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Giant cellular leiomyoma in the broad ligament of the uterus: a case report

Jiao Yan, Yu Li, Xingyu Long, Daocheng Li, Sijin Li

Abstract

BACKGROUND

The treatment of large pelvic masses in postmenopausal women is a challenge for clinical practice. Although ultrasound or magnetic resonance imaging (MRI) can be used to determine size and location of the mass, it is still hard to achieve a preoperative diagnosis. The majority of cellular leiomyomas are diagnosed histopathologically after surgery. We reviewed the differential diagnosis and surgical management of a rare case of cellular leiomyoma in the broad ligament of the uterus.

CASE SUMMARY

A 52-year-old Chinese woman without sexual history was admitted to the First Affiliated Hospital of Guangzhou University of Chinese Medicine for the first time. The patient had a 1-year history of progressive abdominal enlargement as well as a 2-year history of menopause, and complained of frequent abdominal pain and low-grade fever. Computed tomography (CT) of abdomen showed a solid cystic mass (29.4 cm × 18.8 cm × 37.7 cm) in the pelvis and abdomen. Moreover, the blood routine test result indicated a baseline cancer antigen 125 (CA-125) level of 187.7 U/mL and C-reactive protein of 109.58 mg/L. Subsequently, retrograde hysterectomy and bilateral adnexectomy were performed on the patient. On the histopathologic examination of the sample extracted from the patient, the result suggested that this patient suffered from a rare cellular leiomyoma in the broad ligament.

CONCLUSION

Clinicians need to constantly improve diagnosis and treatment for the great challenges posed on clinical assessment, differential diagnosis, and surgical management.

INTRODUCTION

Cellular leiomyoma arising in the broad ligament of the uterus is extremely rare, with only five cases reported to date. Furthermore, the preoperative diagnosis of cellular leiomyoma poses a great challenge for clinicians. On the one hand, its clinical presentation is similar to that of ovarian cancer, such as enlargement of abdominal masses and increased tumor markers (CA125) after the menopause [1]. On the other hand, all cellular leiomyomas in the broad ligaments were diagnosed by intraoperative observation and postoperative pathology. In our study, the sixth case of giant cellular leiomyoma in the broad ligament with cystic and mucinous lesions was reported.

CASE PRESENTATION

Chief complaints

Recurrent abdominal distension for 1 year, accompanied by left upper abdominal pain for 4 days.

History of present illness

By virtue of low-grade fever and abdominal pain, the woman was admitted to the emergency department of the First Affiliated Hospital of Guangzhou University of Chinese Medicine for 4 days.

History of past illness

Except for congenital neurofibroma accompanied with local pigmentation throughout the body and prominent skin with scattered small masses, she hadn't specific previous medical history.

Personal and family history

The patient, unmarried and childless, was a 52-year-old Chinese postmenopausal woman with an asexual life history. She hadn't drug history or family history. Besides, the patient had no history of smoking or drinking.

Physical examination

Abdominal examination discovered an abdominal mass with a size corresponding to the abdominal size at gestation of 37 wk (located below xiphoid process). Anal examination result revealed a large solid cystic mass with well-defined borders in the pelvic abdomen.

Laboratory examinations

The elevated cancer antigen 125 (CA125, a baseline level of 187.7 U/mL) level.

Imaging examinations

As for the computed tomography (CT) result, a solid cystic mass (29.4 cm × 18.8 cm × 37.7 cm) was also found in the pelvis and abdomen.

FINAL DIAGNOSIS

Right broad ligament uterine leiomyoma

TREATMENT

The patient was successfully subjected to retrograde abdominal hysterectomy and bilateral adnexectomy.

OUTCOME AND FOLLOW-UP

The samples were sent for histopathological examination, and the results included that the uterine masses were composed of proliferating smooth muscle cells and arranged in

a braided shape; the cell morphology was slightly changed and no abnormality was observed.

