

Abdominal Ectopic Pregnancy: Challenging Obstetrical Paradox in Series of Three Cases

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ABSTRACT

Abdominal Ectopic Pregnancy (AEP), a rare life-threatening obstetrical complication, is defined as ectopic implantation within the peritoneal cavity outside the uterus, tubes, ovaries, or intra-ligamentous locations. A series of three rare cases of early AEP is presented: (First case: a 32-year-old, gravida 2, para 1; Second case: a 31-year-old, gravida 3, para 2; and third case: a 39-year-old gravida 4, para 2). All the cases were of early (<20 weeks) primary AEP and they all presented with acute abdomen, unstable vitals, and haemoperitoneum. The first case had omental implantation with a gestational sac of (3×3) cm and was diagnosed by strong clinical suspicion with intraoperative confirmation for the same, whereas, the second and third cases had implantation on Pouch of Douglas (POD) and sigmoid colon respectively; and was diagnosed by sonography with intraoperative confirmation during laparotomy. All three cases were successfully managed by a multidisciplinary team approach, blood transfusions, and emergency laparotomy with complete removal of the placenta. They had a good postoperative recovery. The reports of histopathology of tissues retrieved peroperatively from the implantation sites showed chorionic villi which confirmed the product of conceptions. Presentation of the present case series provides an opportunity to illustrate a rare variant of ectopic pregnancy with a challenging obstetric dilemma and to discuss the importance of strong clinical suspicion for such a grave maternal condition, on the part of the attending obstetrician, to make an early diagnosis and prompt treatment, so that, maternal morbidity and mortality can be avoided.

Keywords: Haemoperitoneum, Laparotomy, Sonography, Uterus

INTRODUCTION

Abdominal pregnancy is a rare form of ectopic gestation and potentially life-threatening obstetrical complication in terms of maternal morbidity and mortality. This is thought to represent around 1-1.5% of all ectopic pregnancies, with an estimated incidence varying from 1:10,000 to 1:30,000 pregnancies worldwide [1,2].

Abdominal pregnancy refers to an ectopic pregnancy that has implanted in the peritoneal cavity, external to the uterine cavity and fallopian tubes [3]. The placenta of abdominal pregnancy is often found to be attached to reproductive organs with subsequent rupture into the peritoneal cavity. Direct attachment of the placenta to the uterine serosa, omentum, bowel, and mesentery is also found [4]. Though the most frequent sites of placental implantation are the Pouch of Douglas (POD), which accounts for 55% of cases. It can also occur in mesosalpinx, the omentum, the peritoneum of the abdominal or pelvic walls, and the space between the anterior uterine wall and the bladder [1,5].

The symptoms and signs vary according to the implantation site. If the implantation site is in the pelvic cavity, early diagnosis is easily confused with tubal ectopic pregnancy [6], and only 20-40% of cases are diagnosed before surgery [7]. It increases the risk of fatal intraperitoneal haemorrhage, primarily because of the risk of massive bleeding from partial or total placental separation [8]. Therefore, it is important to diagnose and effectively manage this rare type of pregnancy, in order to reduce maternal morbidity and mortality.

CASE SERIES

Case 1

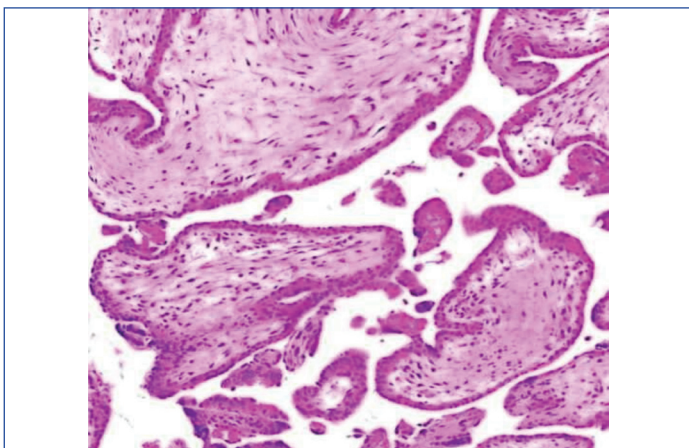
A 32-year-old, gravida 2, para 1, at six weeks of pregnancy attended the Obstetric Emergency Unit with complaints of severe pain in the abdomen of two days duration, and mild occasional bleeding per vaginum for seven days. She had a history of irregular intake of

