



A Case of Adenomyosis during Pregnancy Requiring Cesarean Hysterectomy

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Abstract

Background: Adenomyosis occurs when endometrial tissue is found within the myometrium. The risks of adenomyosis in pregnancy are well described and include preterm delivery and preterm premature rupture of membranes, however only a few case reports describe the risks of delivery in the setting of extensive adenomyosis.

Case Presentation: We report the case of a 36-year-old G2P0010 at 38 week and 4 days gestation who presented in labor. The fetus was in the breech presentation so she underwent a primary Cesarean section. At the time of surgery, extensive transmural adenomyosis with decidualization was noted, ultimately requiring a Cesarean hysterectomy due to intra-operative hemorrhage.

Conclusion: This case emphasizes the importance of early clinical suspicion of adenomyosis in pregnancy to mitigate potentially life-threatening hemorrhage.

Keywords: Transmural adenomyosis; Cesarean hysterectomy; Obstetrical emergency

Introduction

Adenomyosis is defined as the presence of endometrial tissue within the myometrium. Phenotypically, adenomyosis commonly manifests as an enlarged, globular uterus that can result in heavy menstrual bleeding and dysmenorrhea but is often asymptomatic [1]. The true prevalence of the disease is difficult to ascertain because the definitive diagnosis can only be made at the time of histologic assessment of hysterectomy specimens. Nonetheless, prior studies have described adenomyosis rates up to 50% of uterine ultrasound studies in gynecology patients [2]. There is an association between adenomyosis and adverse pregnancy outcomes, including preterm delivery and Preterm Premature Rupture of Membranes (PPROM) [3]. However, reports regarding the implications of adenomyosis in term gestations and potential delivery complications are limited. We present a case of severe transmural adenomyosis at term requiring Cesarean hysterectomy at the time of delivery.

Case Presentation

A 36-year-old G2P0010 Asian woman at 38 weeks 4 days gestation presented via emergency medical transport with vaginal bleeding and contractions. She moved to the United States from China 6 weeks prior to presentation. The patient had not yet established prenatal care in the United States but reported regular prenatal care in China with dating confirmed by 3-month ultrasound. Her history was significant for a history of uterine fibroids and a dilation and curettage for a spontaneous abortion.

On presentation, she was in latent labor and reported the onset of vaginal bleeding 30 minutes prior. Ultrasound demonstrated a fetus in complete breech presentation and a right lateral placenta. Fetal heart tracing was category II with intermittent variable decelerations. The decision was made to proceed with Cesarean section. Upon entry into the abdominal cavity, approximately 200cc hemoperitoneum was noted. Additionally, an area of hypervascularity was noted on the right lateral wall of uterine corpus, with a normal appearing lower uterine segment. Given the concern for invasive placentation, an intra-operative consultation with Maternal Fetal Medicine and Gynecological Oncology was requested. A low transverse uterine incision was made and a male infant was delivered in footling breech presentation. Apgars were 8 and 9, birth weight 2940 grams, and arterial cord gases pH 7.27/O2 62.9/HCO3 28.2/BE -1. The placenta delivered spontaneously and intact without difficulty, and there was no evidence of invasive placentation.

After exteriorizing the uterus for closure of the hysterotomy, active bleeding was identified from

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Figure 1: Uterus with transmural adenomyosis.

The image on the right shows the uterine specimen anatomically oriented with posterior surface to the left and anterior to the right. The expanded globoid corpus can be appreciated in the center of the photo, and the adenomyosis as an ovoid, friable, hemorrhagic dark red transmurial lesion, on the serosal surface. The myometrium is best appreciated as whirled beige areas adjacent to the endometrium and extending to abut the serosal surface. The image on the left depicts the specimen after bivalving along the sagittal plane through the cervical Os and endometrial cavity. The endometrial cavity is seen in the center of the specimen as the dark red-brown, softened area extending in a triangular shape from apex to base, with the triangle's base toward the center of the specimen.



