

Uncommon Cause of Ante Partum Hemorrhage: Cervical Varicosities Associated with Placenta Previa. Case Report and Literature Review

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Abstract

Varices are common in pregnancy but cervical varices are rare and it could be a cause of massive antepartum hemorrhage. We present a case report of cervical varices, in a 31-year-old woman with placenta previa. The clinical course, imaging findings along with the outcome are being presented. In addition a literature review of published cases was done for the purpose of establishing a possible management plan.

Keywords: Cervix; Varices; Placenta Previa; Hemorrhage; Pregnancy

Abbreviation

MRI: Magnetic Resonance Imaging

Introduction

The causes of antepartum hemorrhage are diverse, varied and multifactorial. Antepartum hemorrhage complicates 2 - 5% of pregnancies [1]. The two most important causes of antepartum hemorrhage are placenta previa and abruptio placentae constituting more than 50% of cases. Cervical varices are rare cause of antepartum hemorrhage and only limited number of cases were reported [2-4]. In the presence of cervical varices and placenta previa, the exact origin of bleeding in case of antepartum hemorrhage, can be uncertain. Antepartum hemorrhage can be severe in this situation, which can lead to serious maternal and fetal morbidity. Therefore, it is important to report such rare case, so it will add to our understanding to the problem and the management options possible.

Case Presentation

A 31-year-old Gravida 1 Para 0 with no previous medical or surgical history, presented to emergency department in Women Wellness and Research Center at 18 weeks of pregnancy complaining of painless unprovoked vaginal bleeding of about 50 ml. This pregnancy was spontaneous, and its course was normal until that event. Sterile speculum examination was done and no cervical abnormality was noticed initially. Transvaginal ultrasonography demonstrated a posterior low lying placenta covering the internal orifice of the cervix. Placental lacunae with exaggerated posterior cervical and retro cervical vascularity were also noted at the time of the scan (Figure 1).



Figure 1: (a) Placenta completely covering the internal cervix with multiple lacunae suggestive of blood vessels. (b) Color Doppler imaging confirming the vascularity of the lacunae.

The patient was admitted for observation with the working diagnosis of threatened miscarriage. Bleeding stopped the same day. During her hospital stay, there was no further bleeding. She was discharged after three days with a follow up appointment to be seen in the feto-maternal unit. Transvaginal ultrasonography was done after 2 weeks at 20 weeks of gestation, confirming the diagnosis of placenta previa. At this stage, sterile speculum examination showed a mesh of engorged smooth woven cord like structures. The cervix appeared, on the scan, with complex matrix blood vessels mainly venous suggestive of cervical varices (Figure 2).



Figure 2: (a) Complex matrix vascularity on the color Doppler, (b) Same image without Doppler.

Ten days after the ultrasonography, the patient was admitted again at 21 weeks of gestation, with moderate unprovoked vaginal bleeding. The clinical examination showed the same structure in the cervix but with no active bleeding. She was kept under observation in the hospital with the diagnosis of antepartum hemorrhage, with its origin either from cervical varices or the placenta previa.

After extensive counselling of the parents, from a multidisciplinary team involving Obstetricians, Anesthesiologists, Neonatologists, regarding the origin of bleeding and the possible outcomes for both mother and fetus, the decision was made for conservative management unless there is bleeding enough to cause fetal or maternal compromise. The aim was to observe and stabilize the patient with blood transfusions and to try to reach the age of fetal viability for possible delivery according to the clinical situation.

Although, careful speculum examination showed no active bleeding from varices, cervical varices were considered the most likely cause of antepartum hemorrhage in view of the absence of abruptio or placental separation on the different scans.

During this hospital stay, the patient had frequent repeated, painless, unprovoked vaginal bleeding, leading to a drop of hemoglobin from 10.5 to 7 g/dl. The estimation of total blood loss was about 2500 ml, and patient needed replacement by blood transfusions.

At 23 weeks of gestation, Magnetic Resonance Imaging (MRI) of the pelvis was performed but failed to clearly identify the cervical varices (Figure 3).

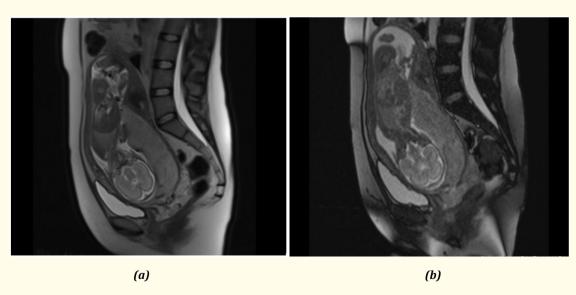


Figure 3: MRI of the pelvis in T1 (a) and T2 (b). Placenta Previa type III with no MRI evidence of Placental invasion.

At 24 weeks of gestation, considering the risk of preterm delivery, antenatal corticosteroids were given to accelerate fetal lung maturity.

