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CASE REPORT**

Gynaecol Perinatol 2017;26(3-4):136-140

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## PLACENTA PRAEVIA PERCRETA WITH INITIAL BLADDER AND PARAMETRIAL INVASION: A CAUSE OF LIFE THREATENING HEMORRHAGE AFTER REPEATED CESAREAN SECTION

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Case report

*Keywords:* placenta praevia percreta, cesarean section, hemorrhage

**SUMMARY.** Placenta percreta is a rare, but potentially life-threatening condition associated with high maternal mortality and morbidity rates, and caused by severe obstetric hemorrhage. Due to rising cesarean section rates, an increased incidence of different forms of adherent placentas (accreta, increta and percreta) has been observed. Although unsuspected during the antenatal period, diagnosis at the time of labor is usually secondary to the inability to define a cleavage plane. The associated hemorrhage can be substantial, and hysterectomy is frequently required. Definitive surgical management is the traditional treatment strategy; however, several authors have recently reported their experiences with conservative management, and some of them had success with this approach. We describe a case of massive, post-cesarean vaginal hemorrhage that occurred in the third postpartum period as the result of a misunderstood placenta percreta invading the parametria and bladder. Post-cesarean hysterectomy, bladder wall repair, and unilateral internal iliac artery ligation were performed to control massive intraoperative hemorrhage. There should be high rate of suspicion for placenta percreta with bladder and parametrium invasion in the evaluation of pregnant women with a history of Cesarean delivery and placenta previa at Cesarean section.

### Introduction

Initially described in 1900 by Alexandroff,<sup>1</sup> placenta percreta is a potentially life threatening condition with fewer than 100 cases reported in the literature.<sup>2-4</sup> The overall incidence of placenta accreta, placenta increta and placenta percreta is between one in 540 and one in 93,000 deliveries.<sup>4,5</sup> Clark et al. undertook a review of 73,819 deliveries involving 52 postpartum hysterectomies for abnormal placentation, of which 76% were placenta accreta, 18% were increta, and 6% were percreta.<sup>5</sup> Although the exact etiology remains unknown, it is believed that the entity is the result of an absent decidua basalis and incomplete development of the fibrinoid layer. This is supported clinically by the observation of its marked increase in the setting of placenta previa and prior uterine surgery, especially previous Cesarean delivery.<sup>1,2</sup> Other antenatal risk factors include increased maternal age, increased parity, and an unexplained elevation in maternal serum [alpha]-fetoprotein.<sup>2,3</sup>

The degree of invasion by the placenta has been histologically differentiated into three categories. Placenta accreta occurs when trophoblastic tissue invades beyond the normal fibrinoid layer and attaches directly to the underlying myometrium. When the placental tissue extends into the myometrium it is referred to as pla-

centa increta, and when it extends beyond the uterine serosa as placenta percreta.

Placenta percreta can be a very challenging situation for the obstetrician, and this complication of pregnancy is rarely diagnosed prenatally. The increased utilization of Cesarean delivery today is strongly associated with a higher frequency of placenta praevia. An increase in frequency from 1 in 1000 pregnancies in 1950 to 101 in 1000 in 1985 has been noted.<sup>5,6</sup> Given the known association between placenta previa and placenta accreta/percreta, it is not unreasonable to suggest that the increased Cesarean delivery rate has directly contributed to the rising incidence of placenta accreta/percreta. We know that 75 percent cases of placenta percreta are associated with placenta praevia. Approximately 25 percent of women with placenta praevia and one previous Cesarean delivery have an accreta/percreta, whereas almost 50 percent with placenta praevia and two prior Cesarean deliveries have placenta accreta/percreta.<sup>6</sup> The risk of placenta previa is linked to the number of prior Cesarean deliveries, with a risk of 0.65 percent after one, 1.8 percent after two, 3 percent after three, and 10 percent after four Cesarean deliveries.<sup>5</sup>

When one considers the patient demographics faced by the modern obstetrician, it is expected that we will have to deal with this clinical scenario more frequently.

