CEOG Clinical and Experimental Obstetrics & Gynecology

Iatrogenic parasitic myoma on the peritoneum of the right pelvic wall

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Summary

Uterine myoma, the most common form of uterine tumor, occurs in approximately 25% of reproductive-aged women. Parasitic myoma, which outgrows its uterine blood supply and obtains a secondary blood supply from another organ such as the omentum, is rare. It is extremely rare if it is on the peritoneum of the right pelvic wall. Only a few cases have been found in this location so far. Here, the authors report an interesting case of parasitic myoma on the peritoneum of the right pelvic wall. They conclude with seven key points, which should be paid more attention to avoid iatrogenic parasitic myoma.

Key words: Parasitic myoma; Iatrogenic parasitic myoma.

Introduction

Uterine myoma, which is the most common form of uterine tumor, occurs in approximately 25% of reproductive-aged women [1]. Parasitic myoma is a rare type of pedunculated subserosal myoma, which is partially or completely separated from the uterus and receives alternative blood supply from another source such as the omentum and mesenteric vessels [2, 3]. There are three theories regarding the pathogenesis of parasitic myomas, and the most prevalent one is iatrogenic cause. Most patients initially have non-specific symptoms including pelvic pain, dyspareunia, or abnormal vaginal bleeding. A diagnosis of parasitic myomas is often an incidental event to an operation for concomitant myomas. They can sometimes cause secondary symptoms, which depends on their location [4]. Further examinations such as measurement of CA 125 concentration and MRI are usually performed. The authors report an interesting case of parasitic myoma on the peritoneum of the right pelvic wall. They conclude with seven points below, which should be paid more attention to: 1) systematic survey the entire cavity, 2) complete removal of tissue pieces, 3) maintain awareness of the potential risk, 4) irrigate the abdomen and pelvis, 5) exclusion of malignancy is mandatory, 6) include parasitic myoma in the differential diagnosis and 7) long-term follow-up.

Case Report

A 33-year-old woman, gravida 6, para 1, came to the authors' clinic in August 2013, complaining of pain in the abdomen for two days. She had undergone a laparoscopic myomectomy at another medical institution in 2007. She did not use contraception regularly. She used emergency contraceptive pill (levonorgestrel 0.75 mg) six to seven times per year. Clinical examination revealed a bulky uterus with a mass in the pouch of Douglas but separate from uterus. Ultrasound examination revealed a hypoechoic solid mass of 7.3×6.0×4.2 cm in the Pouch of Douglas but separate from uterus (Figures 1A, 1B). MRI: a lesion in the Pouch of Douglas. Low signal intensity on T2WI images (Figures 2A, 2E), intermediate signal intensity on T1WI images (Figure 2B), high signal intensity on DWI images (Figure 2C), and homogeneous enhancement (Figure 2D) were found. The signal intensity of the lesion in enhancement phase was lower than that of uterus muscle. Concentrations of CA125, AFP, CEA, and CA199 were normal. Papanicolaou smear was normal. The patient was adequately informed of the possible risks and benefits of laparoscopic surgery and signed a written consent agreeing to undergo laparoscopic exploration. The case was approved by the Institutional Review Board of the Peking University First Hospital. laparoscopic myomectomy was performed. In addition, a large parasitic myoma (approximately seven cm in diameter) was found attached to posterior peritoneum (Figures 3A, 3B), and the pedicle division seemed to be on the posterior uterine wall (Figure 4). Myomectomy was performed – first coagulating and then cutting the pedicle of the myoma. A small second parasitic myoma (approximately three cm in diameter) was found attached to the left mesosalpinx (Figure 5), which was also removed laparoscopically. The pelvic mass weighed 140 grams. The tumor was removed using a morcellator. Pathological diagnosis was leiomyoma with hyaline de-

