
Atypical presentation of placenta accreta spectrum: a case series of spontaneous hemoperitoneum in the third trimester

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Abstract

Background: The indications of emergent termination of pregnancy in placenta accreta spectrum (PAS) include vaginal bleeding, true labor pain or premature rupture of membranes (PROM). Some cases may have atypical presentation as a result of presence of intra-abdominal bleeding rather than vaginal bleeding. Delayed diagnosis of these cases leads to catastrophic complications to both mother and fetus.

Patients and Methods: This was a case series of patients with PAS who underwent emergent delivery due to atypical presentation by hemoperitoneum with absence of any of the classic symptoms that indicate urgent delivery, including vaginal bleeding, true labor pain or PROM. Hemoperitoneum was diagnosed by finding free fluid in the abdomen or pelvis by ultrasonography.

Results: Seven patients were included in this study. The atypical presentation included: 1) shortness of breath; 2) chest tightness; 3) vague abdominal pain and discomfort; 4) shoulder pain; 5) repeated vomiting and sweating; 6) syncope; and 7) cardiac arrest. Laparotomy was performed for all patients and variable amounts of intra-abdominal bleeding were found. The cause of bleeding was either rupture of surface vessel of uterovesical neovascularity or minute focal penetration of the placenta. Cesarean hysterectomy was performed in 5 patients while resection of lower uterine segment with uterine preservation was performed in 2 patients. The newborn was living in 6 patients and dead in one patient who had presented by cardiac arrest.

Conclusion: Any patient with PAS who have atypical presentation must be put under observation with close monitoring of the general condition for signs of hypovolemia, and ultrasonography should be performed by expert sonographer for early detection of hemoperitoneum. Confirmation of the diagnosis indicates emergent laparotomy, better via longitudinal incision, considering cesarean hysterectomy as a first line treatment.

Keywords: PAS, Placenta previa, Placenta accreta, Hemoperitoneum.

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Introduction

There was a dramatic increase in the incidence of placenta accreta spectrum (PAS) over the past several decades as a direct result of increased rates of cesarean delivery (CD), with reported incidence of 1 in 533 deliveries in 2002 (1, 2), compared to 1 in 4000 deliveries in 1970 (3). Massive peripartum bleeding is the real danger and at the same time, the main challenge associated with this problem, as it can lead to multisystem organ failure, disseminated intravascular coagulopathy (DIC), need for intensive care unit (ICU) admission, hysterectomy and even maternal mortality (2, 4-7).

Multiple retrospective cohort studies of PAS disorders have documented that the maternal morbidity was reduced when care was provided in centers of excellence. These centers should provide a dedicated management plan through a multidisciplinary team (MDT) that is experienced in management of such cases (8-11). The components of the team cooperate in order to improve the outcome through providing accurate prenatal diagnosis, dealing with complex pelvic surgery, managing all anesthetic considerations regarding these patients including establishing safe massive transfusion protocols if needed, solving transfusion problems, and finally caring of the patient during postpartum period and managing any complications during this critical period (12).

It was found that emergent delivery of patients with PAS was associated with poorer outcomes and increased maternal morbidity when compared with planned scheduled delivery even when performed in specialized centers by well-trained MDT (13). This had led to controversy about the optimal timing of delivery in PAS, as it should balance the maternal risks of emergent delivery against the neonatal complications of prematurity. The suggested time for delivery is between 34 and 36 weeks of gestation with consideration of earlier delivery in

the presence of recurrent attacks of vaginal bleeding, uterine contractions and premature rupture of membranes (PROM) (12).

Although applying these guidelines had led to reduction in emergent deliveries in PAS, there are still some cases in which the diagnosis of the presence of emergent situation indicating delivery may be difficult and not straightforward due to presence of a typical presentation making these cases easily missed with catastrophic maternal and neonatal complications. Therefore, we aimed in this study to highlight the importance of suspicion, early detection and management of cases of PAS presented with atypical clinical picture.

Patients and Methods

This was a case series of patients with PAS who were presented during the period from September 2018 through August 2023 in Mansoura University Hospital (MUH), Egypt. The study was approved by the Mansoura Faculty of Medicine Institutional Research Board (Code No. R.23.07.2274. R1). The main inclusion criterion was women with PAS who underwent emergent delivery due to atypical presentation by hemoperitoneum with absence of any of the classic symptoms that indicate urgent delivery, including vagina bleeding, true labor pain or PROM. The diagnosis of PAS was made by presence of signs of invasive placentation by ultrasonography in the form of intraplacental lakes, loss of retroplacental hypoechoic line, myometrial thinning, and uterovesical hypervascularity. Hemoperitoneum was diagnosed by finding free fluid in the abdomen or pelvis by ultrasonography.