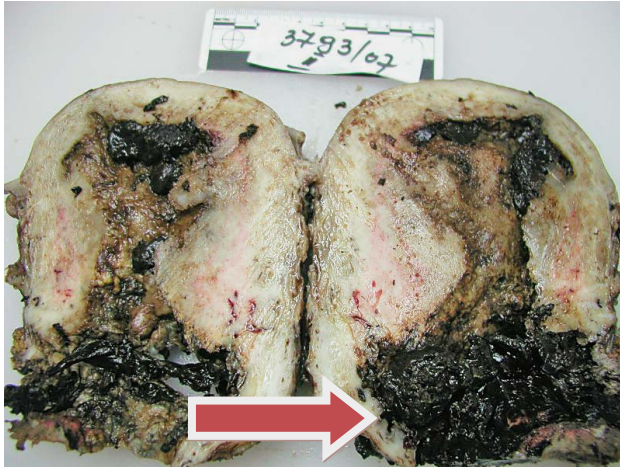


Figure 1. The arrow indicates trophoblast tissue on the inner side of isthmus towards the cervix, penetrating through the muscular wall (*placenta previa percreta*)



Figure 2. An accessory lobe with its own blood vessels (*placenta succenturiata*)

## Case report

A 33-year-old, pregnant woman, multigravida, with a history of one Cesarean section, was admitted to the Department of Gynecology and Obstetrics, General Hospital Pula at 28 weeks of gestation because of mild, painless vaginal bleeding. Ultrasound examination showed complete placenta praevia. After this episode, the patient was discharged home at her own request with recommended home support, bed rest and medical supervision.

At 36 weeks of gestation the patient was again admitted to our department because of severe vaginal bleeding and hemorrhagic shock. Physical examination demonstrated tachycardia, hypotension and anemia. The laboratory blood analysis showed increased levels of hemoglobin (9.8 gm/dL) and hematocrit (29%) with normal platelet rate. Furthermore, her coagulation profile was within normal limits. We immediately started a resuscitation procedure with an infusion of crystalloid

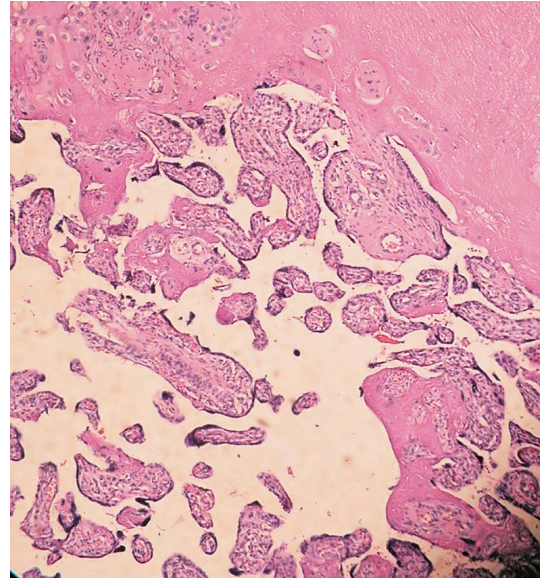


Figure 3. Thin, blood-imbibed fibrous wall with penetrating chorionic villi (HE, 100×)

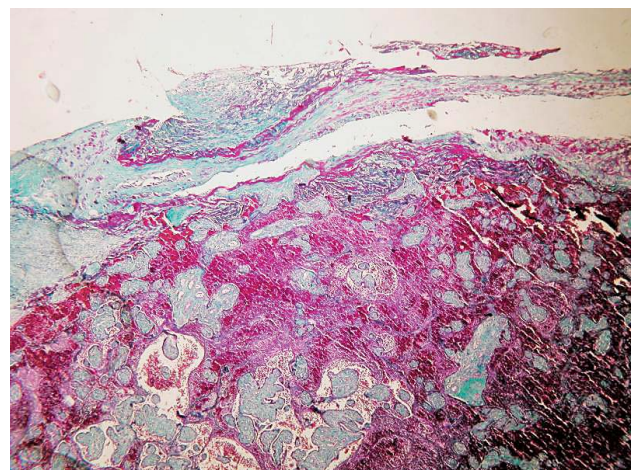


Figure 4. Lacerated fibrous wall with chorionic villi on the external wall side (Malory, 100×)