Revised manuscript accepted for publication April 22, 2015

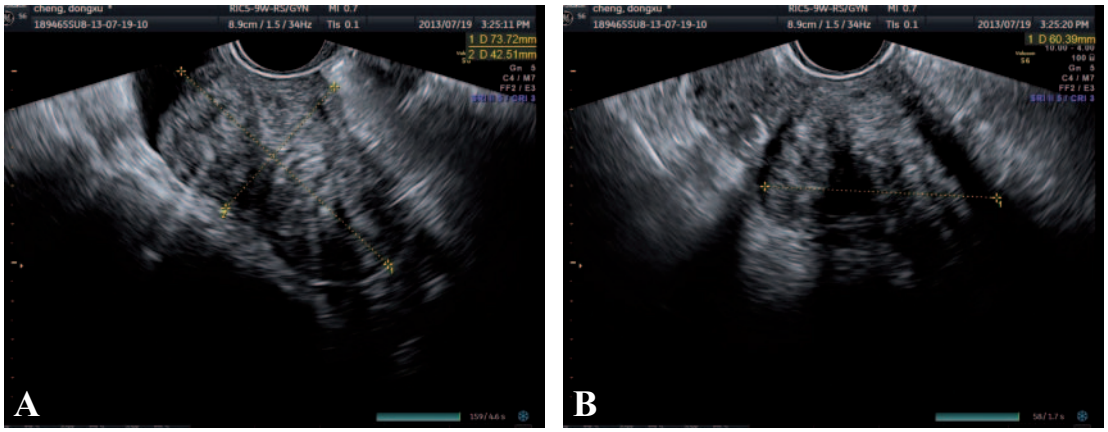

Figure 1. — A mass of 7.3 cm (A) ×6.0 cm (B) ×4.2 cm (A) in the Pouch of Douglas but separate from uterus there is a hypoechoic solid tumor.

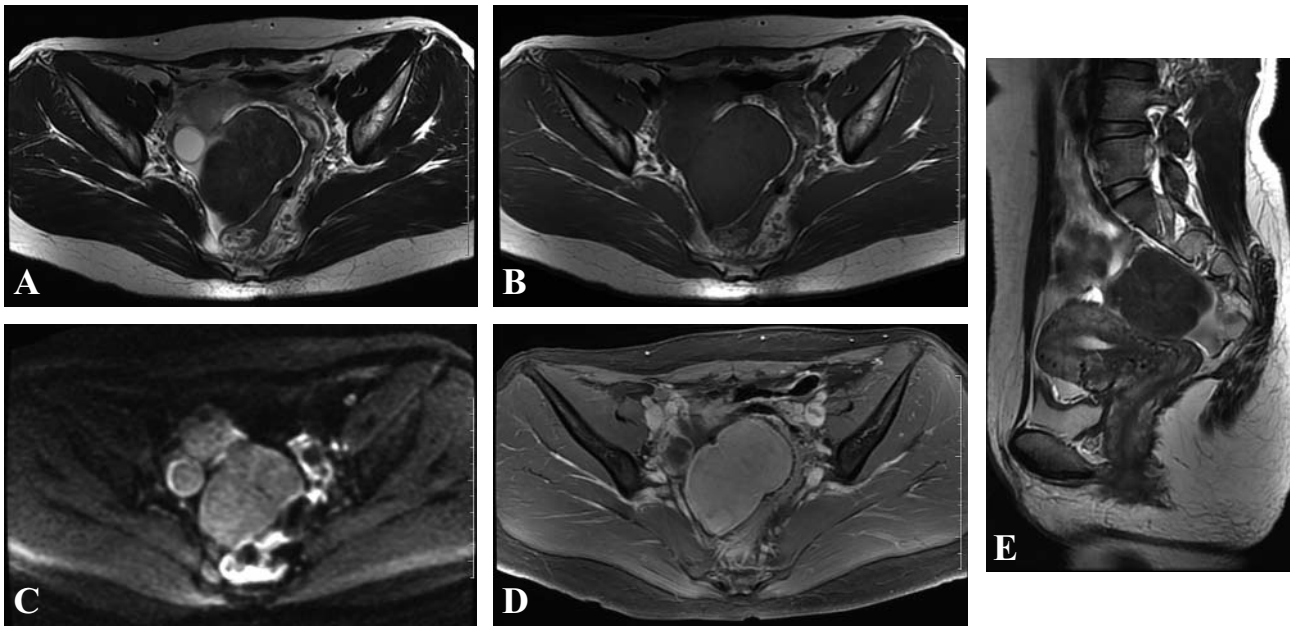

Figure 2. — A lesion in the Pouch of Douglas. Low signal intensity on T2WI images (A, E), intermediate signal intensity on T1WI images (B), high signal intensity on DWI images (C), and homogeneous enhancement (D) were found. The signal intensity of the lesion in enhancement phase was lower than that of uterus muscle.