After one month from her date of admission, at 26 weeks of gestation, the patient had recurrent severe fresh vaginal bleeding, about 2000 ml, with continuing ongoing vaginal bleeding, leading to unstable vital signs. The decision was taken for delivery by emergency cesarean section and vaginal examination under general anesthesia. The blood bank was notified, and six units of crossmatched packed red blood cells were available. The outcome of the cesarean section was a delivery of a baby boy of 1020g with Apgar score of 8 and 9 at 1, and 5 minutes respectively. The baby was admitted to neonatal intensive care unit for severe prematurity. The cesarean section confirmed the diagnosis of placenta previa posterior type III and no placental separation was found. The estimated blood loss during cesarean section was 500 ml, but active vaginal bleeding continued after completing the cesarean section. Vaginal examination under anesthesia was performed. Cervix was hyper vascular with multiple varices noted all around with active bleeding. Hemostasis was achieved by ligation of cervical varices using a figure of 8 hemostatic sutures at 1, 3, 7, 9 and 11 o'clock. The total blood loss during surgery was 3500 ml, with about 3000 ml coming from cervical varices. Patient received seven units of blood transfusion.

The postoperative period was uneventful with normal lochia and no active vaginal bleeding. The patient was discharged at day five post-cesarean section. She was seen six weeks later in the outpatient clinic with a normal postpartum course and normal looking cervix on vaginal examination. Transvaginal ultrasonography was performed and cervical varices have disappeared completely (Figure 4). Baby was discharged from hospital 6 weeks after cesarean section in good condition.



Figure 4: (a) Aspect of the cervix in transvaginal ultrasonography showing absence of varices and (b) same image with color Doppler.

Discussion

Varicose veins, in general, are common in pregnancy with a prevalence of up to 40% [5]. However, cervical varicosities are quite rare in pregnancy with only 22 cases reported in the literature [2-4,6,8-12].

In normal pregnancy, vaginal and uterine veins form vaginal and uterine venous plexuses along the sides of the vagina and cervix, and tributaries drain into the internal iliac veins. The exact mechanism of formation of cervical varices is unknown, however few theories were given.

Firstly, compression of the internal iliac veins by the gravid uterus could cause formation of dilated vessels similar to the mechanism of lower limb varicosities, vulvar varicosities or hemorrhoidal plexus. This could explain the formation of cervical varices in twins pregnancy and polyhydramnios cases. However, out of the 22 cases reported in the literature, only one was associated with polyhydramnios and one with twins pregnancy [13]. These findings were absent in our case.

Secondly, in cases of placenta previa, increased blood drainage of the placental bed locally could result in greater venous pressure and formation of varicosities as explained by Sammour, *et al* [6]. Out of the 22 cases reported in the literature, 16 were associated with placenta previa [13]. In cases of placenta previa, specially abnormally adherent ones, neovascularization can occur and lead to the formation of new vessels as mentioned by Chou., *et al* [7]. In the case we presented, placenta previa was present.

Finally, Diethylstilbestrol exposure in utero is said to be a possible cause leading to the formation of cervical varices [13]. Out of the 22 reported cases, four had this exposure in utero [13]. In our case, patient had no exposure to Diethylstilbestrol as mentioned in the history of the patient.

The presence of cervical varicosities along with placenta previa can lead to severe antepartum hemorrhage and preterm delivery as described in our case. The literature review confirms this statement with an average blood loss of 1636 ml and maximum noted of 5000 ml [14]. The need for blood products is also increased with transfusion rate of 45.5% [14]. There is one reported case of hysterectomy for ruptured cervical varices associated with severe hemorrhage [9].

Severe antepartum bleeding can lead to iatrogenic preterm delivery, which can increase the fetal morbidity and mortality. The average gestational age at delivery, as reported by Peng., et al. is 33 weeks [14].

In cases of antepartum hemorrhage, physicians should consider the differential diagnosis of cervical varices specially in presence of any of the risk factors such as: placenta previa, uterine overdistention or Diethylstilbestrol exposure.

In assessing the differential diagnosis, speculum vaginal examination in early gestational age can miss the diagnosis of cervical varices as shown in our case. Our first vaginal examination, done at 18 weeks, was not conclusive, and the second one done at 21 weeks showed a mesh of engorged smooth woven cord like structures.

In a context of sever antepartum hemorrhage along with the presence of placenta previa, the exact origin of bleeding, either from cervical varices or placental bed, can be difficult to establish, unless active continuous bleeding is seen in an appropriate setup, like operating theater, as it was shown in our case.

Also, cervical varices can emerge either from the endocervix or from the external part of the cervix as mentioned by Tanaka., *et al* [15]. These authors, in their review of 14 cases of cervical varices, have identified five cases where varices were emerging from the external part and four cases from the endocervix. In cases where cervical varices are emerging from the endocervix, speculum vaginal examination will show no abnormalities and could eventually miss the diagnosis.

On the other hand, the review of reported cases suggests to avoid any vaginal examination such as speculum examination, Papanicolaou smear, cervical biopsies or intercourse [14].

In view of all this, the best way to establish the diagnosis of cervical varices is by transvaginal ultrasonography with color and pulse Doppler [3,12-14]. In presence of placenta previa, physicians should localize the placenta precisely and look for any vessels in the surroundings such as vasa previa, and they should also screen the cervix to look for any cervical varices in the cervix or around it. This mapping of the placenta and cervix is best done by transvaginal ultrasonography, but it can also be done by abdominal probe [3].

