Case Reports

Spontaneous uterine rupture in a nulligravida female presenting with unexplained recurrent hematometra

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Summary

Spontaneous rupture of an unscarred uterus in reproductive-age women is exceedingly rare, especially in the context of dysfunctional bleeding and a patent cervical canal. A 25-year-old nulligravida female, who reported recent onset of metromenorrhagia and anemia, was initially admitted for surgical management of unexplained hematometra requiring dilation and curettage. The patient remained with intermittent vaginal bleeding for the following six months on continuous progestin therapy. She then re-presented with enlarged hematometra and uterine rupture, which was surgically repaired. Despite exhaustive conservative treatment to preserve fertility, hysterectomy was eventually required due to recurrent uterine rupture. Idiopathic recurrent hematometra can result from the rare combination of uncontrolled dysfunctional bleeding and absence of outflow obstruction.

Key words: Uterine rupture; Unexplained hematometra; Hysterectomy.

Introduction

Hematometra refers to retrograde accumulation of the menstrual blood in the uterine cavity usually as result of lower genital tract obstruction, most commonly at the level of the cervical canal and involving iatrogenic, infectious or malignant etiologies [1]. It is rare for a hematometra to be present in the absence of an identifiable cause. In a pre-puberal girl, unexplained hematometra has been reported in an initially obstructed and overdistended hemiuterus despite its surgical correction [2]. However, persistence of hematometra in the absence of outflow obstruction is exceedingly rare in a young nulliparous female. The following is a description and discussion of an unusual case of uterine rupture in a young female with unexplained recurrent hematometra.

Case Report

A 25-year-old nulligravida female presented to the emergency department with a three-month history of intermittent heavy vaginal bleeding, new onset of dizziness, and fever. The patient reported a previously regular 28-day interval menses since menarche at age 13 and denied any prior history of sexual activity. Her past medical history was significant for migraines with aura. A transvaginal ultrasound (TVS) depicted a distended blood-filled uterine cavity measuring 8.4 x 6.8 x 7.6 cm, consistent with the diagnosis of hematometra. Intravenous antibiotics were initiated for presumed endometritis, and she underwent an uncomplicated dilation and curettage (D&C) where approximately 200 cc of dark blood was drained from the uterine cavity. Interestingly, the cervical canal was dilated quite easily and a vaginal exam revealed blood from the cervical canal prior to the procedure. Blood transfusion was given for persistent anemia. The final pathology of the specimen revealed only a late secretory endometrium. Since the patient's history of migraines with aura contraindicated the use of estrogen therapy, depot medroxyprogesterone acetate was recommended as treatment for her dysfunctional uterine bleeding (DUB). During the following six months, the patient remained with controlled but intermittent vaginal bleeding. A follow-up pelvic examination one month later revealed a normal size uterus. Two weeks after the third dose of depot medroxyprogesterone acetate, the patient was readmitted to the hospital with lower abdominal pain and uterine bleeding. A pelvic computed tomography (CT) scan revealed a 7.7 x 7.5 x 5 cm blood-filled uterine cavity with a markedly thin uterine wall. A pelvic magnetic resonance imaging (MRI) scan confirmed a 4 cm uterine wall rupture with blood in the peritoneal cavity (Figure 1). The patient underwent an exploratory laparotomy to repair the uterine defect and drainage of intrauterine blood clots. A bleeding diatheses work-up was completely within normal limits. Leuprolide acetate was started in an attempt to cease her menstrual function. A few days following hospital discharge, the patient was readmitted to the hospital with recurring hematometra, an enlarged uterus approximating 16 weeks in size, and an imminent uterine rupture as depicted by MRI. The options of hypogastric artery ligation or selective uterine arterial embolization were discussed, but they were deferred due to the patient's desire for future fertility and for conservative management. Diagnostic laparoscopy revealed an intact uterus without free fluid in the abdomen. Under laparoscopic guidance, the cervix was easily dilated followed by evacuation of 300 cc of intrauterine blood clots. Again no apparent mechanical outflow obstruction was found. A normal pelvic arteriography excluded the possibility of vascular malformation as the cause of the uncontrolled bleeding. After considerable discussion of the benefits of estrogen administration and the risk of stroke in a young woman with severe and uncontrolled DUB and history of migraine with aura, the decision was made to proceed with intravenous equine conjugated estrogen administration. After 24 hours of intravenous estrogen therapy, no improvement in uterine bleeding was noted, as reflected by a progressively enlarging uterus and by a significant reduction in hematocrit. Images from a repeat MRI

