuse vaginal bleeding with secondary anemization, once the UAE was no longer effective due to reabsorption of the embolizing material. She needed transfusion with 2 more blood units.

A multidisciplinary committee decided to practice a second UAE –this time with non-resorbable material– followed by a surgical hysteroscopy, in which RPOC intimately attached to the myometrium were completely resected with bipolar loop (Figure 5). The patient is now asymptomatic.

Results and Discussion

On the first case, patient presented the typical clinical features and risk factors of AVM: Massive vaginal bleeding after two consecutive curettages for RPOC. The US findings included the classical highly-specific signs: multiple hypoechoic myometrial spaces with systolic and diastolic multidirectional turbulent high-velocity flow seen on color Doppler, and prominent parametrial vessels [1,7-9].

On the second case, the US image during the first attempt of echo-guided curettage –which showed a highly vascularized myometrial zone with pulsatile vascularization in continuity with the endometrial cavity– as well as the clinical presentation and the background of curettages prior to the current pregnancy, led the medical team to initially suspect AVM. Nevertheless, the patient did present the three sonographic signs with higher PPV for a RPOC diagnosis: A well- defined endometrial mass, a thickened endometrium, and endometrial vascularization [1,10]. In RPOC, endometrial vascularized component is seen, while in AVM the abnormal vascularization is located exclusively in the myometrium. This is the key point on the US differential diagnosis between the two entities.

Besides that, US has a key role on assessing the type and intensity

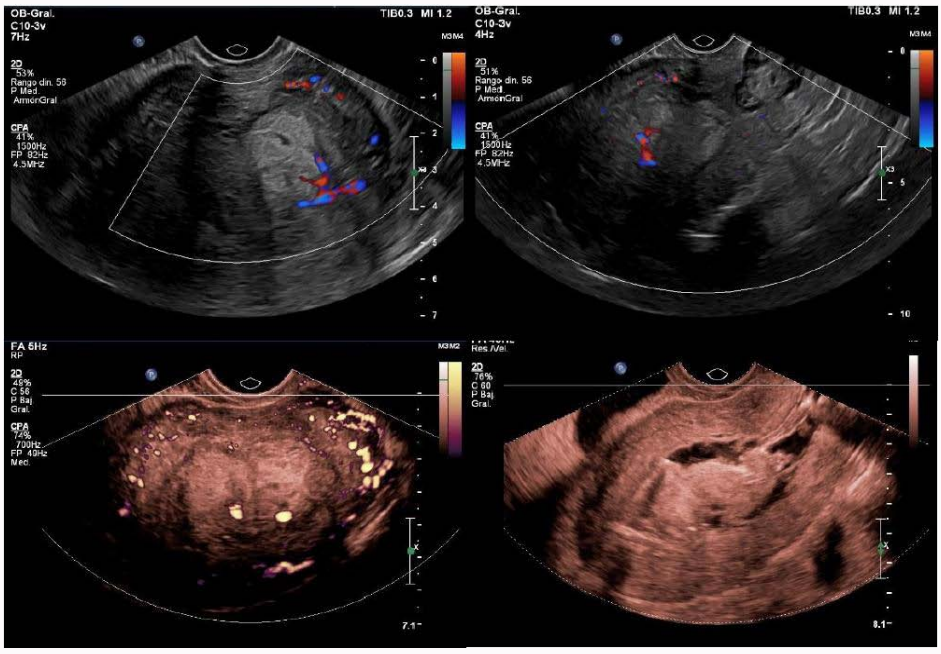

Figure 4: Sonohysterography showed a 26 mm heterogeneous endometrial cavity with a solid heterogeneous mass of 53 mm × 22 mm × 41 mm, color score 3, suggestive of RPOC.

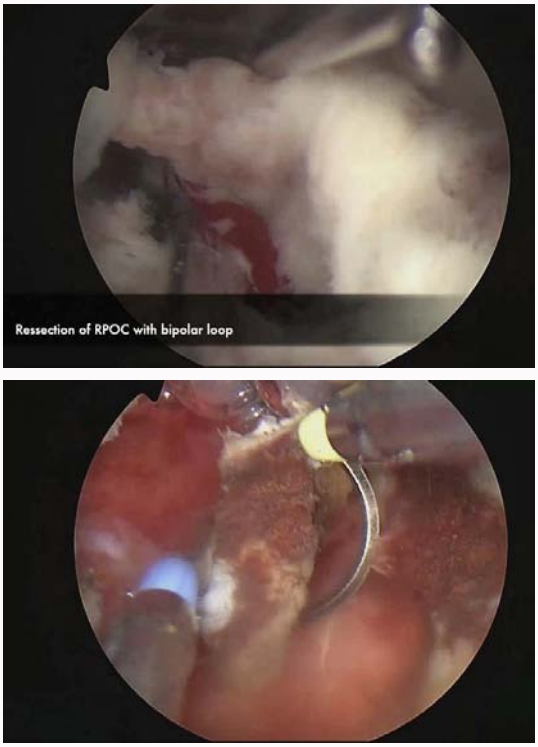

Figure 5: RPOC intimately attached to the myometrium were safely and completely resected with bipolar loop within one single hysteroscopic attempt after UAE.

of the vascularization in RPOC and, consequentially, in anticipating its surgical-treatment associated bleeding risk. In RPOC Kamaya Scale [10] (Figure 6) or Gutenberg Classification [11,12] (Figure 7) types 2 and 3 –as the second case we present, in which endometrial moderate-intense vascularity with equal flow in the endometrium and surrounding myometrial zone was found–, it is worth considering a two-phase treatment: for instance, UAE as a first step to decrease active bleeding and/or risk of intra- surgical bleeding, followed by the final surgical –preferably hysteroscopic– resection of RPOC [6].