Magnetic Resonance Imaging (MRI) is another tool that can be used in the diagnosis of cervical varices. MRI is more beneficial in equivocal cases or suspicion of abnormally adherent placenta [12]. Sagittal T2 views may be useful in imaging vascular dilatation in the cervix and can study the blood flow [12]. But it remains expensive and not always available in an emergency situation. In our case, Magnetic Resonance Imaging failed to identify vessels or varices in or around the cervix, maybe because of focusing on looking for an abnormally invasive placenta.

The management of ruptured cervical varices is not standardized. The rarity of cases reported makes it difficult to establish a clear plan of management or guidelines. The majority of authors agree on bed rest, pelvic rest (avoid intercourse, minimize speculum examination) and antenatal corticosteroids when fetal viability is achieved [2,8-10]. If cervical varices are discovered early in pregnancy and are not associated with significant bleeding, Chyjek., *et al.* in a case report, proposed emergency cervical cerclage in association with bed and pelvic rest [13]. In their case, cervical varices were discovered at 15 weeks of gestation and emergency cervical Mac Donald cerclage was placed. Delivery was achieved at 39 weeks.

However, the risk of severe hemorrhage, due to iatrogenic rupture of cervical varices during the cervical cerclage, and the absence of randomized trials in literature, where only case reports are available, makes it difficult to standardize cervical cerclage in these situations. More studies are required to recommend any antenatal interventions and weigh pros and cons.

In our case, active bleeding continued after completing the cesarean section and was only stopped once the ligation of varices was performed vaginally. It is possible that this ligation could be the treatment of active bleeding from cervical varices in case of antepartum hemorrhage. Once the origin of bleeding is confirmed to be from cervical varices, elective cervical varices ligation antenatally in a trained hands, could stop the bleeding, as demonstrated in our case, and probably allow pregnancy to continue till term. The major challenge in a context of cervical varices along with the presence of placenta previa is to clearly identify the origin of bleeding, from the placenta or the cervical varices.

In the review by Tanaka., *et al.* of nine case of cervical varices associated with placenta previa, the delivery was 37 weeks in three cases, 36 weeks in one case, 34 weeks in one case, 32 weeks in two cases and 27 weeks in one case [15]. The high incidence of preterm delivery in these cases is explained by the risk of severe antepartum hemorrhage leading to emergency cesarean section as reported in our case.

The role of the physician is to optimize the time of delivery whilst avoiding fetal or maternal compromise. Multidisciplinary meetings involving obstetricians, anesthesiologists, pediatricians, hematologists and even interventional radiologists is of high importance. Patient should be aware of all risks (preterm birth, emergency cesarean section, multiple blood transfusions) and the responsible treating team should be ready for intervention at appropriate time.

Among the 22 cases described in the literature, one case delivered vaginally at 31 weeks [8] and one had second trimester abortion at 17 weeks [16]. All the other cases had either elective or emergency cesarean section. Emergency cesarean section was also performed in our case. Cesarean section is the preferred mode of delivery in case of cervical varices in order to minimize vaginal manipulations and avoid any rupture of varices during the vaginal delivery.

Other modalities to control bleeding, such as uterine artery embolization, have been reported in the literature in certain conditions. In situation where termination of pregnancy is indicated in a non-viable fetus, Lesko., et al. used prophylactic uterine artery embolization in

a case of twins pregnancy presented with significant bleeding at 17 weeks [16]. In their case, after counselling, parents opted for termination of pregnancy and uterine evacuation was performed after uterine artery embolization. According to the authors, it helped to prevent significant blood loss during the intervention.

Also, uterine artery embolization was used immediately post cesarean section in the two cases presented by Chyjek., *et al* [13]. The blood loss in these two cases was 1000 ml.

We did consider prophylactic uterine artery catheter placement before planned cesarean section and planned to be discussed in future multidisciplinary meetings, but pregnancy was terminated at 26 weeks in a life threatening situation.

The evolution of cervical varices in the post-natal period seems to be good as varices tend to completely disappear. In the review of nine cases presented by Tanaka., *et al.* seven cervical varices have disappeared immediately after delivery, one after three days and another one after uterine artery embolization [15].

In our case this was confirmed by transvaginal ultrasonography done at six weeks after delivery showing normal looking cervix. This could be explained by the sudden drop of the venous pressure generated from the placental bed after delivery, and the collapse of cervical varices after ligation.

Conclusion

Cervical varices associated with placenta previa are rare and could be potentially life threatening. In these circumstances physicians should be aware of this association and they should diagnose cervical varices in order to avoid iatrogenic rupture or misleading diagnosis of bleeding placenta previa. Cervical mapping by transvaginal sonography is important in such cases to establish the origin of bleeding. Multidisciplinary team involvements are also of high importance and the treating team should be ready for massive hemorrhage or preterm delivery.

Elective cervical varices ligation could be an option in skilled hands in order to prolong the pregnancy, if diagnosis is made early.

Conflict of Interest

The authors declare no conflicts of interest or financial relationships related to this study.

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