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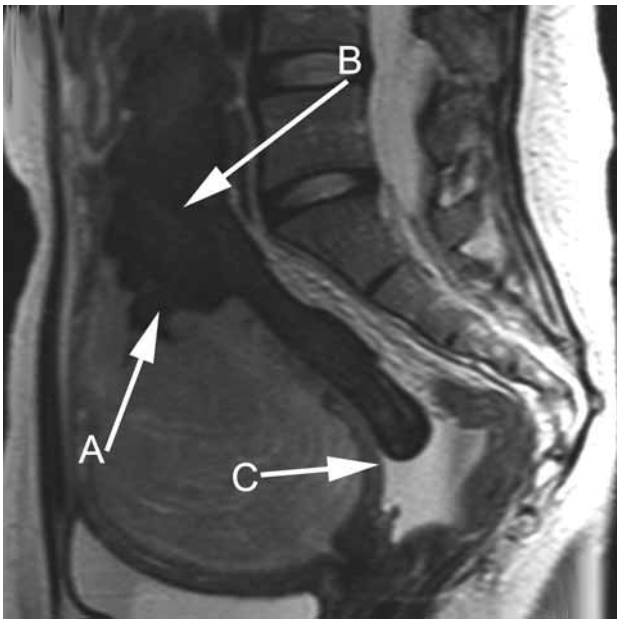

Figure 1. — T2 weighted magnetic resonance imaging depicted a markedly distended uterus filled with blood. Arrow A indicates the site of uterine wall rupture in the fundal area. Arrow B indicates blood clots extravasating into the abdominal cavity, and arrow C indicates the markedly thinned myometrium.

scan were consistent with recurrent uterine rupture. Furthermore, developing fever and increasing lower abdominal tenderness were consistent with endometritis. The patient received a blood transfusion followed by a supracervical hysterectomy. The final pathology revealed a 4.5 x 1.5 cm uterine wall defect with extensive myometrial edema and hemorrhage. Chronic endometritis and adenomyosis were also present.

Discussion

The etiology of the unexplained hematometra formation in the patient described remains a mystery, especially given the previous history of normal menses since menarche and recent onset of self-perceived metromenorrhagia. It is plausible that the suspected endometritis, discovered during an earlier admission, might have resulted in cervical obstruction or sub-occlusion since endometritis has been suggested as a possible etiology for cervical obstruction and acute hematometra formation in a recent report [1]. However, this would not have justified the subsequent recurrent events. Other speculative but possible etiologies include: obstruction of the endocervical canal with debris, blood clots or endometrial tissue resulting in outflow obstruction and retention of menstrual blood within the uterine cavity. Similar to the patient we presented here, uncontrolled uterine bleeding, secondary either to dysfunctional uterine bleeding or intrauterine arterial malformation, have been reported to result in large hematometra in postmenopausal women and young women, respectively [3, 4]. The question remains whether in the setting of severe uterine bleeding, the hematometra can still be formed despite a patent cervical canal. Unfortunately, a definite conclusion can not be drawn solely based on isolated case reports.

The most intriguing aspect of this case was the rapid reaccumulation of the blood in the uterine cavity despite the drainage of hematometra and advanced mechanical cervical dilation. Since no identifiable causes of outflow obstruction could be recognized, we suggested that the over-distended uterine cavity might have resulted in permanent functional impairment leading to hypocontractility of the myometrium and inability to expulse retained blood in the uterine cavity. The analogy can be compared to a deflated balloon unable to return to the initial maximum contractile state. The patient's history of persistent vaginal bleeding and the observation of an easy cervical dilation corroborated the absence of outflow obstruction. Donnez *et al*. have recently reported a case of didelphic uterus with an obstructed hemivagina resulting in ipsilateral hematocolpos and hematometra. Despite the resection of the vaginal septum, hematometra recurred in the hemiuterus in the absence of mechanical obstruction. The same author has suggested a probable loss of contractility of the myometrium and absence of recovery of normal uterine cavity after over distension as the cause of recurrent hematometra. If the hypothesis is proven to be correct, due to the potential damage of the myometrium with an over-distended uterine cavity, early detection and treatment of a large hematometra might be a logical approach to prevent its recurrence. Further studies are needed to confirm the speculation.