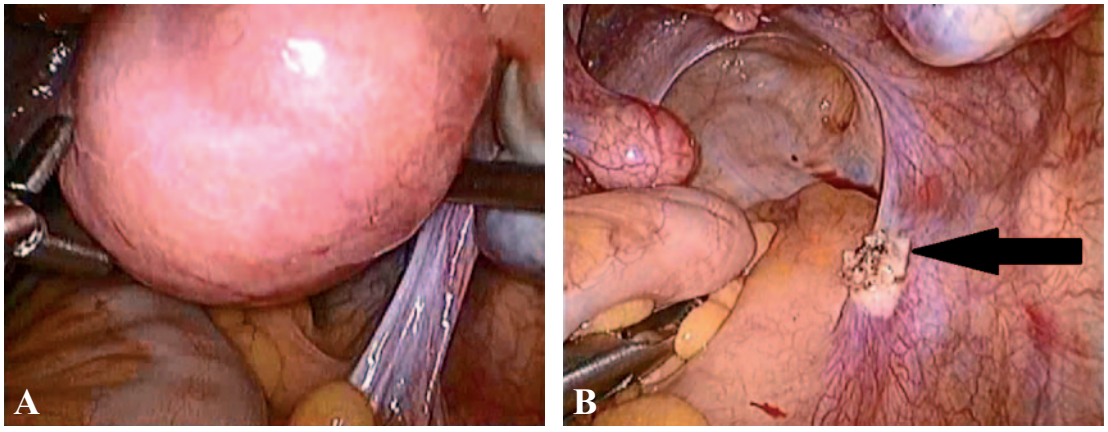

Figure 3. — (A) A large parasitic myoma (approximately 7 cm in diameter) was found attached to posterior peritoneum. (B) After cutting the pedicle of the myoma. The arrow shows the pedicle attached to posterior peritoneum.

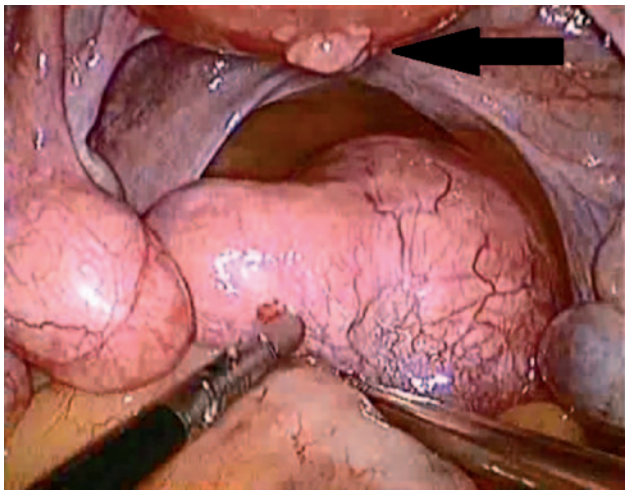

Figure 4. — The arrow shows the pedicle division, which seems to be on the posterior uterine wall.