oral contraceptive pills. She had no history of endometriosis, Pelvic Inflammatory Disease (PID); nor any Sexually Transmitted Diseases (STD). On examination, the patient was pale, her pulse rate was 140 beats per minute and feeble, blood pressure was 80/60 mmHg. Her abdomen was moderately distended with remarkable generalised tenderness and guarding, which prevented adequate palpation. Per vaginal examination revealed that the uterus was tender and almost 10 weeks in size. Bilateral fornices could not be assessed well. Cervical motion tenderness was elicited. Abdominal tapping was done and showed a bloody tap (positive tap test). No Ultrasonography (USG) report was available. The patient was resuscitated with intravenous fluids with noradrenaline support; Foley catheterisation and packed Red Blood Cells (RBC) requisition were done. A clinical diagnosis of ruptured ectopic pregnancy was made and she was prepared for emergency exploratory laparotomy.

The abdomen was opened with a low-midline incision under general anaesthesia. About two litres of haemoperitoneum and almost 500 g of clots were noted. Uterus was found to be of normal size, and bilateral tubes and ovaries were healthy. An ectopic gestational sac (3×3) cm was found to be implanted over the gut omentum close to the sigmoid colon [Table/Fig-1]. The omentum was ligated by silk sutures. The ectopic mass was clamped, and a partial omentectomy was performed to remove the gestational sac. A general surgeon was present during the laparotomy. After securing haemostasis, the peritoneum was washed with one litre of normal saline. An intraperitoneal drain was inserted and the abdomen was closed in layers. The specimen of ectopic mass with adhered omentum was sent for Histopathological Examination (HPE). The postoperative period of the patient was uneventful. She was transfused with three units of packed RBC and was discharged on postoperative day 5. Histopathological report of the ectopic mass showed a product of conception [Table/Fig-2].



[Table/Fig-1]: Ectopic gestational sac over gut omentum- intraoperative finding.



[Table/Fig-2]: Slide showing chorionic villi in conception product (H&E 100X) (Case one).

Case 2

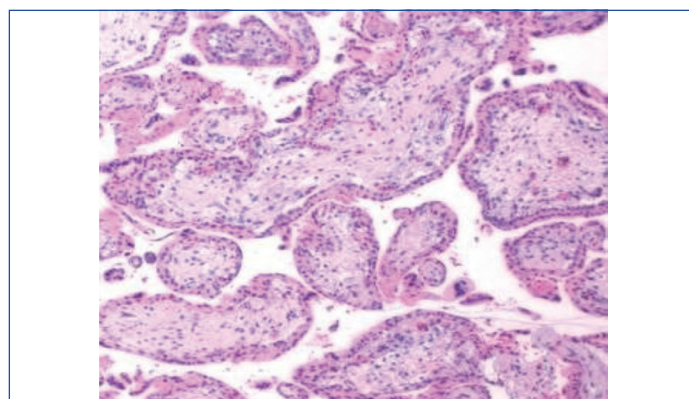
A 31-year-old pregnant lady, gravida 3, para 2, was transferred from a nearby district hospital to the Obstetric Emergency Unit of the Institution with complaints of severe pain in the abdomen for the last 24 hours and intermittent bleeding per vaginum for the last five days. This was a postcaesarean pregnancy with two living issues and her last childbirth was almost two years back. She had around six weeks of amenorrhoea with irregular menstruation and could not remember her last menstrual period. The patient had neither history of PID nor STD. She did not use any contraception and had no history of endometriosis. On examination, she was moderate to severely pale, her pulse rate was 110 beats per minute with low volume, blood pressure was 100/60 mmHg. Muscle guard, rigidity, moderate distension, and tenderness were elicited on abdominal examination. Bimanual examination revealed a bulky uterus, tender boggy mass palpable in the posterior fornix. Abdominal paracentesis revealed a frank bloody tap. Transabdominal USG revealed an empty endometrial cavity with a heterogenous mass measuring 5.5x5.4 cm, with a foetal pole of eight weeks gestation inside the gestational sac and located in the POD close to the right adnexa. There was free fluid in the paracolic gutter and pelvic cavity. The patient was resuscitated with intravenous fluid and an urgent requisition of three units of packed RBC was sent for emergency blood transfusion.

