



Complete placenta previa and increta after radical trachelectomy: A case report

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ABSTRACT

Background: Placenta accreta spectrum (PAS) disorders are increasing in incidence and represent a significant contributor to severe maternal morbidity in the US. Prior uterine surgeries other than cesarean section are important, yet less common, risk factors for PAS.

Case: This is a case of a 43-year-old woman with a prior history of cervical cancer necessitating radical trachelectomy. She was subsequently diagnosed with a complete placenta previa with a high degree of suspicion for PAS. Multidisciplinary teams convened to plan for delivery. A cesarean hysterectomy was performed at 32 weeks. Final surgical pathology confirmed the presence of morbidly adherent placenta invading the vaginal cuff.

Conclusion: Patients who are diagnosed with early-stage cervical cancers have the option of fertility-preserving surgical management. Serial ultrasound evaluations, specifically looking for PAS, might be warranted in post-trachelectomy pregnancies.

1. Introduction

Placenta accreta spectrum (PAS) refers to varying degrees of pathological implantation of the placenta, whether as a whole or focal, to the myometrium. Prevailing theories for the underlying causes of PAS center around disruption of the endometrial-myometrial interface, usually due to antecedent iatrogenic causes (myomectomy, dilation and curettage, cesarean incision) (Liu et al., 2021). The rate of PAS is increasing in the United States (Wu et al., 2005), along with cesarean delivery rates, necessitating uniform diagnostic criteria and management guidelines. Often multidisciplinary teams are involved in the process of anticipation, planning, and delivery of these high-risk pregnancies in order to mitigate varying degrees of complexity (Society of Gynecologic Oncology et al., 2018). Abnormal placentation can lead to life-threatening hemorrhage, even when it is anticipated. Planned cesarean hysterectomy is the current standard of care for management of cases deemed to be high risk for, or with definitive diagnosis of, PAS (Liu et al., 2021). In cases of patients with atypical pelvic anatomy, operative planning and management is oftentimes complicated; this is further confounded in pregnancies where PAS is suspected.

Despite a decrease in the incidence of cervical cancer over recent decades with widespread availability of HPV vaccines as well as Pap tests (Bedell et al., 2020), there remains a population of patients who are

diagnosed with early-stage disease during their childbearing years. Only in such specific group of patients fertility-sparing surgery is a plausible option (Kasuga et al., 2021). This procedure, which is typically a radical trachelectomy (RT) either through vaginal, abdominal or laparoscopic approach, has been demonstrated to have an overall pregnancy success rate of 23.9 % (Smith et al., 2020). RT is a rare fertility-sparing procedure for FIGO 2018 stage IA2-IB1 cervical cancer (Nitecki et al., 2020). It is associated with higher risk for antepartum complications including miscarriage, preterm birth and preterm premature rupture of membrane (Kasuga et al., 2021; Smith et al., 2020). Prophylactic transabdominal cerclage is typically offered for prevention of preterm delivery, although there is limited data demonstrating that it improves the chance for a term delivery (Kasuga et al., 2021).

Here we describe a rare case of PAS in a post-trachelectomy uterus.

2. Case report

This is a case of a 43-year-old gravida-1, para-0 patient who presented to our obstetric care at 23 weeks' gestation following her move to the United States. Her medical history was significant for stage 1B1 cervical cancer diagnosed in 2015 and treated with three cycles of neoadjuvant chemotherapy (Paclitaxel and Cisplatin). Patient underwent a radical trachelectomy in 2015 with placement of an abdominal

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cerclage in the remaining tumor-free cervical stroma prior to embryo transfer. Subsequently she had in-vitro fertilization with donor eggs and her partner's sperm. She had no other significant medical or surgical history and did not use toxic substances. Her vital signs and all laboratory values were within normal limits. During an initial sonographic assessment, a complete placenta previa was noted, with marked vascularity at the intersection between the lower uterine segment and the vagina. Absent myometrial-vesical interface, presence of lacunae in the posterior aspect of the placenta as well as posterior placental bulge were also identified (Fig. 1). A subsequent non-contrast MRI of the abdomen and pelvis at 23 + 6 weeks confirmed complete placenta previa with a large focal bulge in the posterior-inferior aspect of the placenta at the lower uterine segment, and no visible myometrium surrounding the placenta in this region, with high suspicion for PAS. There was no evidence of bladder involvement (Fig. 2).