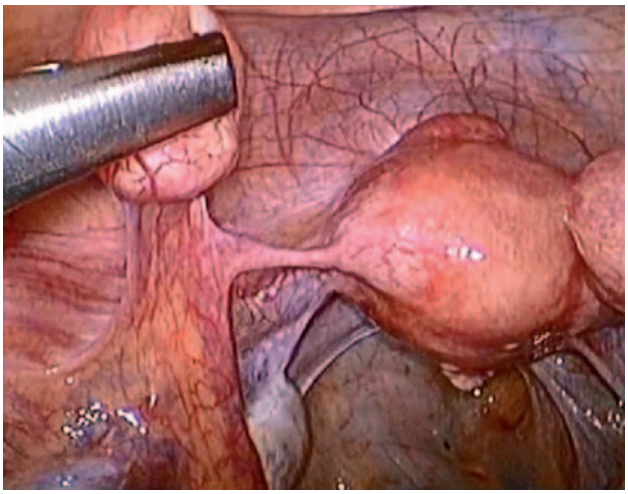

Figure 5. — A small second parasitic myoma (approximately three cm in diameter) is found attached to the left mesosalpinx.

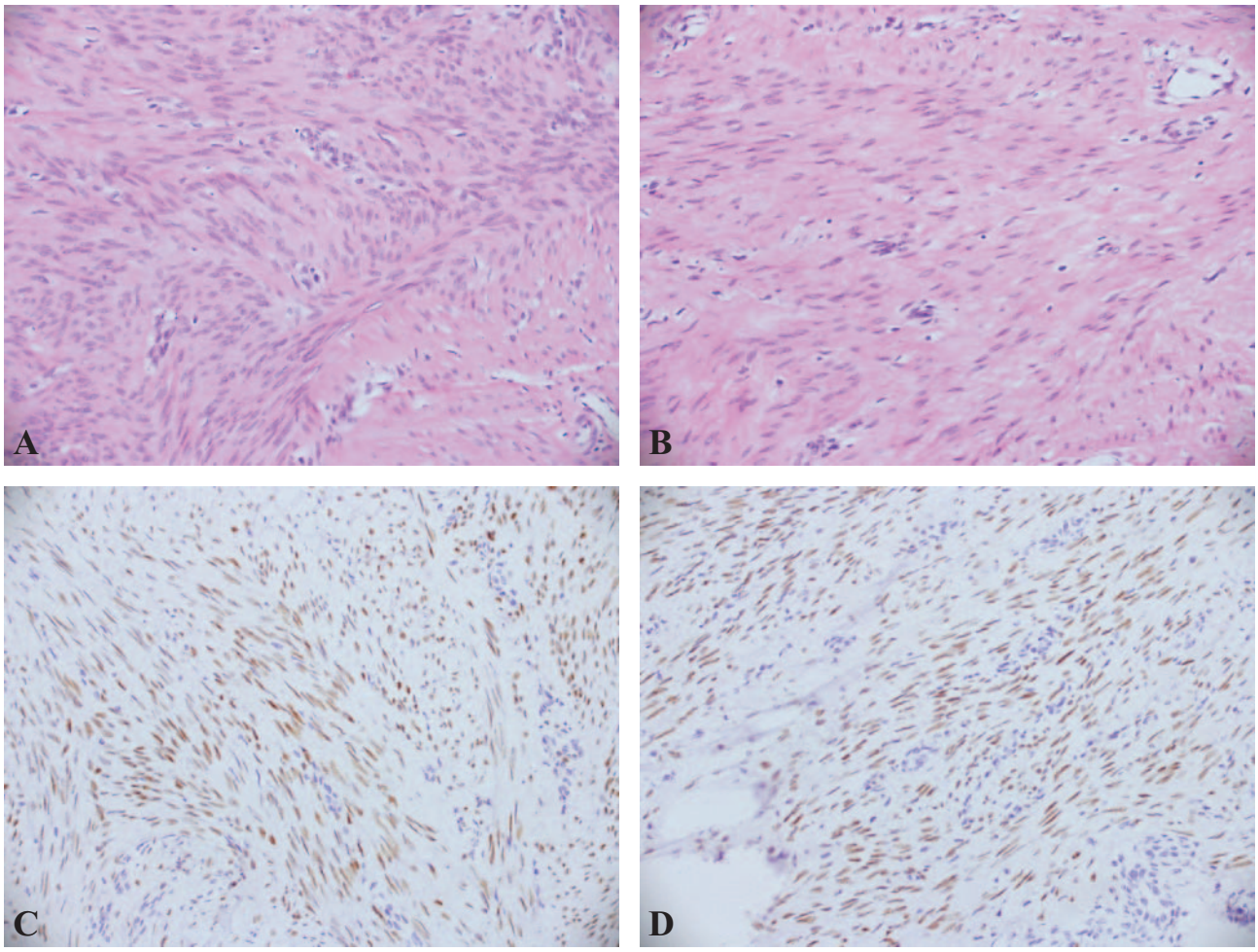

Figure 6. — (A) Intersecting fascicles of cytologically bland, spindled cells with cigar-shaped nuclei and eosinophilic cytoplasm (HE ×20). (B) An area with hyaline degeneration (HE ×20). The spindled cells of leiomyoma express ER (C) and PR (D) diffusely.

generation, positive for estrogen receptor, and progesterone receptor (ER 90%++, PR90%++) (Figures 6A-6D). The patient had an unremarkable postoperative period and is asymptomatic to date.

Discussion

Parasitic myoma is a rare type of pedunculated subserosal myoma, which is partially or completely separated from the uterus and receives alternative blood supply from another source such as the omentum and mesenteric vessels [2, 3]. Although the exact mechanisms for the subsequent growth of parasitic myoma are not well understood [5], it seems reasonable to hypothesize that exposure of retained myoma fragments to sex steroid hormones and/or growth factors may result in the growth of parasitic myoma later [5]. Parasitic myomas were found most frequently parasitizing to the omentum, and second most frequently parasitizing to the sigmoid colon [4, 6-14]. However, the present authors report an interesting case of parasitic myoma on the peritoneum of the right pelvic wall, with positive estrogen and progesterone receptors (ER 90%++, PR90%++). Moreover, she used emergency contraceptive pill six to seven times per year.

Parasitic myoma, which seems to be a rare disorder developing from the natural history of pedunculated myomas, has become increasingly reported over the last decade [4, 6-14]. Parasitic myomas were described for the first time by Kelly and Cullen [15]. There are three theories regarding the pathogenesis of parasitic myomas. The first hypothesis is that parasitic myoma spontaneously develops from pedunculated myoma, and it needs more blood supply to grow and to maintain itself, so that it adheres to its surrounding structures and separates itself from the uterus. The second hypothesis is that parasitic myoma is associated with previous uterine surgery. During the morcellation of myomas, small particles of myoma may be left in the pelvic, which may implant in normal tissues [4]. The third hypothesis is that parasitic myoma is associated with restriction of blood supply to the uterus. Therapeutic administration of gonadotropin-releasing hormone agonists restricting blood supply is effective in treating the symptoms of a myomatous uterus. Similarly, other minimally invasive procedures such as uterine artery embolization and MR(f)US are also used to manage symptomatic myomas. Some researchers suggested that parasitic myomas developed after these two kinds of treatments [14, 16]. Parasitic myoma is considered iatrogenic disease in the last two hypotheses.

