

An interesting case of large multi-leiomyomas in uterine cervix with concomitant pregnancy and review

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Summary

The incidence of uterine leiomyoma in women aging from 35 to 50 years is reported to be 70–80%. Generally, leiomyoma is classified according to the locations and directions of growth, and the most common types including subserosal, intramural, and submucous myoma. Only about 0.4% of leiomyoma develops in the cervix of uterus, and it would be even rare to find a cervical leiomyoma with a size similar to a full-term uterus, as reported here, which poses a challenging situation for gynecologists.

Key words: Cervical leiomyoma; Uterine myoma.

Introduction

Uterine leiomyoma in women aging from 35 to 50 years is reported to be 70–80% [1]. Leiomyoma is generally classified according to the locations and directions of growth, and the most common types including subserosal, intramural, and submucous myoma. Only about 0.4% of leiomyoma develops in the cervix of uterus [2]. An even more rare finding would be a cervical leiomyoma with a size similar to a full-term uterus. The authors report such a case which poses a clinical challenge for gynecologists.

Case Report

A 43-year-old, G6P2 woman, visited her primary physician for 12 weeks' amenorrhea. The primary findings included uterine leiomyoma and a large pelvic mass. Amenorrhea was caused by the unexpected pregnancy. The woman was then referred to the authors' hospital for further treatment. She reported a history of non-symptomatic uterine leiomyoma for seven years, and the latest ultrasound follow-up was performed three years ago, which showed an intramural leiomyoma (about 6×7 cm in size). One year ago, the patient felt abdominal distension and this symptom was deteriorating since then; however, no intervention was given. The cervix was not exposed on routine vaginal examination. On abdominal examination, a plump abdomen with a size similar to a full-term pregnancy was noted. No tenderness or pain was reported during palpation, and the bottom of the uterus reach the xiphoid. The woman's serum β -hCG was 22,909.4 mIU/ml and hemoglobin was 92 g/L. The tumor makers, including CA19-9, CA125, CEA, and AFP were within the normal range. Routine blood biochemical tests revealed no abnormality either. Ultrasound imaging revealed a large mass within the abdominal-pelvic cavity with close relationship to the uterus (Figure 1). A few liquid areas and abundant bloodstream signal were noted in the

Doppler scanning. Transvaginal ultrasonography also revealed a 3.9×2.1×4.5 cm fertilized egg in the uterine cavity, and an intramural hypoechoic nodule (8.9×5.4×8.1 cm in size) located in the left-lateral wall of the uterus. Gigantic uterine leiomyoma was highly expected, and curettage was not possible, therefore exploratory laparotomy was scheduled directly. Exploratory laparotomy revealed an enlarged uterus, dwelling on a huge abdominopelvic mass. This factual tumor below the posterior pelvic peritoneum bilaterally developed to pelvic sidewalls, extending from the pelvic floor to the diaphragm. It was smooth and had a clear boundary between neighboring structures. Another two similar masses originated from cervix were also noted, with the size of 6×7 cm and 15×15 cm, respectively. The anterior one pushed the bladder upwards while the right lateral one was within the broad ligament, which was stretched to the pelvic sidewall. The right broad ligament was opened and a extruded ureter was noted. The adhered ureter was gently separated from the broad ligament by fingers and smooth forceps. Then the mass was entirely removed which was confirmed to be leiomyoma by intraoperative pathological test. Due to the large size of the abdominal-pelvic masses, the surgical field was poorly exposed and therefore the surgical procedure was difficult. Since this current pregnancy was completely unexpected for this patient, and she had no intention to have anymore children, finally hysterectomy and bilateral salpingectomy were performed, which could decrease the blood loss and prevent injuries to the neighboring organs/structures. It is worth noting that there was an extensive parametrial venous plexus, which connected the posterior cervix to this large myoma via a vascular peduncle (Figure 2). This thick peduncle was sutured and ligated repeatedly, but the bulky vessels within the peduncle still caused massive blood loss. The hemoglobin level dropped to 62 g/L and three units of red cell suspension was transfused to correct anemia. Then the abdominal aorta and inferior vena cava was check by a specialized vascular surgeon and all the abdominal and pelvic organs were kept intact. Finally a drainage tube was left in the pelvis for early detection of postoperative hemorrhage. The removed leiomyoma weighed 4,500