The patient presented to labor and delivery at 24 + 6 weeks with moderate vaginal bleeding and was admitted for close monitoring, steroids for fetal lung maturation, and contingency planning with a multidisciplinary team that included Ob, MFM, NICU, Gyn-Onc, nursing, Radiology, and Interventional Radiology (IR). A decision was made to keep the patient admitted for close observation until delivery, and plans were made for delivery with cesarean hysterectomy in the event of maternal or severe fetal decompensation. The patient was hemodynamically stable at admission and remained so despite intermittent mild-moderate bleeding during the first three weeks. She received IV iron sucrose to optimize her hemoglobin level (which reached 11.8 g/dl on the day of surgery). A repeat MRI at 28 weeks revealed unchanged focal lobulation of the placenta filling the lower uterine segment with absent placental-myometrial interface. T2-weighted imaging revealed new onset increased low signal within the left aspect of the placenta anterior to the focal placental bulge with absent interface between the placenta and the bladder wall, without extension into lumen of bladder (Fig. 3). This raised suspicion for placenta percreta. Delivery with cesarean hysterectomy was planned for 32 weeks' gestation, with a bleeding mitigation plan in place. The patient received a second rescue dose of steroids in preparation for the delivery.

On the day of delivery, the patient received epidural anesthesia prior to IR's placement of occluding balloon catheters within the internal iliac arteries under fluoroscopic guidance. Prior to delivery, cystoscopy and ureteric stent placement were performed by the urology team. At the time of cystoscopy, the bladder lumen appeared hyper vascular but there were no visual lesions consistent with placental tissue.

A live 1590 g female infant was delivered via *trans*-fundal incision, with APGAR scores of 9 and 9 at 1 and 5 min respectively. The hysterotomy was subsequently closed and the hysterectomy started with the

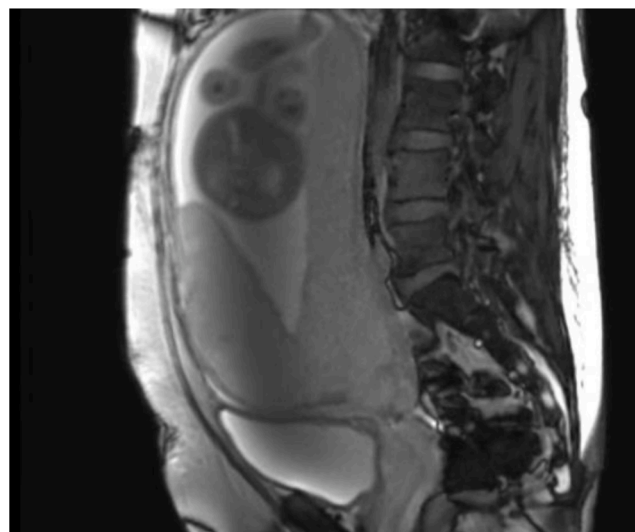


Fig. 2. Non-contrast MRI abdomen/pelvis at 23 week's gestation. Large focal bulge in the posterior inferior aspect of the placenta in the lower uterine segment and no visible myometrium surrounding the placenta in this region. The placental tissue in this region is heterogeneous and associated with a low signal band.

placenta still in-situ. The hysterectomy was performed with close attention to the dissection of the utero-ovarian vascular bundle. Location of the utero-vaginal junction was confirmed via intra-operative vaginal exam. After removal of the specimen, no remaining placental tissue was identified within the vagina (Fig. 4). A cell saver was utilized throughout the surgery, and the patient was transfused back 200 cc of autologous red blood cells plus 1 unit of packed red blood cells and 1 unit of fresh frozen plasma intra-operatively. The total estimated blood loss was 1000 mL. The preoperative hemoglobin was 11.8 GM/DL and the postoperative hemoglobin the following day was 12.2GM/DL. Patient had an uncomplicated postpartum course and was discharged from the hospital on postpartum day four. The final histopathology confirmed placenta accreta/increta.

