

Puerperal spontaneous prolapse of large pedunculated uterine submucosal myoma after full-term vaginal delivery

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Summary

Complications of submucosal myomas during pregnancy are rare and can be divided into those occurring during pregnancy, at delivery, and in the puerperium. The authors present two similar cases of unusual complication of submucosal uterine myoma in the puerperium. The two patients were admitted to this department for protruding vaginal mass after full-term vaginal delivery. Each experienced sudden severe abdominal pain, and felt a large vaginal mass after defecation or coughing. Combining gynecological examination and MRI, the authors accurately diagnosed the cases as spontaneous prolapse of large pedunculated uterine submucosal myoma. After anti-infective therapy, the 45-year-old woman underwent total laparoscopic hysterectomy; the second case, a 31-year-old woman was treated by a conservative surgical approach, including partial vaginal myomectomy and elective hysteroscopic myomectomy. This is the first report describing spontaneous prolapse of pedunculated uterine submucosal myoma in the puerperium after full-term vaginal delivery, and MRI played a key role in the diagnosis and treatment.

Key words: Uterine; Submucosal myoma; Prolapse; MRI.

Introduction

Uterine myoma is the most common benign tumor of the female genital tract. A prospective study found myoma prevalence during pregnancy to be around 10.7% [1]. Uterine myoma is associated with numerous pregnancy complications, including premature delivery, placental abruption, abnormal fetal position, and postpartum hemorrhage [2, 3]. Due to their special location, submucosal myomas more easily induce pregnancy and postpartum complications in women of reproductive age than intramural and sub-serosal myomas. Patients have even received hysterectomy for severe postpartum hemorrhage caused by submucosal uterine leiomyoma to avoid a life-threatening condition [4].

Here the authors present two cases of large pedunculated uterine submucosal myoma with local infection that spontaneously prolapsed into the vaginal cavity after full-term vaginal delivery. The report was approved by the local ethics committee. To the best of the present authors' knowledge, no similar case has been described previously in the literature.

Case Report

Case 1

A 45-year-old woman, gravida 1 para 1, presented with a protruding vaginal mass after full-term vaginal delivery. The patient had regular menses, with the last one on September 2, 2015. At five weeks pregnancy, ultrasound indicated an intramural fibroid

measuring 6.3 cm in the posterior wall. The pregnancy was smooth without pain or other special complications. At 38 weeks pregnancy, she received forceps delivery to help the healthy baby due to fetal distress. The placenta was spontaneously delivered, without postpartum hemorrhage. At the beginning, the puerperium was all without incidence, with normal lochia and no abdominal pain. Twenty days after delivery, the patient had sudden lower abdominal pain without fever or nausea and vomiting; however, lochia amounts increased to menstrual blood loss volume. These symptoms gradually decreased after three days without treatment. Fourteen days after abdominal pain onset, she felt a perineal mass which hardly regressed after stool. When she arrived to the present department, gynecologic examination revealed the presence of a large vaginal mass measuring 6 cm in diameter, covered with pus moss and protruding from the cervix. Palpation of uterus fundus was unsatisfactory for thick fat of the abdominal wall. Hematologic investigation revealed white blood cell count and hemoglobin within the normal ranges (8460/mm³ and 14.5 g/dl, respectively). Differential diagnosis of prolapsed submucosal or chronic uterine inversion was made. Ultrasound imaging revealed a uterine size of 9.6×7.8×6.6 cm; there was an 8.7×6.5×6.2 cm non-homogeneous mass in the lower part of the posterior wall (Figure 1). Sagittal T1-weighted MRI with contrast enhancement showed the mass had a wide pedicle connected to the posterior wall; the prolapsed vaginal mass portion showing diffuse homogeneous low signal intensity without enhancement, indicative of necrosis (Figure 2). The mass was diagnosed as a prolapsed large submucosal myoma with infection. The patient did not desire further pregnancies and feared myoma recurrence. After three days of anti-infective therapy, total laparoscopic hysterectomy and bilateral salpingectomy under general anesthesia was performed. Pathological data confirmed submucosal leiomyoma with red degeneration and active growth, with no evidence of malignancy. The patient recovered uneventfully, and was discharged in a sta-