Figure 2: Cross section of focal adenomyosis.

A cross section shows a focus of adenomyosis located deep within the myometrium. The center of which is pink with an area of softening radiating outward. The texture can be appreciated by a smoother, more edematous and homogeneous appearance than the adjacent tan, firm, whirling cut surface of the uninvolved myometrium.



Figure 3: Cross section of transmural adenomyosis.

A cross section shows the specimen oriented with endometrial cavity superior, serosal surface inferior. Cross sections reveal mottled myometrium with pink-red areas of soft, fleshy changes and adjacent friable dark red-brown lesions, representing decidualized adenomyosis with new and old hemorrhage, respectively.

the serosal side of right the lateral and posterior uterine walls. These areas were hypervascular, friable and denuded. There was significant compromise of the right uterine wall with a suspected full-thickness defect. Uterine tone was normal but the cavity lesions and denuded areas were actively bleeding. The bilateral fallopian tubes appeared abnormally dilated and were adherent to the posterior and lateral aspects of the uterine wall, as were the ovaries. The rectosigmoid colon was also densely adherent to the posterior uterine wall. During the posterior lysis of adhesions, there was a significant amount of bleeding encountered from the abnormal vascularity and the denuded uterine sidewalls. We attempted conservative management by applying pressure as well as figure-of-eight sutures, however active bleeding continued. Aggressive resuscitation ensued as the patient became hemodynamically unstable and coagulopathic. She required a massive blood product transfusion consisting of 14 units of packed red blood cells, 2 packs of platelets, 2 pooled cryoprecipitate for 20 units of cryoprecipitate total, and 8 units of fresh frozen plasma. Given the lesions in the uterine wall and the degree of hemorrhage with hemodilutional coagulopathy, the decision was made to proceed with an emergent hysterectomy. The total estimated blood loss was 8,275cc.

Following surgery, the patient was transferred to the Intensive Care Unit. Her post-operative course was complicated by a right upper extremity deep venous thromboembolism requiring anticoagulation, followed by the formation of an incisional hematoma requiring evacuation. She was discharged home on post-operative day nine.

Pathological examination of the hysterectomy specimen revealed extensive, transmural decidualized adenomyosis with hemorrhage.

Gross pathologic assessment demonstrated a globoid shaped uterine corpus with thickened myometrial walls that were focally thinned with friable, hemorrhagic changes of overlying serosal surfaces (Figure 1). On cut sections, these focally thinned, friable areas along with other hemorrhagic areas within the myometrium were determined as extensive, transmural, decidualized adenomyosis (Figures 2 and 3). Representative sections of the specimen were submitted for microscopic evaluation (Figure 4), and no endometriosis was noted.

Discussion

Adenomyosis is defined as a benign, non-neoplastic invasion of endometrium into myometrium or endometrial gland-like tissue located within the myometrium. Microscopically this appears as ectopic endometrial glands and stroma surrounded by hypertrophic and hyper-plastic myometrium [1]. The edges of an adenomyosis lesion are not well defined or delineated from the surrounding myometrium. The presented case is unique from a pathologic perspective given the presence of exaggerated adenomyosis and the lack of coexisting endometriosis.

While adenomyosis is a disease commonly associated with menstrual complications, there are also pregnancy related risks. There are progesterone receptors on endometrial tissue and the elevated progesterone levels in pregnancy result in stromal decidualization of adenomyosis foci [1]. This heavily decidualized adenomyosis can then lead to uterine wall weakness, resulting in uterine rupture or uncontrolled bleeding during pregnancy. Thus, it is important to counsel patients regarding the potential implications of suspected adenomyosis where clinically relevant and to consider maternal-fetal outcomes.