After proper counselling, an emergency exploratory laparotomy was planned in view of massive intraperitoneal haemorrhage due to separation of abdominal ectopic and impending shock. Under general anaesthesia, a median laparotomy was performed. There was moderate haemoperitoneum (one litre) with almost 500 g of clots in the abdomen. The uterus was bulky, and bilateral adnexa were normal and healthy, but there was an irregular bleeding tissue (partially separated placenta) around 4x3 cm in the POD, close to the right posterior surface of the uterus [Table/Fig-3]. The placental tissues were removed by dissecting them carefully, by digital separation from POD and posterior surface of the uterus, and sent for HPE. Bleeding from the implantation site was secured by bipolar

cautery and compression by a local haemostatic agent. After abdominal closure, uterine curettage was done and the specimen was sent for HPE to rule out retained product of conception. She was transfused with two units of packed RBC and her postoperative period was uneventful. The patient was discharged on the seventh postoperative day. Histopathology report revealed that the tissue removed from POD was the product of conception [Table/Fig-4], and the specimen of uterine curettage was decidual cast.



[Table/Fig-3]: Haemoperitoneum with partially separated placenta in POD, peroperative view.



[Table/Fig-4]: Histopathology picture of product of conception showing chorionic villi (H&E, 100X) (Case two).

Case 3

A 39-year-old, gravida 4, para 2, was referred from a district hospital after being diagnosed with Abdominal Ectopic Pregnancy (AEP) and presented with dull aching pain and huge haemoperitoneum. On examination, the patient was alert, conscious, and cooperative; blood pressure was 100/70 mmHg, pulse rate-130 beats per minute; severe pallor. Physical examination revealed huge distension of the whole abdomen with tenderness, rigidity and muscle guarding. Per vaginal examination showed os closed, cervix tubular, slightly drawn up, no bleeding per vaginum. Abdominal paracentesis revealed a positive 'tap' test for haemoperitoneum. Ultrasonography revealed a single, live foetus, 13 weeks two days, with gross intraperitoneal haemorrhage and implantation that, seemed to occur outside the uterus, adjacent to gut loops in the right adnexal region. The patient was resuscitated with intravenous fluids and an urgent blood requisition was done and the patient was prepared for emergency exploratory laparotomy after proper counselling.

Laparotomy was performed by infraumbilical median incision and about three litres of haemoperitoneum was revealed. An ensac foetus of about 14 weeks in size was found in the peritoneum with cord and placenta firmly adhered to the appendices epiploic of the sigmoid colon and partial adhesion with left conu and adnexa [Table/Fig-5]. Blunt dissection and adhesiolysis was performed to release amnion-omental adhesion. General surgery, urology, and vascular surgery services were consulted during the laparotomy. The uterus was 10 week size, both sided tube and bilateral ovaries were found to be normal and healthy, and preserved. No rent was found in the uterus.



[Table/Fig-5]: Intraoperative finding of foetus 14 weeks size, removed from the gestational sac attached to sigmoid colon.

There was continuous oozing from the placental bed adhered to the gut wall. The general surgery team ligated the site of appendices epiploicae with '1-0' silk sutures to arrest the active bleeding sites on the gut and omentum. Bipolar cautery was also used to achieve haemostasis. About two litres of normal saline peritoneal wash was given. An intraperitoneal drain was placed in the POD and the abdomen was closed in layers. A specimen of ectopic mass with placenta was sent for HPE. The postoperative period of the patient was uneventful. The patient received four units of packed RBCs and was discharged on the 14th postoperative day.

DISCUSSION

Abdominal pregnancy is an alien variation of ectopic pregnancy with high incidence, reported in women of developing countries with limited-resource settings, probably due to low socio-economic status, increased incidence of PID, endometriosis, history of infertility, tubal reconstruction surgery, pregnancy with an intrauterine device. Although these risk factors can predispose to AEP, only 50% of women with AEP are found to have any associated risk factors [1]. Cases of the present series did not have any of these risk factors. The risk of maternal and perinatal death from abdominal pregnancy

is very high. The maternal mortality in abdominal pregnancy may range from 0.5% to 18% [1]. The risk of maternal death in abdominal ectopic is 7-8 times more than the mortality, that occurs in tubal ectopic and 90 times higher than the mortality found in intrauterine gestation. The risk of perinatal mortality rate is 40-95% [1,9].