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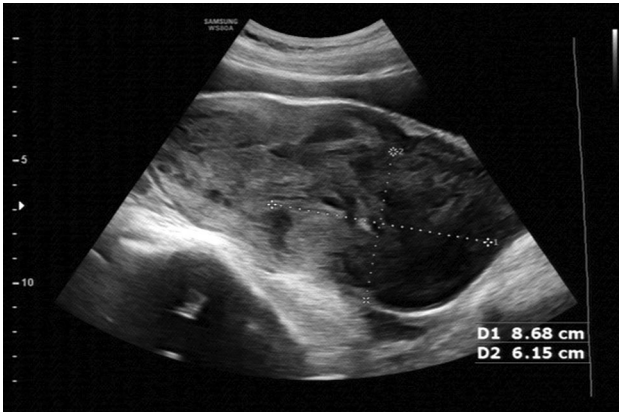


Figure 1. — Ultrasound imaging of the myoma in a 45-year-old woman.

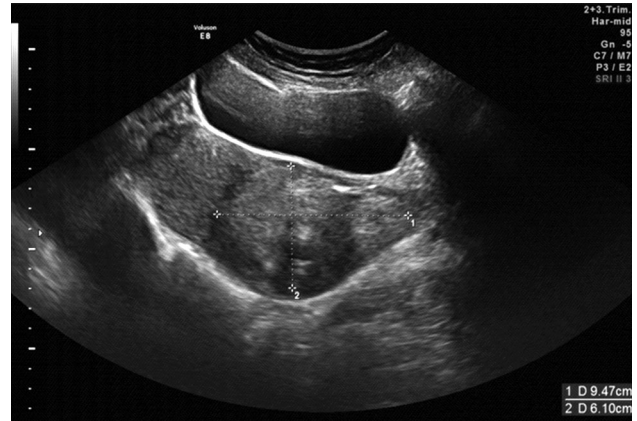


Figure 3. — Ultrasound imaging of the myoma in a 31-year-old woman.



Figure 2. — Sagittal T1-weighted MR image with contrast enhancement of the prolapsed submucosal myoma in a 45-year-old woman.



Figure 4. — Sagittal T1-weighted MR image with contrast enhancement of the prolapsed submucosal myoma in a 31-year-old woman.

ble condition five days after surgery.

Case 2

A 31-year-old woman, gravida 1 para 1, visited the present department for protruding vaginal mass after full-term vaginal delivery. The patient had regular menstrual periods with the last menstruation on October 30, 2015. Pre-pregnancy examination found an intramural fibroid of 5 cm in diameter in the anterior wall. Without treatment, she became pregnant, and the pregnancy process was smooth. She underwent full-term natural vaginal delivery of a healthy baby at the local hospital 50 days prior. She had no postpartum hemorrhage, and the lochia disappeared two weeks after delivery. Twenty days post-delivery, she had sudden lower abdominal pain after a severe cough, and felt a spherical tumor in the vagina. Abdominal pain was self-mitigated without

fever or vaginal bleeding; she did not consult a doctor because of fear. In the last ten days, the patient had a malodorous smell that even she could not stand. Gynecologic examination revealed that a protruding vaginal mass covered with pus moss had prolapsed out of the cervix. Hemoglobin was 9.2 g/dl, for white blood cell count of 6840/mm³, and C-reactive protein of 18.0 mg/L. Ultrasonic imaging revealed a 9.5×6.4×6.1-cm non-homogeneous mass originating from the lower endometrial cavity (Figure 3). MRI showed the same results as the above case, except that the mass was connected to the anterior wall of the uterine cavity and larger (Figure 4). Based on these data and the experience from the above patient, the lesion was also diagnosed as submucosal uterine fibroid associated with infection. After three days of anti-infective therapy, transvaginal myomectomy was carried out under general anesthesia. During the operation, the vaginal part of the mass was