Results

Seven patients were included in this study. Table 1 presents the demographic and clinical characteristics of the patients. All patients had history of at least one prior CD. Three patients were presented in emergency unite

while the other 4 patients were admitted from the outpatient clinic for elective delivery, but the emergency presentation occurred in the inpatient department. The clinical presentation of hemoperitoneum included: 1) shortness of breath; 2) chest tightness; 3) vague abdominal pain and discomfort; 4) shoulder pain; 5) repeated vomiting and sweating; 6) syncope; and 7) cardiac arrest. All patients presented in the third trimester (32-37 weeks of gestation). The fetus was living in 6 patients before delivery with no signs of fetal compromise. The fetus was dead in the seventh case that arrived to hospital with cardiac arrest. Also, the uterine wall appeared intact with no evident hole as evaluated by ultrasonography just before

delivery.

The operative and postoperative characteristics of the patients were displayed in table 2. Longitudinal abdominal incision was performed in 6 patients and transverse incision was performed in only one patient. Cesarean hysterectomy was performed in 5 patients while resection of the lower uterine segment (LUS) and uterine preservation was performed in 2 patients. All patients needed transfusion of variable amounts of packed red blood cells (RBCs) and fresh frozen plasma (FFP). Only 2 neonates required admission to neonatal care unit (NCU) due to premature delivery at 32-33 weeks and low birth weight below 2500 gm.

Table 1. Demographic and clinical characteristics of the 7 patients

| | Patient 1 | Patient 2 | Patient 3 | Patient 4 | Patient 5 | Patient 6 | Patient 7 |
|--|---------------------------------|---|---------------------------------|-------------------------------------|--|--------------------------------|----------------|
| Age (years) | 30.12 | 31.37 | 26.75 | 25.98 | 30.58 | 29.73 | 25.16 |
| Height (cm) | 162 | 164 | 164 | 167 | 166 | 168 | 165 |
| Weight (kg) | 68 | 66 | 70 | 92 | 79 | 102 | 72 |
| BMI (kg/m²) | 25.91 | 24.54 | 26.03 | 32.99 | 28.67 | 36.14 | 26.45 |
| Gravidity | 4 | 5 | 2 | 4 | 5 | 3 | 4 |
| Parity | 2 | 4 | 1 | 3 | 3 | 2 | 2 |
| Prior cesar-ean delivery | 2 | 4 | 1 | 3 | 3 | 2 | 2 |
| Prior vaginal delivery | 0 | 0 | 0 | 0 | 0 | 0 | 0 |
| Prior miscarriage | 1 | 0 | 0 | 0 | 1 | 0 | 1 |
| Underwent MRI | No | Yes | Yes | No | No | Yes | No |
| Site of presentation | Inpatient department | Inpatient department | Inpatient department | Emergency unit | Emergency unit | Inpatient department | Emergency unit |
| Clinical presentation | Shortness of breath and syncope | Chest tightness and shortness of breath | Shortness of breath and syncope | Vague abdominal pain and discomfort | Chest tightness, shortness of breath and shoulder pain | Repeated vomiting and sweating | Cardiac arrest |
| Gestational age at delivery (weeks) | 32.14 | 37.71 | 32.71 | 35.71 | 37.14 | 36.14 | 32.14 |

Table 2. Operative and postoperative characteristics of the 7 patients

| | Patient 1 | Patient 2 | Patient 3 | Patient 4 | Patient 5 | Patient 6 | Patient 7 |
|--|-----------------------|-----------------------|-----------------------|---|---|-----------------------|------------------------|
| Abdominal incision | Longitudinal | Longitudinal | Longitudinal | Longitudinal | Longitudinal | Transverse | Longitudinal |
| Operative procedure | Cesarean hysterectomy | Cesarean hysterectomy | Cesarean hysterectomy | Resection of LUS and uterine preservation | Resection of LUS and uterine preservation | Cesarean hysterectomy | Cesarean hysterectomy |
| Intraoperative transfusion | | | | | | | |
| <i>Packed RBCs (units)</i> | 8 | 7 | 9 | 3 | 6 | 6 | 10 |
| <i>FFP (units)</i> | 8 | 7 | 9 | 2 | 6 | 6 | 10 |
| Intraoperative complications | Bladder injury | No | No | No | No | No | Bladder injury and DIC |
| Postoperative packed RBCs transfusion (units) | 0 | 0 | 0 | 0 | 2 | 2 | 5 |
| Neonatal weight (gm) | 2100 | 3200 | 1800 | 2500 | 3100 | 3000 | 2000 |
| NICU admission | Yes | No | Yes | No | No | No | IUFD |