solution and a transfusion of packed red blood cell. An emergency Cesarean section was performed. The abdomen was entered through a Phannenstiel incision. The fetus, delivered through a classical incision, weighed 2800 g and had an Apgar score of 10 estimated at one and five minutes after the delivery. The placenta was tightly adherent to the lower uterine segment, and was manually removed. After a double layer closure and few haemostatic sutures the bleeding stopped. After the Cesarean section the patient was admitted to the Intensive Care Unit. However, after two hours, re-exploration was required due to ongoing vaginal hemorrhage. There was persistent diffuse bleeding from pelvic structures, and a supracervical hysterectomy and left hypogastric ligation were performed. The tear and the hemorrhage from the bladder base performed a dissection of lower uterine segment. The bladder defect was repaired in two



layers, based on consultation with a surgeon. Three days after the operation, the patient experienced multiple problems, including abdominal distention and meteorism. An abdominal X-ray examination indicated the presence of multiple aeroliquid levels, and paralytic ileus was diagnosed. After two days of support with nasogastric suction and parenteral feeds, the passage was restored. The patient received 10 units (U) of packed red blood cells and 7 U of fresh frozen plasma, and was discharged home on post-operative day 16.

## Histopathology

An analysis of placenta and uterus was performed by the Department of Pathology and Forensic Medicine, General Hospital Pula, Croatia.

On gross examination of the uterus, trophoblast tissue remained within lower part of uterus towards the cervix, penetrating through the fibrous wall (*placenta praevia percreta*, *Figure 1*).

On the gross examination, the placenta was discoid, lacerated and covered by coagulated blood. A small accessory lobe, connected to the main part of the placenta by its own blood vessels, was found (*placenta succenturiata*, *Figure 2*).

Microscopic analysis of tissue specimens from the lower uterine segment showed a thin, blood imbibed fibrous wall with penetrating chorionic villi (*Figure 3 and 4*).

## Discussion

Maternal mortality and morbidity are significantly increased by placenta percreta. Mortality, secondary to hemorrhage and its complications, can be as high as 10 percent.<sup>7</sup> Significant intraoperative blood loss may necessitate massive blood transfusion with the attendant complications of disseminated intravascular coagulation (DIC), transfusion reactions, alloimmunization, fluid overload, and less commonly, infection. Surgical morbidity includes: hysterectomy, bowel injury, urological injuries (including a 2 to 3 percent risk of ureteral trauma), and bladder lacerations that may require partial vesical resection. The patients are also at increased risk for thrombotic events. Despite prophylactic antibiotic therapy, there is a high incidence of sepsis and infectious morbidity. Also, secondary acute respiratory distress syndrome (ARDS) is not uncommon in these patients. Postoperative bleeding necessitates re-exploration in up to 7 percent of patients with placenta accreta/percreta.<sup>7</sup>

Management options for placenta percreta consist of surgical removal of the uterus and involved tissues or, alternatively, conservative treatment with the placenta left in situ after delivery. Based on a questionnaire issued to members of the Society of Perinatal Obstetricians,<sup>3</sup> O'Brien *et al.* reported that 93% of cases of placenta percreta were managed with total hysterectomy and removal of involved tissues.

Placenta percreta that invades the myometrium and serosa to the urinary bladder is a rare condition that may cause catastrophic obstetric and urologic complications.<sup>8</sup> The largest meta-analysis performed regarding this condition includes 54 cases, of which 44% of the women were reported to have had a partial or total cystectomy. Three maternal deaths were reported among the 54 cases.<sup>8</sup> The management of this rare situation is complicated and remains challenging. A collaborative team approach to management is imperative, because it is necessary to discuss all of the various options with the patient and the appropriate consultants.