Figure 5. — Gross picture of the prolapsed submucosal myoma from a 31-year-old woman.

found to be about 10×10 cm (Figure 5), and it was difficult to expose the cervix. The visible part was cut with cold knife, suturing the tumor wound to stanch bleeding. Ultrasound monitoring suggested the residual tumor in the lower uterine cavity was 5×6 cm (Figure 6). Considering that the myoma was too large and still associated with infection, hysteroscopic surgery was not appropriate for possible induction of uterine infection and transurethral resection of the prostate syndrome. The patient was administered three cycles of GnRH agonists to reduce the remaining part of the myoma. Afterward, elective hysteroscopic surgery was performed to completely remove the remaining fibroid without any complication (Figure 7). Histopathological examination confirmed the diagnosis of a submucosal leiomyoma with red degeneration. The patient was discharged the day after surgery.

Discussion

Myomas are the most common uterine benign tumors, and occur in approximately 20% to 30% of women of reproductive age [5]. However, a study assessing hysterectomy specimens revealed myomas in approximately 74% of samples from premenopausal women [6]. Based on the uterine layer affected, myomas are categorized as submucosal, intramural, and sub-serosal. Submucosal myomas whose prevalence ranges from 5% to 8% [7, 8], are those in contact with, or distorting the endometrial cavity. Due to their special location, submucosal myomas usually cause

more adverse effects on fertility and are more easily associated with pregnancy complications and adverse obstetric outcomes, including subfertility, miscarriage, preterm labor, intrauterine growth restriction, and postpartum hemorrhage. Mounting evidence suggests that hysteroscopic resection results in improved outcomes in such conditions [9, 10]. Submucosal myomas easily form peduncle and as foreign bodies in the uterine cavity, often cause uterine contraction to be squeezed out of the cervix into the vagina or perineum in the non-puerperal period. However, pedunculated large uterine submucosal myomas with local infection that spontaneously prolapse into the vagina in the puerperium are scarcely reported. To the present authors' knowledge, this is the first report presenting this complication of submucosal myomas.

The above two patients had regular menses without abnormal uterine bleeding. Before pregnancy or in the first trimester, ultrasonography indicated the existence of inter-mural myomas measuring 5×6 cm in diameter, which is very common in women of reproductive age. However, it is extremely rare that such intramural myomas grow to submucosal myomas and prolapse out of the uterine cavity into the vagina. The mechanism remains unclear, and the authors speculate that it might be related to cervical relaxation, thin uterine walls, and rapid myoma growth during



Figure 6. — Ultrasound imaging of the residual tumor in the lower uterine cavity in a 31-year-old woman.

pregnancy and in the puerperium [11-13]. Sudden increase of abdominal pressure might be a predisposing factor, such as severe cough or hard stool, as observed in the above patients. In MRI data, submucosal myomas were clearly located in the lower part of the uterine cavity, and this special location might also play an important role.

It is well known that uterine inversion can present as a protruding mass in the vagina in the puerperium. Uterine inversion is a rare condition classified as puerperal and non-puerperal. Puerperal uterine inversion is more common, and occurs immediately after the third stage of labor, or within 24 hours post-partum; it is often associated with trauma while forcing the umbilical cord or the endometrial fundus, when the placenta is not stripped [14]. Non-puerperal uterine inversion occurs more than four weeks post-delivery, and is mostly related to benign myomas and seldom associated with malignancy after 45 years [13]. It is suspected when gynecologic examination detects a protruding mass in the vagina or vulva, with the uterine fundus not palpated by bimanual examination [15]. In addition, uterine inversion has characteristic ultrasonography images which reveal a hyperechoic mass in the vagina, with a central hypoechoic H-shaped cavity in transverse images and a U-shaped groove from the fundus to the center of the inverted part in longitudinal images [16, 17].