It is classified as primary or secondary based on its implantation site. Primary implantation is rare, with the incidence being 0.6-1.6% of all cases reported worldwide [1,9]. According to Studdiford's criteria (1942), the diagnosis of primary abdominal pregnancy is based on the following: 1) normal tubes and ovaries; 2) absence of an utero-placental fistula; and 3) attachment exclusively to a peritoneal surface early enough in gestation to eliminate the likelihood of secondary implantation from primary site [10]. Secondary abdominal pregnancy is the most common and frequent following rupture of tubal ectopic [9]. The most common sites of placental implantation are the POD, which accounts for 55% of cases, followed by the mesosalpinx, the omentum, the peritoneum of the abdominal or pelvic walls, and the space between the anterior uterine wall and the bladder [1]. The least common form of abdominal pregnancy is the omental pregnancy [11]. The first case of the present series, was located in the omentum, the second case was in POD and the third case was found in the peritoneum and gut wall (sigmoid colon). In the present series, the uterus, bilateral tubes, and ovaries appeared normal and the authors could not find any utero-peritoneal fistula and all three cases were detected in the early gestational period (<20 weeks). Therefore, the cases of the present series seemed to be early primary abdominal pregnancies. In 2021, one similar case was reported by Dorjey Y et al., in Bhutan [12].

Tabulated illustration with significant features of a few recent published case reports worldwide on early primary abdominal pregnancy has been depicted in [Table/Fig-6] [11,13-16].

S. No.	Study (year)	Age (years)	Parity	Risk factors	Gestational age (weeks)	Presentation	HCG (IU/L)	Imaging findings (USG, if not specified)	Outcome
1	Yasin NZHM et al., (2020) [16]	31	P2+1	Not known	Not known	Acute abdomen, pale, tachycardia	-	-	Exploratory laparotomy- haemoperitoneum, uterus and fallopian tubes normal, 'omental mass between omental fold, partial omentectomy performed
2	Yip SL et al., (2016) [14]	31	-	Not known	6 weeks	Cramping epigastric pain radiating to the supra pubic region	11,803	Empty uterus with free fluid between liver and kidneys, gestational sac outside uterus	Laparoscopy revealed haemoperitoneum, normal fallopian tubes and uterus and an omental ectopic gestation; mini laparotomy was performed for excision of the mass.
3	El Farouqi A et al., (2022) [15]	26	P0+0	Not known	18 weeks	Abrupt onset abdominal pain with vaginal bleeding	-	MRI revealed an intraperitoneal gestational sac in the left flank	Exploratory laparotomy revealed an oval foetal sac implanted directly into the abdominal cavity with a normal appearance of the uterus and fallopian tubes
4	Jayanthi R et al., (2019) [11]	26	P1+0	Not known	8 weeks 1 day	Severe lower abdominal pain, pale, hypotensive	-	Empty uterus with endometrial thickness 6 mm, minimal to moderate amount of free fluid in POD	Exploratory emergency laparotomy- haemoperitoneum of 500 mL, both fallopian tubes normal, omental ectopic identified in right lumbar region, excised with bipolar energy source
5	Bashiru JB et al., (2021) [13]	23	P1+0	Intake of abortifacient	18 weeks 1 day	Severe abdominal pain, vaginal bleeding	-	Live 18 weeks 5 days abdominal pregnancy	Exploratory laparotomy- haemoperitoneum, well formed extra uterine gestational sac at the superior surface of uterus
6	Present study (Case 1)	32	P1+0	Not known	6 weeks	Acute abdomen, hypotension, tachycardia	-	-	Exploratory laparotomy- haemoperitoneum, ectopic mass adhered to gut omentum close to the sigmoid colon, partial omentectomy performed
	Present study (Case 2)	31	P2+0	Not known	6 weeks	Severe pain in abdomen and intermittent bleeding per vaginum	-	Empty endometrial cavity with a gestational sac in POD close to right adnexa	Emergency exploratory laparotomy- haemoperitoneum of about 1 litre, partially separated placental tissue close to the right posterior surface of the uterus that was removed by digital separation
	Present study (Case 3)	39	P2+1	Not known	13 weeks 2 days	Dull aching pain, huge haemoperitoneum, hypotension	-	Gestational sac of 13 weeks 2 days close to gut loops in right adnexa, outside uterus	Exploratory laparotomy revealed 3 litres haemoperitoneum and an ensac foetus found in the peritoneum with cord and placenta firmly adhered to the appendices epiploicae of sigmoid colon and partial adhesion with left conu and adnexa