Conclusion

Early diagnosis of both RPOC and acquired AVM is key to establishing an appropriate therapeutic plan to quickly solve the situation and to avoid iatrogeny. AVM are frequently over-diagnosed. Therefore, taking the medical antecedents into consideration is useful since AVM are rare in the absence of a previous intra-uterine manipulation and tissular injury. US features for AVM are specific and allow an accurate diagnosis orientation until arteriography is performed. In the case of RPOC, US is not as specific, but it is key on the differential diagnosis, orientating whether the abnormal vascularization is located in the myometrium or in the endometrium. US- obtained information may be also useful when planning the therapeutic approach to RPOC and to assess the risk of intra-surgical bleeding.

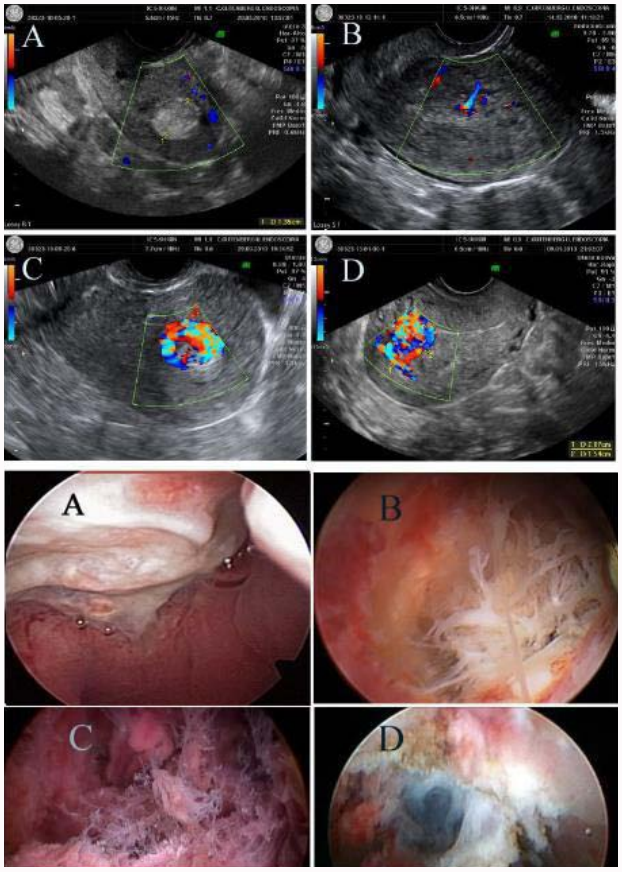

Figure 7: Ultrasonographic and hysteroscopic patterns of RPOC. Gutenberg Classification. A- Type 0: Hyperechogenic avascular mass; white mass with no clear structures. B-Type 1: Different echoes with minimal or no vascularization; well-defined avascular chorionic villi. C- Type 2: Highly vascularized mass confined to the cavity; well vascularized chorionic villi. D- Type 3: Highly vascularized mass with highly vascularized endometrium; aneurism over myometrium in the implantation area [12].

Personal Communications

Part of the work here presented has been presented on a virtual poster to the 2022 ISUOG Congress.

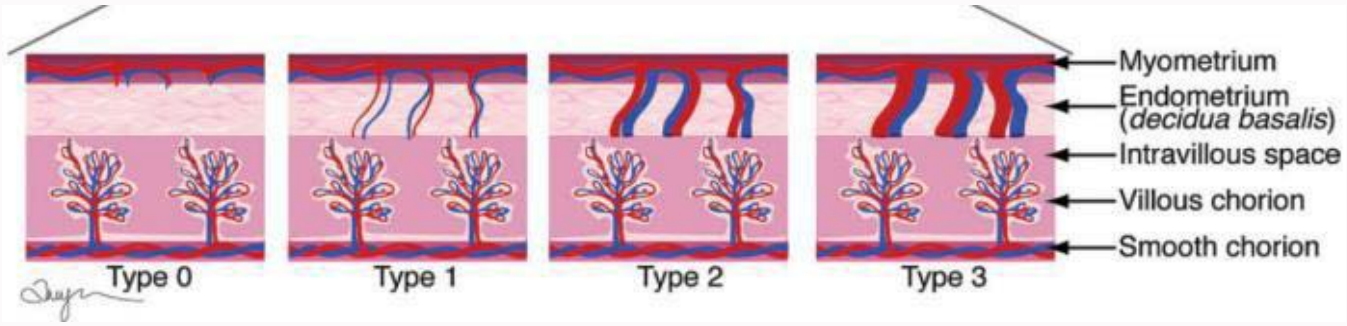

Figure 6: Classification of RPOC vascularity types according to Kamaya Scale [1].

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