Discussion

The PAS usually manifests itself by vaginal bleeding while the occurrence of intra-abdominal bleeding is an unusual finding in PAS patients leading to delayed diagnosis of such life threatening condition. Deficiency of decidua basalis is the hallmark pathologic finding present in PAS which gives access to extravillous trophoblast to invade larger vessels beyond the junctional zone. The presence of hypervascularity at placental bed observed by prenatal imaging and at time of delivery is suggestive of presence of neovascularization around the scar area in invasive placentation (14).

This intra-abdominal bleeding that occurs with invasive placentation may result from spontaneous rupture of a surface vessel of this neovascularity or minute focal penetration of

the placenta through the markedly thinned hypervascular uterine serosa over an area of few millimeters of the LUS. The rate of accumulation of blood is variable and may be rapid leading to early manifestations of hypovolemic shock or it may be gradual, and with this later condition, the patient may be in a stable general condition until late in the disease (mimicking the subacute disturbed ectopic pregnancy), making another difficulty in the diagnosis.

The atypical presentation of our patients was in the form of chest tightness, difficulty in breathing, vague abdominal pain, repeated vomiting, sweating, shoulder pain syncopal attack and cardiac arrest. In all patients, the diagnosis was made by presence of free fluid in the abdomen, especially in the hepatorenal and lienorenal gutters by transabdominal sonography or in Douglas

Pouch by transvaginal sonography (Figure 1). The seventh case had cardiac arrest once arrived to the hospital that was reversed after cardiopulmonary resuscitation and there was massive hemoperitoneum.

At time of presentation, there was no obvious abnormality in fetal heart beat pattern, and fetal movements were good in 6 patients and in the seventh patient who presented after cardiac arrest, the fetus was dead. Also, there was no gapping of the myometrium when assessed by ultrasonography.

The gestational age at occurrence of this condition was variable between 32 and 37 weeks of gestation which highlights the importance of presence of high index of suspicion when dealing with PAS patients even at earlier gestational ages. Also, the antenatal follow up was unremarkable in all patients except in one patient (patient number 3) who had a diagnosis of cesarean scar pregnancy in the first trimester and this patient had history of only one prior CD. After counselling, the patient decided to continue pregnancy despite the risk. Early hospitalization since 28 weeks of gestation was done and ultrasonography in the third trimester revealed marked thinning of the myometrium and placental bulge (Figure 2).

We have reviewed several case reports in literature of uterine rupture in PAS (15-19). The data of these cases are inconsistent as some of them were not be directly related to pathophysiology of PAS. Also, in our opinion, we think that the use of term of rupture uterus in our patients may be misleading. This term is used frequently by obstetricians to describe complete loss of the integrity of the uterine wall. At time of diagnosis of rupture uterus, the fetus is usually dead or severely compromised and dies within very short time. Also, the fetus with the amniotic sac may be present outside the uterus.

In our patients, as described earlier, there was either rupture of surface vessel or minute focal penetration of the placenta (Figures 3 and 4) which is a unique complication of invasive placentation. The overall integrity

of the uterus was not lost with the fetus and placenta remained intrauterine. In one of our patients, we had about 3 hours between the time of diagnosis and the laparotomy as this case was Rh negative and there was no available blood, and despite this, the fetus was born living with no obvious hypoxemia. Only in the last patient, the fetus was dead because the mother had cardiac arrest.

In all of our patients, laparotomy was done depending on the presence of free fluid by ultrasonography and in all patients, variable amounts of hemoperitoneum were found (about 3 liters in the most critical one). We think that there is no role for expectant management because we found the bleeding site to be active in all cases. Also, the theoretical risk of presence of non-obstetric source of bleeding, like rupture spleen, must not delay the decision of laparotomy.