Urologists should consider this entity when evaluating pregnant women with hematuria. Gross hematuria is rare even when the bladder is invaded, and it occurred in only 6 of the 27 previously reported cases.<sup>9–14</sup> Microscopic hematuria associated with the other clinically suspicious features should also alert the urologist to possible placental invasion of the bladder. Uterine rupture is a dreadful outcome of placenta percreta, although it has not been reported in cases of vesical involvement. However, a patient of Bakri *et al.* presented with a uterovesical fistula.<sup>15</sup>

Most patients with placenta percreta require hysterectomy. When the diagnosis is established early, delivery may be considered at 34 to 35 weeks of gestation since the incidence of prepartum hemorrhage markedly increases beyond week 36.<sup>16</sup> After a high vertical hysterotomy is performed to remove the fetus, manual removal of the adherent placenta should be avoided because it results in massive hemorrhage. Instead, immediately proceeding to hysterectomy is recommended. In cases of bladder involvement, care must be taken not to attempt to dissect the bladder away from the lower uterine segment, as this results in torrential bleeding. Anterior bladder wall cystotomy is particularly helpful for defining dissection planes and determining whether posterior bladder wall resection is required. Price *et al.* describe a modified posterior approach to hysterectomy that involves en bloc removal of the posterior bladder wall, followed by reconstruction when it is impossible to separate the bladder from the lower uterine segment.<sup>16</sup> After bladder wall resection and reconstruction adequate drainage via urethral and suprapubic catheters is required. In patients with massive intraoperative hemorrhage, immediate compression of the infrarenal aorta may significantly decrease bleeding. Isolation and temporary occlusion of the infrarenal aorta may then enable the surgeon to assess and manage the situation more effectively. Hallak *et al.* report that a transvaginal pressure pack is useful when consumptive coagulopathy makes hemostasis impossible using conventional surgical methods.<sup>18</sup>

The antenatal diagnosis of placenta accreta has been described using both ultrasound and magnetic resonance imaging when there is clinical suspicion.<sup>19–21</sup> Neither modality is 100% sensitive or specific. Ultrasound findings described include the following: an absence of the hypoechoic layer normally seen between the pla-

centa and myometrium, numerous placental lacunar vascular spaces, and the extension of tissue with placental density into the myometrium or serosal layer.<sup>2,6</sup> Color Doppler imaging has been used in evaluation and can be helpful in defining the accompanying abnormal vascularity.<sup>19–21</sup> T2-weighted magnetic resonance imaging findings include focal myometrial thinning and an indistinct myometrial-placental border.<sup>19,21</sup>

When invasion of the urinary bladder by placenta praevia accreta is preoperatively detected, the literature mentions two methods. The first is to reduce blood perfusion to the uterus by occluding the anterior division of the internal iliac arteries. The inflation of balloons localized in the internal iliac arteries immediately and rapidly decreases bleeding in the surgical field. Besides decreasing blood flow to the uterus and reducing hemorrhage, this action allows for better exposure for the surgical staff. The second technique is to perform a subtotal hysterectomy, removing most of the placental volume and leaving the portion of the placenta attached to the posterior wall of the bladder in situ. It is believed that by not attempting to remove this portion of the placenta, it is possible to prevent bladder injury and the need for a partial or total cystectomy.

The use of methotrexate for conservative treatment of placenta percreta has been suggested by some, but others argue against it.<sup>22,23</sup> In this case, the patient's initial [beta]-hCG level was low. Because the use of the drug is still disputed, we chose not to use methotrexate post-operatively. The [beta]-hCG levels fell spontaneously, and the placental tissue shrank.