Comment:

The pathophysiology of PAS is not well understood. The prevailing hypothesis suggests that the disruption of the endometrial-myometrial interface from prior uterine surgery allows for abnormal invasion of trophoblasts in the area of the uterine scar (Jauniax et al., 2018; Hussein et al., 2022). Most cases of PAS have been reported in pregnant patients with prior histories of cesarean delivery, myomectomy or

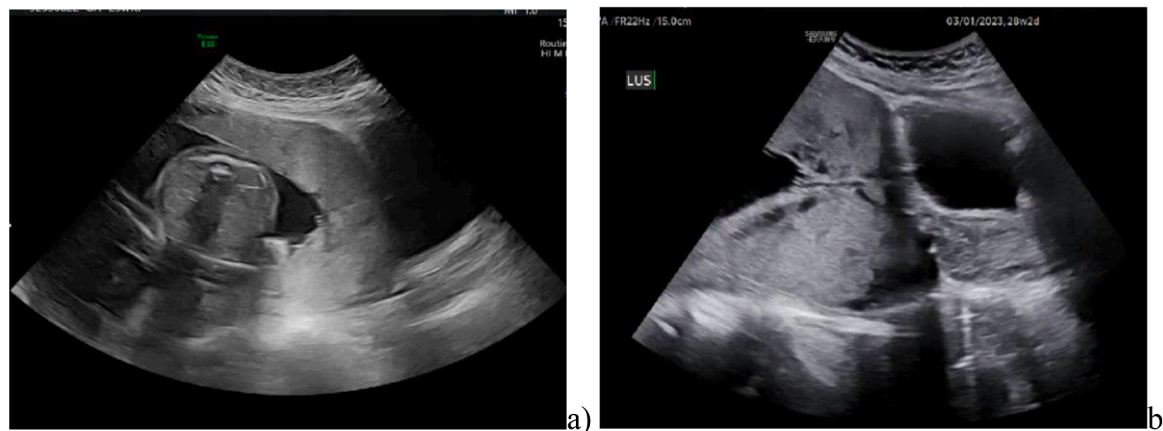


Fig. 1. Transvaginal sonographic images. A). A central placenta previa was seen at 23 weeks' gestational age. The placental-vesical interface is obliterated. There is loss of the normal hypoechoic plane in the posterior myometrium beneath the placental bed. B). Repeat transvaginal ultrasound at 28 weeks' gestational age. There is bulging of the posterior placenta into surrounding region. Multiple placental lacunae seen.

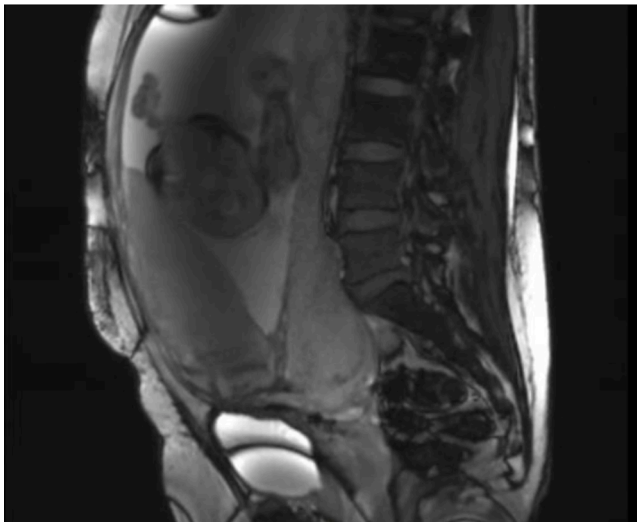


Fig. 3. Repeat non-contrast MRI abdomen/pelvis at 28 week's gestation. Unchanged focal lobulation of the placenta filling the lower uterine segment. Increase in low signal tissue/fibrosis within the left aspect of the placenta anterior to the focal placental/uterine bulge with absent interface between this placental tissue and the wall of the left bladder dome.

dilation and curettage of the uterus. However, to our knowledge (after searching multiple research databases including PubMed, Google Scholar and Cochrane Library), there have been no reported cases of PAS in post-trachelectomy pregnant patients.