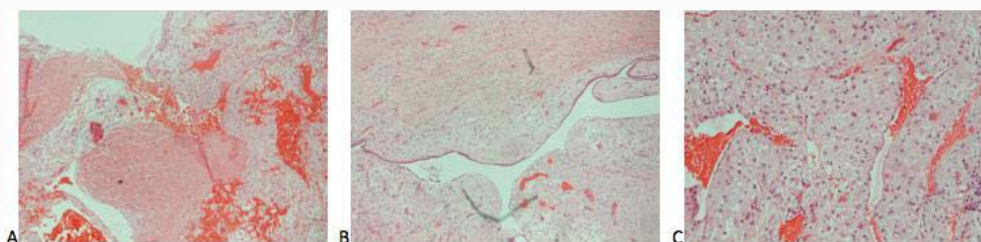


Figure 4: Decidualized adenomyosis, microscopic:

A- Adenomyosis with diffuse decidualization of the uterus, hyperplasia and hypertrophy of myometrial smooth muscle and fresh hemorrhage (4x).

B- Myometrial smooth muscle hyperplasia and haphazard whirling arrangement of myocyte bundles, as well as myocyte hypertrophy (10x).

C- Decidualized adenomyosis includes two cell types; epithelial glandular cells – polygonal with abundant pink cytoplasm, and supporting mesenchymal stromal cells- spindled, inconspicuously intervening between epithelial cells. Prominent interstitial edema identified between the stromal and epithelial cells (20x).

It is well established that adenomyosis is associated with poor obstetrical outcomes including increased rates of preterm delivery, premature rupture of membranes, fetal malpresentation, and cesarean section [3]. There are also case reports of adenomyosis presenting as uterine rupture, mostly preterm or in patients with previous uterine surgery [4,5]. The case presented here uniquely describes a case of transmural adenomyosis resulting in obstetrical hemorrhage due to disruption of the uterine myometrium. Areas of bleeding and myometrial disruption were histopathologically found to represent extensive transmural adenomyosis with marked decidual reaction extending from the endometrium to the uterine serosa.

Imaging can play an important role in the identification of adenomyosis and subsequent intrapartum management. Extensive literature exists on the use of transvaginal ultrasound and MRI to diagnose adenomyosis, with recent literature demonstrating only slightly increased accuracy in diagnosis with MRI, which is made comparable by 3D ultrasound imaging [6]. The most specific findings for adenomyosis on ultrasound are an enlarged globular uterus with sub-endometrial echogenic linear striations and myometrial cysts, with special attention paid to the junctional zone between the endometrium and myometrium. There is limited literature on imaging of adenomyosis in the antepartum period, likely because patients are unaware that they have the condition when they conceive. Early prenatal ultrasound is important, however, because it can be the first indication of a uterine abnormality which may be masked by the growing fetus later in pregnancy.

If adenomyosis is suspected prior to or during pregnancy, providers should have a high index of suspicion for bleeding complications secondary to potential decidualization of that tissue. This is especially true if hemoperitoneum or hyperdecidualization is noted at the time of Cesarean delivery, or if there is concern for intra-abdominal bleeding following vaginal delivery. Other common clinical risk factors such as menorrhagia, dysmenorrhea, advanced maternal age, multiparity, and prior uterine surgery are associated with adenomyosis and should therefore raise our index of suspicion for bleeding risk when caring for women during pregnancy. One study has quoted the rate of postpartum hemorrhage to have increased from 1.9 to 4.2 per 1000 deliveries from 1999-2008, with a doubling of the

rate of peripartum hysterectomy [7]. Furthermore, according to the Center for Disease Control, the pregnancy-related mortality ratios were 15.9 and 17.3 deaths per 100,000 live births in 2012 and 2013 respectively, increased from 10.0 in 1990 [8]. Although the etiologies for the overall rising maternal morbidity and mortality rates in the United States are not clearly identified, careful consideration of risk factors and evaluation of abnormal imaging findings becomes especially important in mitigating catastrophic hemorrhage.

Despite an unexpected hemorrhage and hysterectomy followed by multiple post-operative complications, this case resulted in a healthy mother and infant who were able to return home to their family in China.

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