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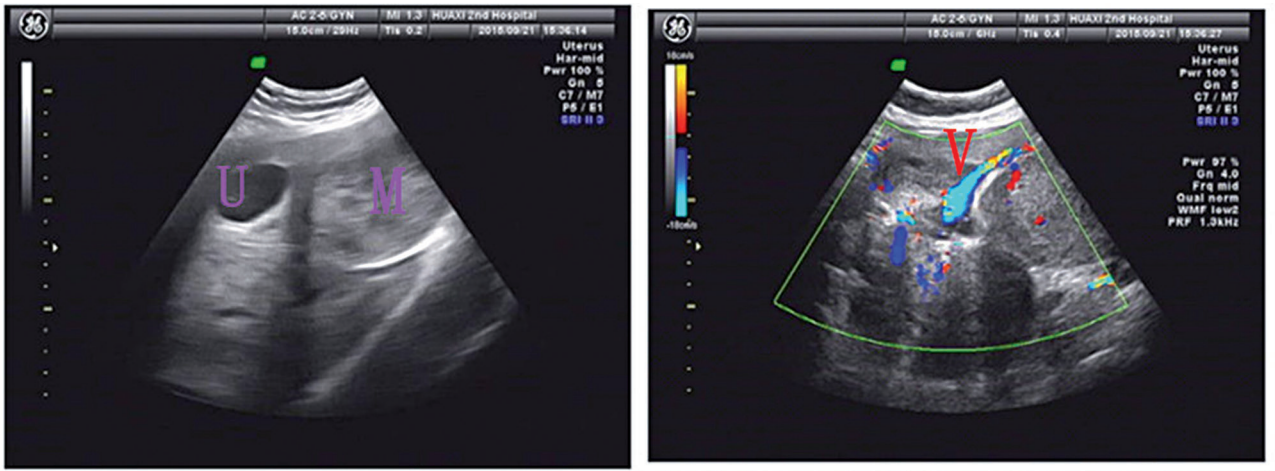


Figure 1. — Transvaginal ultrasonography and Doppler findings (U: uterus, M: mass, and V: vascularity).

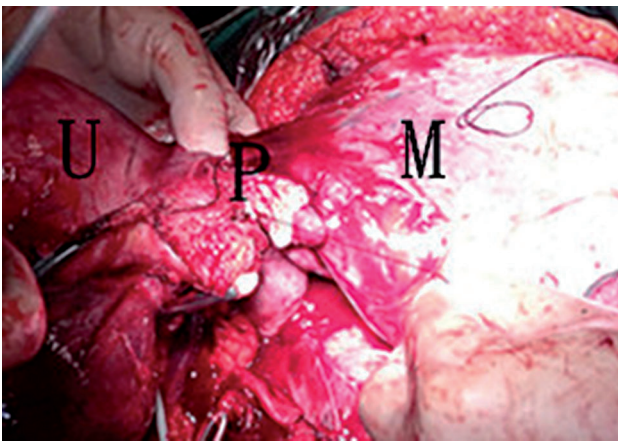


Figure 2. — Peduncle connecting the posterior of cervix to this large myoma (p: peduncle, U: uterus, and M: mass).

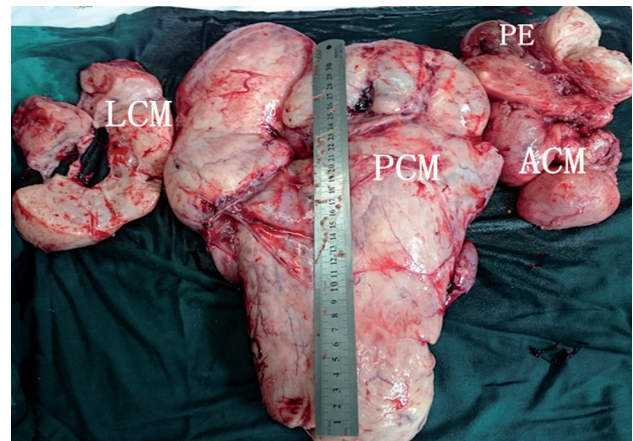


Figure 3. — Complete resection: lateral cervical myoma (LCM), posterior cervical myoma (PCM), and anterior cervical myoma (ACM).

grams (Figure 3) and large areas of myxoid degeneration and interior bleeding inside were found. Postoperatively, the patient received another three units of red blood cells and recovered well, and was discharged on postoperative day 4. The postoperative pathology confirmed leiomyoma with degeneration and placental-villous tissues, and the immunohistochemical result showed that most cells were positive for caldesmon +++ and negative for CD10. Less than 1% cells stained positive for Ki67.