Several reports have been published recently regarding the use of prophylactic occlusive balloon catheters and embolization for the treatment of severe obstetric hemorrhage.<sup>22,23</sup> Ojala *et al.*<sup>24</sup> report the use of this technique in seven patients with abnormal placentation diagnosed prenatally. There were no complications associated with the procedure when performed in a prophylactic setting, and good outcomes were reported. Several complications were associated with emergency embolization, such as vaginal necrosis, paresthesia of the leg, and thrombosis of the popliteal artery. However, conservative management in cases of placenta percreta invading the bladder, including leaving both the uterus and placenta in situ, use of methotrexate, and uterine artery embolization, has not always been so successful. Severe hemorrhage and emergency hysterectomy have occasionally resulted from such conservatism.<sup>25,26</sup> On the other hand, the surgical approach, i.e., total hysterectomy, has been reported as the treatment of choice in cases of abnormal placentation. In cases where the placenta invades the urinary bladder the problems associated with the surgical approach are compounded because hysterectomy alone cannot effectively remove the placenta in its entirety. Hemostasis is difficult to achieve. Bladder resection makes the operation technically more complex, and exposes the bladder and the ureters to the risk of injury. Applying prophylactic occlusive balloon catheters in cases of placenta percreta

invading the urinary bladder combined with subtotal hysterectomy – leaving that portion of the placenta that is adherent to the bladder in situ – is a good strategy that may reduce blood loss and preserve an intact bladder. However, the patient should be informed of the reported complications of the procedure and the limited experience in achieving this goal before asking for her consent.<sup>22–24</sup>

## Conclusion

Placenta percreta, which invades through the myometrium to the serosa and occasionally to adjacent organs, is a rare and complicated form of pathologic placentation. Due to the rising rate of cesarean deliveries, more cases of abnormal placentation are being reported.

The optimal management of a patient with placenta percreta is complex and involves the simultaneous cooperation of members from a number of disciplines. This multidisciplinary team should ideally be headed by the obstetrician. An organized approach allows everyone to understand what to expect, what their particular contribution will be, and reduces confusion when the surgery takes place.

Our experience with such a case has led us to consider the prenatal radiologic techniques used in the diagnosis of placenta percreta, to involve simultaneous cooperation from members of a number of relevant disciplines, and to consider other surgical and non-surgical approaches to care.

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*Paper received:* September 26, 2018., accepted: October 28, 2018.

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## PLACENTA PREVIA PERCRETA S POČETNOM INVAZIJOM MOKRAĆNOG MJEHURA I PARAMETRIJA: UZROK PRIJETEĆEG KRVARENJA NAKON PONOVLJENOG CARSKOG REZA

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*Prikaz bolesnice*

*Ključne riječi:* placenta praevia percreta, carski rez, krvarenje

**SAŽETAK.** Placenta percreta je rijetko, ali potencijalno životno opasno stanje u porodništvu, povezano s visokim morbiditetom i mortalitetom majke, uzrokovano teškim krvarenjem. Zahvaljujući povećanom broju porođaja carskim rezom, primjećuje se i povećana pojava različitih oblika neadekvatnog pripoja posteljice (placenta *accreta*, *increta* i *percreta*). Iako ne postoji sumnja tijekom antenatalnog razdoblja dijagnoza u vrijeme porođaja sekundarna je u odnosu na nemogućnost procjene stupnja razdora. Pridruženo krvarenje može biti vrlo obilno i zbog toga se često mora učiniti histerektomija. Definitivno kirurško zbrinjavanje je tradicionalna strategija liječenja; ipak neki autori nedavno su objavili svoja iskustva s konzervativnim liječenjem i uspjehom u takvom pristupu liječenju. Prikazali smo slučaj masivnog vaginalnog krvarenja, nakon porođaja carskim rezom, koje je nastupilo u trećem postpartalnom periodu, kao rezultat neprepoznate placente perkrete, koja je zahvatila parametrije i mokraćni mjehur. Kako bi se zaustavilo intraoperativno krvarenje učinjena je histerektomija, uz korekciju zida mokraćnog mjehura i jednostrano podvezivanje ilijačne arterije. Kod evaluacije trudnica s prethodnim carskim rezom i placentom previjom trebali bi češće posumnjati na postojanje placente perkrete s infiltracijom mokraćnog mjehura i parametrija.