Although palpation of uterine fundus was unsatisfactory in the current patients for thick fat of the abdominal wall, ultrasonography indicated intact uterine fundus and normal uterine outline. Combined with patient history, the present authors made the diagnosis of prolapse of large pedunculated uterine submucosal myoma, which was the basis for subsequent surgical plans.

Few reports have described the use of MRI findings for the diagnosis of prolapsed submucosal leiomyomas. MRI does not only accurately delineate the pedicle and uterine attachment of the prolapsed myoma, but is also very help-

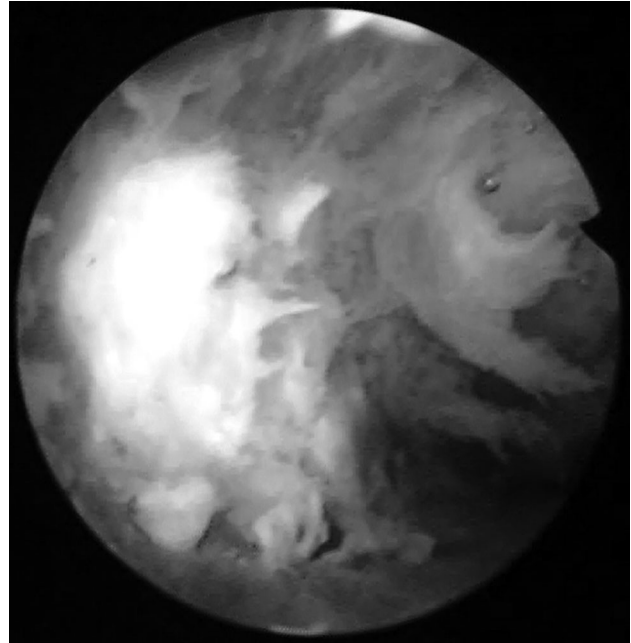


Figure 7. — Hysteroscopic view of the residual tumor after three cycles of GnRH agonist treatment in a 31-year-old woman.

ful in distinguishing malignant tumors, which provides clinicians with important information in planning the surgical approach [18, 19].

For treatment, the present authors believe hysterectomy and myomectomy are both suitable, and the final surgical approach should be based on patient age, requirements for reproduction, and wishes of retaining the uterus [20, 21]. In this study, the first patient was 45-year-old; she did not desire further pregnancies and was very fearful of fibroid recurrence, so laparoscopic hysterectomy was performed after anti-infective treatment. The second patient was only 31-years-old, and a conservation treatment program was adopted as follows. First, the vaginal part of the tumor (which was necrotic) was removed to control infection. Then, she was administered three cycles of GnRH agonists to reduce the remaining part of the myoma. Finally, elective hysteroscopic myomectomy was performed to remove the remaining part. In both patients, the postoperative course was uneventful without any complications.

Conclusion

The authors reported two cases of puerperal spontaneous prolapse of pedunculated uterine submucosal myoma, which is extremely rare after full-term vaginal delivery. The differential diagnosis of non-puerperal uterine inversion and malignancy must be performed. MRI was helpful in reaching the correct diagnosis preoperatively and planning an appropriate surgical approach. After anti-infective treatment, hysterectomy or myomectomy could be considered,

with the final surgical approach depending on patient age, requirements for reproduction, and wishes of retaining the uterus.