We had preferred longitudinal incision for opening the abdomen except in one patient (patient number 6) who was morbidly obese (BMI was 36.14 kg/m²) and in who, transverse incision was performed. Longitudinal incisions have many advantages. First, they are less time consuming and provide rapid access to the abdomen than transverse incisions especially all these patients had one or more prior CD with adhesion formation. Second, longitudinal incisions give access to fundal hysterotomy avoiding the placenta and minimizing the blood loss. Third, they are more exploratory than transverse incisions allowing better gain to retroperitoneal space if needed and also evacuation of all collected blood.

Cesarean hysterectomy was performed in 5 patients and resection of LUS with uterine preservation was performed in 2 patients. In hemodynamically unstable patient, performing cesarean hysterectomy is the safest method and should be done without hesitation. Also, the presence of more severe forms of PAS (Federation of Gynecology and Obstetrics grades 3a diffuse disease, 3b, and 3c) (20) indicates radical treatment.

The problem is that performing cesarean

hysterectomy is very difficult and needs much experience in placenta percreta. The condition is more dramatic and challenging in such cases as the presence of hemoperitoneum may obscure the surgical field and makes dissection in correct planes difficult. Also, the patient instability adds more tension to the surgeon. The most experienced personnel should deal with such cases and follow the well-known rules of peripartum cesarean hysterectomy as delayed bladder dissection and the start by clamping and cutting the upper pedicles first (21). The presence of huge amount of intra-abdominal bleeding should not be frustrating and the surgeon should keep in mind that all active bleeding will be controlled after bilateral clamping of uterine vessels and should keep stable and active until reaching this step. In case of severe hemodynamic instability, surgeon can apply manual aortic compression (22) and we had used this maneuver by the assistant in patient number 3 while performing the hysterectomy.

Hiramatsu described a case of placenta percreta that was presented with massive hemoperitoneum due to rupture surface vessel. Hemostasis was done at bleeding site after delivery of the fetus with delayed hysterectomy after 3 days with interventional radiologic intervention (23). Five of our patients were discharged after a period of 7 to 10 days in stable general condition. Patient number 1 was discharged after 21 days due to presence of bladder injury that was repaired by urologist and the integrity of bladder was confirmed with no fistula before discharge. Patient number 7 had presented with cardiac arrest that was reversed after cardiopulmonary resuscitation had severe intra-operative DIC and had cardiac arrest again in ICU and died after 12 hours of delivery.

Conclusion

Patients with PAS may present with atypical manifestations due to presence of intra-abdominal bleeding rather than vaginal bleeding. These include chest tightness,

difficulty in breathing, repeated vomiting, sweating, vague abdominal pain, shoulder pain, syncope, and even cardiac arrest. Any patient with PAS having any of these manifestations must be put under observation with monitoring of the general condition for signs of hypovolemia and ultrasonography should be performed by expert sonographer for early detection of free fluid in abdomen or pelvis. Confirmation of diagnosis of hemoperitoneum indicates emergent laparotomy better through longitudinal incision and cesarean hysterectomy should be the first line of treatment.