One of the major challenges encountered in our case was the difficulty of interpreting sonographic findings, in part due to post-RT changes in the pelvic anatomy. Ultrasound images revealed increased vascularity at the uterovaginal anastomosis suggestive of post-operative varicosity following RT which could increase the risk for bleeding in pregnancy (D'Antonio et al., 2014). Overall, the absent myometrial-vesical interface, the presence of placental lacunae as well as placental bulge were highly suggestive of PAS. The role of MRI for PAS is unclear,

however, it appears to be beneficial in evaluating PAS in the posterior position, such as in our case, and to potentially assess the depth of PAS invasion in the surrounding regions including the parametrium and bladder (Liu et al., 2021; Tantbirojn et al., 2008).

It is unclear whether PAS could worsen throughout pregnancy, but it has been hypothesized that the extent of invasion could be dependent on the depth of the original scar (Yi et al., 2010). After a second MRI was repeated at 28 weeks, concerns were risen as to whether the placenta could eventually invade into the bladder. The absent interface between the placental tissue and the wall of the bladder could indicate further progression of PAS to placenta percreta, and the necessity for early delivery to minimize maternal morbidity. Optimization of the fetus gestation was also taken into consideration in determining the timing of delivery. After a multidisciplinary meeting between all the departments involved, a planned cesarean hysterectomy was scheduled at 32 weeks gestational age.

To minimize intraoperative blood loss, multiple strategies have been proposed. Intraoperative use of cell salvage, arterial embolization, temporary arterial occlusion using a balloon catheter are some of the adjuvant interventions that were considered to control for possible hemorrhage and reduce maternal complications (Bartels et al., 2023). Multiple case reports in the literature have documented the efficacy of endovascular balloon catheters (Knuttinen et al., 2012; Pini et al., 2023; Nankali et al., 2021). Perhaps, most importantly, Anisodoweleh et al have shown that internal iliac balloon occlusion can significantly mitigate blood loss intraoperatively and reduce the need for perioperative blood transfusion as reported in their systematic review and meta-analysis. In their study, 29 articles were analyzed with a total sample size of 1140 in the control group, and 1225 in the balloon occlusion group, showing a positive effect of the intervention (Kasuga et al., 2019). In our case, the endovascular balloon catheters were placed in the internal iliac arteries by interventional radiology prior to delivery and were inflated at the time of the hysterectomy. Promising outcomes reported in the literature suggest that prophylactic vascular occlusion of the internal iliac artery should be considered as standard care in managing PAS.



Fig. 4. Anterior and posterior view of the uterus following cesarean hysterectomy. Placenta was left in-situ during the closure of the hysterotomy. The surgical stump post-trachelectomy displays a tan-brown, hemorrhage soft placental tissues bulging out of the uterus.

3. Conclusion

Fertility-sparing RT for cervical cancer allows women in their child-bearing age to consider the option of pregnancy. Although not reported in the literature, PAS can arise in the anatomically distorted pelvis following RT. PAS calls for a multidisciplinary approach due to the high mortality and morbidity of the diagnosis. Appropriate contingency plans to minimize blood loss must be discussed in a timely manner before delivery and cesarean hysterectomy. Most importantly, targeted ultrasound assessments to identify signs of PAS should be considered in post trachelectomy pregnancies.

4. Informed consent Statement

The authors confirm that a written informed consent has been obtained from the involved patient who has given approval for this information to be published in this case report.

CRedit authorship contribution statement

Jiahua Chen: Writing – original draft. **Laura Gilroy:** Writing – review & editing. **Howard Minkoff:** Writing – review & editing. **Albert Palileo:** .

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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