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References

- [1] Laughlin S.K., Baird D.D., Savitz D.A., Herring A.H., Hartmann K.E.: "Prevalence of uterine leiomyomas in the first trimester of pregnancy: an ultrasound-screening study". *Obstet Gynecol.*, 2009, 113, 630.
- [2] Somigliana E., Vercellini P., Daguati R., Pasin R., De Giorgi O., Crosignani P.G.: "Fibroids and female reproduction: a critical analysis of the evidence". *Hum. Reprod. Update*, 2007, 13, 465.
- [3] Klatsky P.C., Tran N.D., Caughey A.B., Fujimoto V.Y.: "Fibroids and reproductive outcomes: a systematic literature review from conception to delivery". *Am. J. Obstet. Gynecol.*, 2008, 198, 357.
- [4] Akrivis C.H., Varras M., Bellou A., Kitsiou E., Stefanaki S., Antoniou N.: "Primary postpartum haemorrhage due to a large submucosal nonpedunculated uterine leiomyoma: a case report and review of the literature". *Clin. Exp. Obstet. Gynecol.*, 2003, 30, 156.
- [5] Cook H., Ezzati M., Segars J.H., McCarthy K.: "The Impact of Uterine Leiomyomas on Reproductive Outcomes". *Minerva Gynecol.*, 2010, 62, 225.
- [6] Cramer S.F., Patel A.: "The frequency of uterine leiomyomas". *Am. J. Clin. Pathol.*, 2002, 94, 435.
- [7] Novak E.R., Woodruff J.D.: "Myoma and other benign tumors of the Uterus". *Gynecol. Obstet. Pathol.*, 1979, 8, 260.
- [8] Fedele L., Bianchi S., Dorta M., Brioschi D., Zanotti F., Vercellini P.: "Transvaginal ultrasonography versus hysteroscopy in the diagnosis of uterine submucous myomas". *Obstet. Gynecol.*, 1991, 77, 745.
- [9] Carranza-Mamane B., Havelock J., Hemmings R., Reproductive Endocrinology and Infertility Committee, Cheung A., Sierra S., et al.: "The management of uterine fibroids in women with otherwise unexplained infertility". *J. Obstet. Gynaecol. Can.*, 2015, 37, 277.
- [10] Litta P., Conte L., De Marchi F., Saccardi C., Angioni S.: "Pregnancy outcome after hysteroscopic myomectomy". *Gynecol. Endocrinol.*, 2014, 30, 149.
- [11] Lascarides E., Cohen M.: "Surgical management of nonpuerperal inversion of the uterus". *Obstet. Gynecol.*, 1968, 32, 376.
- [12] Krenning R.A., Dorr P.J., de Groot W.H., de Goey W.B.: "Non-puerperal uterine inversion. Case report". *Br. J. Obstet. Gynaecol.*, 1982, 89, 247.
- [13] Marjolijn de Vries., Denise Arlette Maria Perquin.: "Non-puerperal uterine inversion due to submucous myoma in a young woman: a case report". *J. Med. Case Rep.*, 2010, 4, 21.
- [14] Poon S.S., Chean C.S., Barclay P., Soltan A.: "Acute complete uterine inversion after controlled cord traction of placenta following vaginal delivery: a case report". *Clin. Case Rep.*, 2016, 10, 699.
- [15] Witteveen T., van Stralen G., Zwart J.: "Puerperal uterine inversion in the Netherlands: a nationwide cohort study". *Acta Obstet. Gynecol. Scand.*, 2013, 92, 334.
- [16] Hsieh T.T., Lee J.D.: "Sonographic findings in acute puerperal uterine Inversion". *J. Clin. Ultrasound*, 1991, 19, 306.
- [17] Ali E., Kumar M.: "Chronic Uterine Inversion Presenting as a Painless Vaginal Mass at 6 Months Post Partum: A Case Report". *J. Clin. Diagn. Res.*, 2016, 10, QD07.
- [18] Panageas E., Kier R., McCauley T.R., McCarthy S.: "Submucosal uterine leiomyomas: diagnosis of prolapse into the cervix and vagina based on MR imaging". *AJR Am. J. Roentgenol.*, 1992, 159, 555.
- [19] Kim J.W., Lee C.H., Kim K.A., Park C.M.: "Spontaneous prolapse of pedunculated uterine submucosal leiomyoma: usefulness of broccoli sign on CT and MR imaging". *Clin. Imaging*, 2008, 32, 233.
- [20] Doherty L., Mutlu L., Sinclair D., Taylor H.: "Uterine fibroids: clinical manifestations and contemporary management". *Reprod. Sci.*, 2014, 21, 1067.
- [21] Nair S.: "Contemporary management of fibroids". *Ann. Acad. Med. Singapore*, 2003, 32, 615.

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