[Table/Fig-6]: Published case reports on EAP with significant findings of each case almost similar to present cases [11,13-16].
EAP: Early abdominal pregnancy

Besides, classification based on the site of implantation, abdominal pregnancy can also be classified as early or late depending on the gestational age at which it presents. Early Abdominal Pregnancy (EAP) presents at or before 20 weeks of gestation and late or advanced abdominal pregnancy presents after 20 weeks of gestation [4]. Similar to case one of the current series, omental implantation of abdominal pregnancy was reported by George R et al., Jayanthi R et al., Yip SL et al., Yasin NZHM et al., Ozdemir I et al., [2,11,14,16,17].

In concordance with case two of the present series, implantation of the placenta at POD in abdominal pregnancy was reported by Cagino K et al., Yang X and Ma K, Wong JQE and Lim YH [18-20]. Two cases of abdominal pregnancies reported from Dubai in 2018, also had a placental attachment to POD [5]. Placental attachment to the mesentery of the sigmoid colon like case three of the present series was also observed in the cases of abdominal pregnancy reported by Dubey S et al., and Yildizhan R et al., [3,8]. An ectopic mass adherent to the rectal wall was reported recently by Thang NM et al., from Vietnam [21].

The clinical presentations of abdominal pregnancy are uncertain. In a recent study, most patients had abdominal pain and vaginal bleeding and some patients needed admission for haemorrhagic shock caused by placental separation or rupture of ectopic pregnancy lesions [22]. This finding is consistent with the present cases because all patients in the present series had abdominal pain and haemoperitoneum. Massive haemoperitoneum was recently reported in a case of abdominal pregnancy from a resource-limited setting in Ghana [13]. Intraperitoneal haemorrhage was also noticed in AEP reported by others [23,24]. A case of primary omental pregnancy presented with shock due to severe intraperitoneal haemorrhage was reported from Turkey [17].

Despite wide variation in presentations, severe lower abdominal pain is one of the most consistent findings [6,13], like the cases of the present series. The clinical pictures vary according to the implantation site. If the implantation site is in the pelvic cavity, early diagnosis is easily confused with tubal ectopic pregnancy [6], and only 20-40% of cases are diagnosed before surgery [7]. The first case seemed to be ruptured tubal ectopic preoperatively, but a confirmatory diagnosis was made intraoperatively and by HPE.

Practically, diagnosis of AEP may be late and difficult because patients may remain asymptomatic, or if symptomatic, the symptoms are non specific [25]. So, a high index of clinical suspicion is of paramount importance for its early diagnosis [3]. This is consistent with the diagnosis of the first case of the present series where there was no imaging study except clinical suspicion and laparotomy findings. In many cases, the diagnosis of AEP is not confirmed, until a laparotomy or laparoscopy is performed [26]. In a resource-poor setting, the diagnosis can be made peroperatively [27]. Although serial monitoring of serum beta-Human Chorionic Gonadotropin (HCG) level is a useful tool for clinically suspected tubal ectopic, it is not consistent enough to make the diagnosis of abdominal pregnancy [3]. Transvaginal USG is considered the frontline diagnostic imaging tool with a sensitivity of 99% for the diagnosis of abdominal pregnancy [1,5]. Diagnosis of AEP is often difficult and missed during routine USG [3]. Sonography gives only 50% accuracy for diagnosis of EAP when it is used along with clinical evaluation [4]. The classical USG finding of abdominal pregnancy is an empty uterus, with a gestational sac or mass outside of the uterus, fallopian tubes, and ovaries confirming the diagnosis of abdominal pregnancy [4,5]. Magnetic Resonance Imaging (MRI) serves as an adjunct in cases when sonography is inconclusive or equivocal [5]. To confirm the location of placental and foetal tissue, MRI can be used [1]. MRI may also help in surgical planning by estimating the depth of placental tissue involvement in mesenteric and uterine attachment [28].