DISCUSSION

Leiomyoma, the most common benign tumor in the uterus [2], presents no obvious symptoms in many women with fibromas. Only patients who have symptoms affecting the function of the tumor-adjacent organ, such as urinary frequency and urgency, constipation, pain, or cancer-related anemia will opt to visit the clinic. Given the complex pathogenesis of uterine leiomyoma, the etiology of uterine leiomyoma remains unclear at present. As revealed by the results of clinical observations and laboratory tests, both estrogen and progesterone are involved in regulating the growth of uterine fibroids [3]. In addition, sex hormone level is an essential contributor to uterine fibroids. Interpretatively, several studies have uncovered that the incidence of hormone-dependent uterine leiomyoma is about 70% in women of childbearing age [4] and 80% in 50-year-old women [5]. Uterine fibroids almost form inside the uterus and rarely outside of it. In terms of their histomorphological features, uterine fibroids are mainly classified into intramural fibroids, submembranous fibroids, and submucosal fibroids [6]. Among these histopathological forms, cellular leiomyoma in the broad ligament is the least common type of uterine fibroid (< 5%) [7, 8]. The broad ligament contains embryonic remnants formed by mesonephric (wolffian) ducts. Notably, wolffian ducts give rise to the genital ducts in males, but contract to generate embryonic remnants. These remnants including epithelial and paracellular cells, may spread and form abnormal cysts, or produce some myofibers that can lead to leiomyomas within the broad ligament [9]. So far, only five cases of cellular leiomyoma in the broad ligament have been reported in the literature [7,10-12]. And the comprehensive assessment and diagnosis in these studies are mainly conducted with the aid of tumor markers, ultrasonography and histopathological examination. Nevertheless, little information of cell leiomyoma in the broad ligament is available for its rare occurrence. More challengingly, it is easily confused with the diagnosis of ovarian malignancies. Therefore, a great challenge is

posed for clinicians and pathologists to increase the accuracy of preoperative diagnosis of cellular leiomyoma in the broad ligament.

Patients with cellular leiomyoma in the broad ligament are diagnosed at an advanced stage, with main clinical manifestations of abdominal mass, abdominal distension, and ascites. In line with the findings of previously reported cases, the patient presented with up-regulated CA-125 Levels in this case. However, CA-125 is not a specific marker for cellular leiomyoma in the broad ligament, and will be increasingly expressed in endometriosis and serous ovarian tumors, which therefore increases the difficulty of preoperative diagnosis [1]. These findings also imply, that all cellular leiomyomas in wide ligaments are currently diagnosed by intraoperative observation and postoperative pathology. Accordingly, it is worth mentioning that exploratory laparotomy is the most effective method for making a diagnosis.

In this case, the ring ligation was performed by an attending physician with more than 30 years of clinical surgical experience. Attention should be paid to protecting vital tissues and organs such as bladder, ureter and vital blood vessels during the operation. The tumor was distinctly different from the ovary (Figure 5). Given its complete and clear capsule, the tumor could be preliminarily distinguished from ovarian tumors. Nonetheless, histopathology was the final gold standard should be followed. By the way, postoperative pathological examination of the myometrial wall and bilateral ovaries revealed acute suppurative inflammation, and recurrent low-grade fever in patients may be associated with endogenous fever factors such as tumor growth. In this case report, successful surgical intervention is critical to the prognosis of the patient.

The mass in the patient was so huge that the bladder was moved upward and the boundaries of the interstitial space were unclear. Thus, choosing a conventional hysterectomy has higher risks, like poor hemostasis and excessive intraoperative bleeding. Collectively, we finally chose the retrograde total hysterectomy through Douglas pouch to inhibit bleeding and effectively dissect the uterovesical space. To be specific, the cervix and vagina were exposed in the posterior fornix; blood flow was

blocked in each branch; and the uterus was finally removed after blunt dissection of the uterine space from below to above the level of the bladder triangle.

In addition, hyalinization is the most common secondary phenomenon. With the development of hyaline fibroids, cysts in various sizes were generated after liquefaction and necrosis of muscle cells, which was concluded by reviewing those cases from 2016 and 2020. In a nutshell, large masses were associated with cystic and myxoid lesions. Cystic leiomyomas displayed decreased T1 signal intensity and increased T2 signal intensity (no increase observed in the cystic lesion area) on MRI, while common leiomyomas exhibited lower T2 signal intensity. This perspective has indicated a way for the clinical differentiation of common leiomyomas and cystic leiomyomas [12].

Cellular leiomyoma, a variant of leiomyoma, is one of the most special types of uterine leiomyoma. On the study conducted by Shabana *et al* of 964 female genital leiomyomas, cellular leiomyomas were discovered to be the most common variant of leiomyomas, but they remained clinically easily confused with malignancy due to their specificity[13]. Therefore, the application of bioinformatics to predict relevant specific tumor biomarkers is particularly important for preoperative diagnosis and prognosis. In light of the disease's rarity, it is more crucial to address the mentioned issue with additional research using animal models in subsequent studies.

CONCLUSION

Giant cellular leiomyoma in the broad ligament is a rare clinical neoplasm. The probability of being confused with ovarian malignancy is increased by the elevated CA-125 Levels, as well as cystic and mucinous lesions. All in all, timely and successful surgical intervention is critical for patients indicated for surgery.

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