Most patients have non-specific initial symptoms including pelvic pain, dyspareunia or abnormal vaginal bleeding. Diagnosis of parasitic myomas is often an incidental event to an operation for concomitant myomas, such as the present second parasitic myoma attached to the left mesosalpinx. They can sometimes cause secondary symptoms, which depends on their location [4]. Parasitic myomas occur at various sites, including the port site [7], intestines, peritoneum, and omentum in the abdominal cavity. A history of hysterectomy, myomectomy or presence of concurrent uterine myomas may support the diagnosis in many cases; however, usually the finding of a heterogeneous pelvic mass at transvaginal ultrasonography warrants exclusion of malignancy, and further examinations such measurement of CA125 concentration and MRI are usually performed. Due to its multiplanar detection capability, MRI is the most reliable technique in such cases. It can accurately demonstrate the location of the tumor relative to adjacent structures such as the ureter, bladder, or rectum, which is critical for surgical planning. Typical leiomyomas demonstrate low to intermediate signal intensity on T1-weighted images and low signal intensity on T2-weighted images. Myxoid degeneration and necrosis may be visible as high signal intensity areas on T2-weighted images. Another common variant form seen on both T1- and T2-weighted images is a cobblestone-like appearance caused by hyaline degeneration, with high-signal intensity foci representing areas of infarction caused by rapid growth [17]. The present patient's MRI showed low signal intensity on T2WI images, intermediate signal intensity on T1WI images, high signal intensity on DWI images, and homogeneous enhancement. Serum CA125 in patients with parasitic myoma is normal or slightly increased. The present patient's serum CA125 was normal. Ghamande *et al.* [18] reported a patient with parasitic myoma presenting a CA125 level of 1,539 U/ml. The patient underwent an exploratory laparotomy and resection of a parasitic fibroid following which the CA125 levels decreased and normalized within a month. Ascites are rare symptoms of parasite myoma. Kebapci *et al.* [19] reported a patient with parasitic myoma presenting massive ascites. Ascite and pleural effusion disappeared six months after surgery.