Several published studies reported on the therapeutic regimen including conservative and surgical treatment options for abdominal ectopic. Conservative therapy includes selective placental vascular

embolisation, ultrasound-guided drug injection (methotrexate) in the gestational sac, or maternal systemic drug therapy [29,30]. Expectant management has been attempted successfully in a few cases to achieve foetal maturity [31]. But a long follow-up period may be required in this approach.

For the management of AEP, a surgical procedure in the form of laparotomy or laparoscopy is preferable, and an excellent outcome is achieved by the complete removal of the whole sac containing the foetus and membranes along with the placenta [32]. Laparotomy is preferable in cases, where there is a risk of haemorrhage [33]. EAP with haemodynamically unstable mothers has been treated by laparotomy [14].

Laparoscopic surgery may be opted, if the cases are diagnosed early and do not carry vascular risks [26]. In 2016 Yip SL et al., concluded in a case report on primary omental pregnancy that there are increasing reports of laparoscopically managed EAP among women, who are haemodynamically stable [14]. In fact, after the year 2000, the rate of operative laparoscopy for EAP was 100%, because of significant benefits in terms of minimal blood loss, fast recovery, and a short period of hospital stay in the laparoscopic surgery [4]. Several cases were reported in recently published literature that even with haemoperitoneum, EAP was managed surgically by operative laparoscopy based on a great deal of surgical expertise and technological advances [24,34]. Pre-viable abdominal pregnancy (<24 weeks) is usually treated with laparotomy with removal of the ectopic pregnancy with or without placental removal (if low risk of maternal haemorrhage) [27]. All the patients were treated surgically (laparotomy) in the present series.

Several cases reported globally in published literature in recent times on successful surgical management of early primary AEP either by laparotomy or by laparoscopy, has been depicted in [Table/Fig-7] [2,3,11-16,18,19,23,24,34-41].

Authors who managed EAP by Laparotomy*		Authors who managed EAP by Laparoscopy†
1.	El Farouqi A et al., (2022) [15]	Wilcox A et al., (2022) [36]
2.	George R et al., (2021) [2]	Cagino K et al., (2021) [18]
3.	Bashiru JB et al., (2021) [13]	Chan WV et al., (2021) [37]
4.	Dorjey Y et al., (2021) [12]	Kang OJ et al., (2021) [24]
5.	Katke RD, (2021) [23]	Seo GH et al., (2017) [34]
6.	Yasin NZHM et al., (2020) [16]	Kaya C and Ekin M (2017) [38]
7.	Jayanthi R et al., (2019) [11]	Yang X and Ma K (2017) [19]
8.	Shaheed S et al., (2019) [35]	Cosentino F et al., (2017) [39]
9.	Dubey S et al., (2016) [3]	Nayar J and Nair SS (2016) [41]
10.	Yip SL et al., (2016) [14]	Cheung CS-y and Cheung VYT (2014) [40]

[Table/Fig-7]: EAP managed by laparotomy or laparoscopy in recent published case reports.

*Laparotomy [2,3,11-16,23,35]; †Laparoscopy [18,19,24,34, 36-41]

Massive bleeding from the placental site is a life-threatening and challenging complication of abdominal pregnancy. Generally, complete removal of the placenta is not recommended, and partial removal is required to control intraoperative haemorrhage. Sometimes, it is to be left in-situ and wait for self-involution and resorption [27].

A multidisciplinary surgical approach including gynaecological oncology, vascular surgery, urology, and trauma surgery may be necessitated because of the risk of torrential bleeding, difficult pelvic surgery, and urological complication. The risk of postoperative complications like haemorrhage or infection is to be dealt with through continuous monitoring and follow-up [42]. AEP carried to term is a rare possibility [27,42]. As per the study report, congenital malformations in the newborn are common and the risk of foetal malformations is about 40%, out of these 50% of babies can survive the first week of life [43].

CONCLUSION(S)

Abdominal pregnancy is a rare life-threatening obstetric condition. Its diagnosis and treatment are still great challenges, particularly in resource-poor settings. If detected early, laparotomy is the method

of choice for its treatment. Based on a high index of clinical suspicion along with a multi-disciplinary team approach and a great deal of surgical expertise, all the cases of AEP were successfully managed. Prompt diagnosis, judicious intervention by emergency exploratory laparotomy, and adequate blood transfusion, were the keys to save all the mothers in the present case series.

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