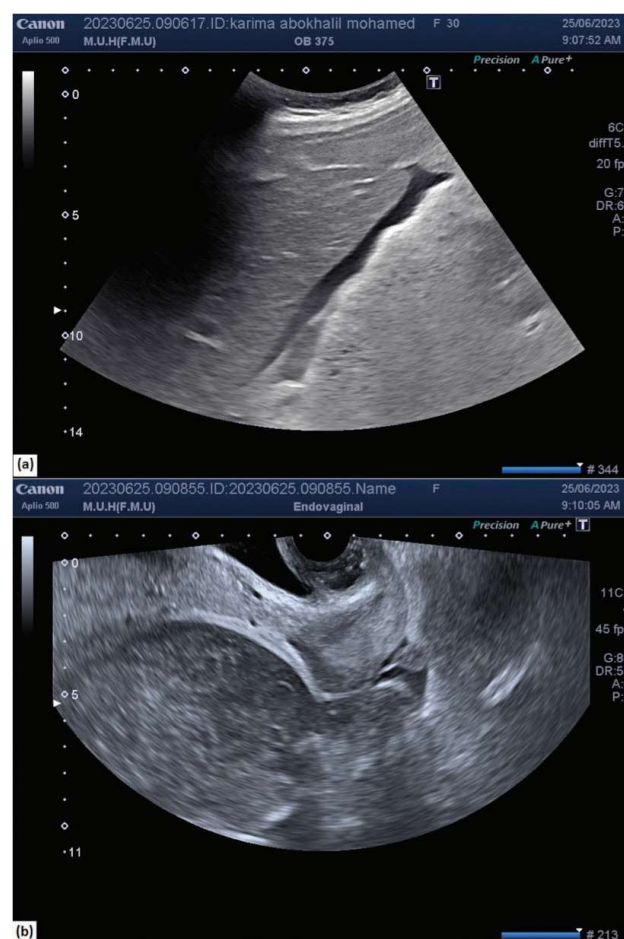


Figure 1. Ultrasonographic diagnosis of hemoperitoneum in patient number 5 through presence of free fluid in hepatorenal gutter by transabdominal sonography (a) and presence of free fluid in Douglas Pouch by transvaginal sonography (b).



Figure 2. Marked myometrial thinning and placental bulge by ultrasonography in third trimester in patient number 3.

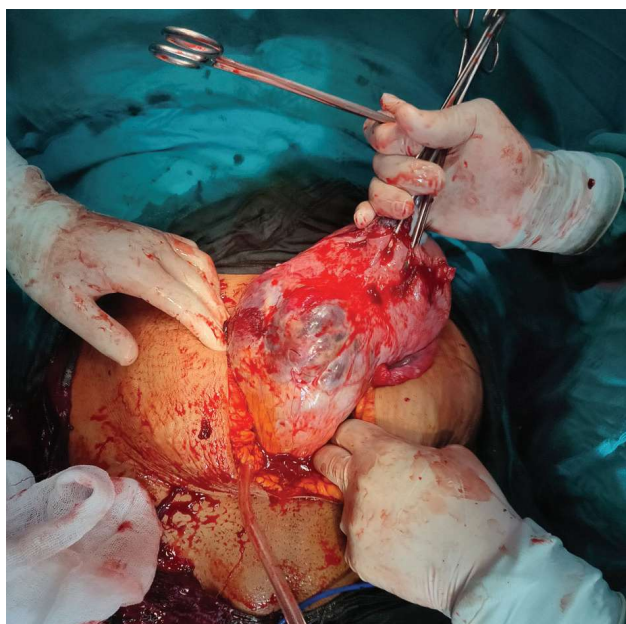


Figure 3. Intraoperative photo of patient number 5 showing placenta previa percreta with the cause of hemoperitoneum (rupture of surface vessel over placental penetration area).



Figure 4. Cesarean hysterectomy specimen of patient number 3 that had intraoperative massive hemoperitoneum due to focal placental penetration.

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Conflicts of Interest

The authors declare that they have no competing interests.