In the last ten years, there has been an increase in the reports of parasitic myomas in the literature, and they are found to be largely associated with previous surgeries. The increasing report of incidence of iatrogenic parasitic myomas in the literature might be caused by increased use of minimally invasive surgery. Advances in laparoscopic techniques and instrumentations have enabled physicians to avert laparotomy in many cases, resulting in practical benefits over conventional surgeries. Since the 1990s, the laparoscopic approach has gained popularity, and currently, total hysterectomy [20], subtotal hysterectomy [21], and myomectomy [22] are frequently performed by laparoscopy in many medical institutions around the world. Laparoscopic myomectomy is usually a feasible option, irrespective of size, site or number of the myomas [23]. However, some long-term complications after morcellation of the uterus have recently been described in the literature [4, 6-14]. There is even a report of a uterine leiomyoma particle growing in an abdominal wall incision [24], and another reported case of parasitic myoma under the dome of the diaphragm [25]. Tissue morcellation, especially in large myomas, may be very time-consuming, therefore, tissue pieces may spread in the abdominal cavity. Morcellation enables removal of large specimens at laparoscopy, however, even using a morcellation bag [26], there is still risk for incomplete removal. According to a recent report, retained

fragments occur in 0.57% of subtotal hysterectomies [11]. This is especially true in obese patients in whom the visceral fat makes inspection of the peritoneal cavity more difficult. These retained fragments usually become infarcted and cause abdominal pain, which necessitates immediate removal of the mass. There have been reports of retained tissue becoming necrotic and causing severe peritonitis [27]. In the present case, the patient had a history of myomectomy. In terms of the iatrogenic pathogenesis of parasitic myomas, it is considered that fragments from incomplete removal of a myoma during the previous operation could have implanted in the posterior peritoneum and formed the stalk.

The present authors conclude the following seven key points below, which should be paid more attention to: First of all, systematic surveying of the entire cavity with meticulous surgical technique is necessary. Second, all tissue pieces that are morcellated should be completely removed. Even small bits displaced into the upper abdomen can result in parasitic fibroids [25]. Third, it is important for us as physicians and surgeons to maintain awareness of this potential risk to the patients. Fourth, placing the patient in reverse Trendelenburg position after morcellation and repeatedly irrigating the abdomen and pelvis with normal saline may be helpful to wash out small pieces in pelvis [6]. After intraperitoneal lavage, a thorough examination of the abdomen and pelvis should be performed and all the remaining tissues should be removed. Fifth, if morcellation is anticipated or required, exclusion of malignancy is mandatory. In all patients who are at risk of uterine malignancy (e.g., postmenopausal or with intermenstrual bleeding) and will undergo the surgical procedure, uterine biopsy and cervical cytologic analysis should be performed before surgery to reduce risk of possible spread after morcellation of a malignant tumor, despite the possibilities of false-negative result or other potential causes leading to the patient's symptoms. Sixth, in patients who have pelvic masses with a history of morcellation, iatrogenic parasitic myomas should be considered in the differential diagnosis [28]. Seventh, it is necessary to perform a long-term follow-up.

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