Discussion

The proliferation of leiomyoma cell depends on estrogen and progesterone [4], which may contribute to the large cervical leiomyoma found in this pregnant female. In previously reported cases, large cervical fibroids were misdiagnosed as ovarian tumors [3, 4], but in the present case the preoperative diagnosis highly reminds of benign

tumor based on age, history, presentations, physical examination, blood and imaging tests, and then exploratory laparotomy which rapidly and directly confirms the nature and origin of this mass. MRI or CT was not taken for the limited accuracy in differential diagnosis.

Matsuoka *et al.* classified cervical leiomyoma into two types: extracervical and intracervical [5], while Chang *et al.* advocated a classification of five types, according to the surgical technique and location [6]. Anterior cervical myoma can lead to frequent urination; the posterior myoma might cause constipation and the myoma expanding laterally to broad ligament might compress the ureter and uterine artery [7]. Moreover, other non-specific symptoms may present, such as menstrual abnormalities, dyspareunia, and postcoital bleeding [6, 8]. Rarely, incarcerated procidentia has also been reported [9]. Furthermore, cervical fibroid

may have influence on fertility [10]. Surprisingly, the present patient reported none of the aforementioned symptoms but abdominal distension and even conceived unexpectedly in spite of such huge myomas.

The treatment for each patient with cervical leiomyoma should be individualized, as recommended in the guidelines. At present conservative therapies such as uterine artery embolization (UAE), gonadotropin-releasing hormone (GnRH) agonists, and magnetic resonance-guided focused ultrasound are optional, but may have limited effect on larger-sized or highly-multiple leiomyoma. Therefore, surgical approach, such as myomectomy or hysterectomy, remains the leading treatment for symptomatic leiomyoma [11]. Considering mainly infeasible curettage, the symptoms, size of mass, patient's age, further fertility plan, and an open hysterectomy instead of myomectomy was planned for this patient.

It is sometimes difficult to avoid injuries to surrounding structures when handling cervical myoma and the estimated blood loss is often much larger than removing myoma in other location. Performing laparoscopic myomectomy for small size has been discussed before [5, 6], but clearly it was not suitable for cervical myoma with excessive growth. Temporary balloon occlusion of the bilateral internal iliac artery combined with laparoscopic myomectomy has also been reported in a nulligravida with large cervical myoma [12], which have not been placed into common use. Several points summarized by the present case are worthy of mention: 1) regarding diagnostic difficulty in abdominopelvic mass, large cervical leiomyoma should be taken into consideration, especially for possibly benign tumor; 2) overgrowth of myoma is always accompanied by rich vascularity, hence sufficient blood product should be available; 3) except history and examinations, exploratory findings combined with intraoperative pathology can help quickly confirm uncertain diagnosis of mass, then an individual operative procedure; 4) as such large and multiple cervical myomas, position of the ureter undoubtedly should be the most essential step for reluctant ureter injury, meanwhile gentle separation of the surroundings is also important for any other injuries; 5) the hysterectomy, instead of myomectomy, should be the first choice, which could facilitate exposure of the surgical field and decrease intraoperative blood loss. Finally, as retroperitoneal myoma from cervix, vascular surgeon should be consulted to prevent and/or repair unexpected vascular injuries.

Such large multiple myomas in the cervix are rarely found in a pregnant female. In addition with medical his-

tory-taking and physical examinations, exploratory laparotomy combined with intraoperative pathology may be a preferred treatment for large masses occupying the abdominopelvis. Furthermore, the procedure should be individualized based on intraoperative findings, and special attention should be paid to minimize blood loss and avoid injuring important neighboring structures such as ureters and blood vessels.

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