REFERENCES

1. Wu S, Kocherginsky M, Hibbard JU. Abnormal placentation: twenty-year analysis. *American journal of obstetrics and gynecology*. 2005;192(5):1458-61.
2. Bailit JL, Grobman WA, Rice MM, Reddy UM, Wapner RJ, Varner MW, et al. Morbidly adherent placenta treatments and outcomes. *Obstetrics and gynecology*. 2015;125(3):683-9.
3. Read JA, Cotton DB, Miller FC. Placenta accreta: changing clinical aspects and outcome. *Obstetrics and gynecology*. 1980;56(1):31-4.
4. Eller AG, Porter TF, Soisson P, Silver RM. Optimal management strategies for placenta accreta. *BJOG : an international journal of obstetrics and gynaecology*. 2009;116(5):648-54.
5. Warshak CR, Ramos GA, Eskander R, Benirschke K, Saenz CC, Kelly TF, et al. Effect of predelivery diagnosis in 99 consecutive cases of placenta accreta. *Obstetrics and gynecology*. 2010;115(1):65-9.
6. Belfort MA. Placenta accreta. *American journal of obstetrics and gynecology*. 2010;203(5):430-9.
7. Silver RM, Landon MB, Rouse DJ, Leveno KJ, Spong CY, Thom EA, et al. Maternal morbidity associated with multiple repeat cesarean deliveries. *Obstetrics and gynecology*. 2006;107(6):1226-32.
8. Shamshirsaz AA, Fox KA, Salmanian B, Diaz-Arrastia CR, Lee W, Baker BW, et al. Maternal morbidity in patients with morbidly adherent placenta treated with and without a standardized multidisciplinary approach. *American journal of obstetrics and gynecology*. 2015;212(2):218.e1-9.
9. Al-Khan A, Gupta V, Illsley NP, Mannion C, Koenig C, Bogomol A, et al. Maternal and fetal outcomes in placenta accreta after institution of team-managed care. *Reproductive sciences (Thousand Oaks, Calif)*. 2014;21(6):761-71.
10. Eller AG, Bennett MA, Sharshiner M, Masheter C, Soisson AP, Dodson M, et al. Maternal morbidity in cases of placenta accreta managed by a multidisciplinary care team compared with standard obstetric care. *Obstetrics and gynecology*. 2011;117(2 Pt 1):331-7.
11. Smulian JC, Pascual AL, Hesham H, Qureshey E, Bijoy Thomas M, Depuy AM, et al. Invasive placental disease: the impact of a multi-disciplinary team approach to management. *The journal of maternal-fetal & neonatal medicine : the official journal of the European Association of Perinatal Medicine, the Federation of Asia and Oceania Perinatal Societies, the International Society of Perinatal Obstet*. 2017;30(12):1423-7.
12. Allen L, Jauniaux E, Hobson S, Papillon-Smith J, Belfort MA. FIGO consensus guidelines on placenta accreta spectrum disorders: Nonconservative surgical management. *International journal of gynaecology and obstetrics: the official organ of the International Federation of Gynaecology and Obstetrics*. 2018;140(3):281-90.
13. Flores-Mendoza H, Chandran AR, Hernandez-Nieto C, Murji A, Allen L, Windrim RC, et al. Outcomes in emergency versus electively scheduled cases of placenta accreta spectrum disorder managed by cesarean-hysterectomy within a multidisciplinary care team. *International journal of gynaecology and obstetrics: the official organ of the International Federation of Gynaecology and Obstetrics*. 2022;159(2):404-11.
14. Jauniaux E, Collins S, Burton GJ. Placenta accreta spectrum: pathophysiology and evidence-based anatomy for prenatal ultrasound imaging. *American journal of obstetrics and gynecology*. 2018;218(1):75-87.
15. Enebe JT, Ofor IJ, Okafor, II. Placenta

- percreta causing spontaneous uterine rupture and intrauterine fetal death in an unscarred uterus: A case report. *International journal of surgery case reports*. 2019;65:65-8.
16. Yang Y, He F. Placenta percreta complicated with uterine rupture. 2022;305(1):291-2.
 17. Boujida S, M'Hamdi O, Flissate F, Baidada A, Kharbach A. Placenta percreta as a cause of uterine rupture in the second trimester: Case report. *International journal of surgery case reports*. 2022;94:107069.
 18. Bouab M, Kiram H, Jalal M, Lamrissi A, Bouhya S. Uterine rupture with massive hemoperitoneum due to placenta percreta in a second trimester: A case report. *International journal of surgery case reports*. 2022;99:107652.
 19. Ogoyama M, Yamamoto K, Suzuki H, Takahashi H, Fujiwara H. Uterine Rupture With Placenta Percreta Following Multiple Adenomyomectomies. *Cureus*. 2023;15(2):e34852.
 20. Jauniaux E, Ayres-de-Campos D, Langhoff-Roos J, Fox KA, Collins S. FIGO classification for the clinical diagnosis of placenta accreta spectrum disorders. *International journal of gynaecology and obstetrics: the official organ of the International Federation of Gynaecology and Obstetrics*. 2019;146(1):20-4.
 21. Kingdom JC, Hobson SR, Murji A, Allen L, Windrim RC, Lockhart E, et al. Minimizing surgical blood loss at cesarean hysterectomy for placenta previa with evidence of placenta increta or placenta percreta: the state of play in 2020. *American journal of obstetrics and gynecology*. 2020;223(3):322-9.
 22. Nieto-Calvache AJ, Palacios Jaraquemada JM, Basanta N, Aryananda RA, Sinisterra-Díaz SE, Rodriguez F, et al. Internal manual compression of the aorta- an effective way to temporarily control pelvic bleeding in obstetrical hemorrhage. *American journal of obstetrics and gynecology*. 2022;227(1):96-7.
 23. Hiramatsu Y. Cesarean Hysterectomy for Placenta Previa Accreta Using Retrograde Abdominal Hysterectomy Approaching from the Posterior Vaginal Wall. *Surgery journal (New York, NY)*. 2021;7(Suppl 1):S38-s45.