Uterine rupture in a non-gravid and unscarred uterus is exceedingly rare. Spontaneous uterine rupture has been reported in young nulligravida women presenting with arterial malformations [3]. Previous reports have acknowledged an association between uterine rupture and adenomyosis, with the inference that characteristic cyclic changes from these "invaded endometrial glands and stroma" may result in chronic inflammation, hemorrhage, and tissue necrosis of the native myometrial layer. This may lead to compromised tissue integrity and consequent myometrial tearing [5]. Although adenomyosis was found in the uterine specimen of the patient presented here, there is currently not enough evidence to establish a direct causal association between adenomyosis and uterine rupture. Theoretically, the intermittent uterine wall distension might have predisposed the infiltration of endometrial cells deep into the myometrium, thus accounting for the adenomyosis lesion found within the myometrium in the case described. While the plausible explanation for the uterine rupture might be obvious – excessive intraluminal pressure exceeding the distensible capacity to weaken and thin the myometrium leading to its rupture – the underlying mechanism leading to large hematometra formation remains elusive in the absence of evident lower genital tract obstruction.

Estrogen therapy is the mainstay in the treatment of life-threatening or uncontrolled DUB in women of reproductive age. Given the patient's history of migraines with aura, estrogen therapy was medically contraindicated due

to the presumed increased risk of stroke. Evidence from a meta-analysis of six case-control studies suggested that patients with migraines who use estrogen therapy were two to four times more likely to have an ischemic stroke as compared to non-user migraine patients; the risk is even greater when migraines are accompanied by aura [6]. While there is consensus to avoid estrogen therapy in women of reproductive age with migraines, the controversy remains as to whether or not estrogen therapy, when medically indicated, could be safely used in selective young healthy females having migraines with aura [7]. Although speculative, it is possible that early intervention with intravenous estrogen could have ameliorated the patient's uncontrolled DUB and prevented the progressively enlarging hematometra formation leading to uterine rupture. Traditionally, progestin has been successfully used as an alternate to estrogen for the control of DUB when estrogen is medically contraindicated. Intermittent vaginal bleeding might occur while on progesterone therapy due to endometrial atrophy, but rarely does uncontrolled and persistent bleeding requiring multiple blood transfusions occur, as is presented here.

The physiopathology behind the uncontrolled bleeding of our patient is unclear. The patient reported here failed to respond to pharmacological suppression of the endometrium and the uterine bleeding continued despite the use of estrogen, progesterone or GnRH agonist. A similar experience was reported by Donnez *et al*., to prevent hematometra recurrence in one of the hemiuteri before the definitive surgical removal [2]. Shreikant *et al*. reported a similar unsuccessful experience to control the endometrial bleeding in a postmenopausal woman presenting with recurrent hematometra using a high dose of medroxiprogesterone acetate [4]. Whether a different pharmacological regimen would be more effective in preventing the recurrence of unexplained hematometra is currently unknown. In our patient, hysterectomy was eventually indicated due to the uncontrolled bleeding and ascending pelvic infection. The optimal treatment for hematometra in the absence of mechanical obstruction remains an area for further exploration.

Conclusion

In summary, this case illustrated persistent hematometra in a young nulligravida woman in the absence of any evident mechanical obstruction of the lower genital tract. The challenging aspect of the management was the ineffectiveness of a conservative approach with subsequent development of recurrent hematometra in a young patient who desired fertility preservation. The uncontrolled and recurrent nature of her uterine pathology led to uterine rupture and eventually necessitated hysterectomy as the definitive treatment. The optimal treatment for the unexplained hematometra deserves further investigation.

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