

# A diagnostic dilemma and surgical attention of a giant broad ligament leiomyoma with cystic degeneration: case report

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Background: Broad ligament leiomyoma is the most common extrauterine leiomyoma, but with a very low incidence rate. When broad ligament leiomyoma is giant and undergoes degeneration, it poses both clinical evaluation and radiological difficulty in differentiating from an ovarian tumor. Meanwhile the changes of anatomical structure increased the difficulty during surgery. Conclusions: Thus, the diagnosis of a cyst-solid complex adnexal mass with a marginally raised serum level of CA125 might be a broad ligament leiomyoma with degeneration besides a ovary tumor. During surgery of myomectomy, we should be meticulous hemostasis and avoid ureter and bladder injury and other complications.

#### Kevwords

Broad ligament leiomyoma; Degeneration; Diagnosis; Myomectomy

#### 1. Introduction

The broad ligament is a double-layered sheet of mesothelial cells originating from 2 layers peritoneal folds and it connects the sides of the uterus to the lateral walls and floors of pelvis. Between the 2 layers tissues, there are connective tissue, smooth muscles, nerves, and blood vessels [1]. Tumors of the broad ligaments are rare. The most common solid tumor of the broad ligament is a leiomyoma [2]. Broad ligament leiomyoma is usually asymptomatic. In uncomplicated cases (e.g., small and no degeneration) broad ligament leiomyoma is clinically interpreted before surgery as a solid adnexal mass which is separated from both the uterine body as well as the ovary. However, if the leiomyoma reaches a significant size, it can lead to various degeneration when the innervation and nutritional status for the leiomyoma changes. Then it may pose greater diagnostic difficulty than when it occurs in the uterus [3]. This is one such case report where a diagnosis of the malignant ovarian tumor was made and finally, the operation and histopathology confirmed the diagnosis of a giant broad ligament leiomyoma with cystic degeneration.

#### 2. Case report

A 46-year-old, married woman, admitted to Qingdao Central Hospital in February 2016 and presented with the complaint of abdominal distension for a month, along with mild abdominal pain. She had no complaints related to menstrual irregularities. The associated complaints of bladder or bowel, anorexia, or weight loss were also not reported. She had one past full-time normal vaginal delivery 22 years ago. Special medical or surgical history and the history for a family member with genital malignancy were not reported. Upon physical examination, her vital signs were normal and we could found an obviously swollen belly as the size of the full-term pregnancy. Abdominal palpation revealed a giant firm mass that extended to the subdiaphragmatic area. No abdominal rebound pain or tenderness was observed. Per vaginal examination revealed a normal vulva, vagina, and vaginal portion of the cervix but fornices were full. Per pelvic examination, we could find a giant cyst-solid mass with a diameter of about 30 cm, the uterus and adnexa were not palpable and we could not determine the mass originated from the uterus or the adnexa.

The laboratory investigation reports showed that hemoglobin was 11.0 g/dL and the serum level of cancer antigen 125 (CA125) was 38 U/mL, while Carcino Embryonic Antigen (CEA) and serum alpha fetal protein (AFP) and HE 4 levels were normal. Kidney and liver function tests were within normal limits. Abdominopelvic computed tomography (CT) scan on 13-02-2016 revealed a well-defined, heterogeneous, and mixed density mass measuring 35 × 30 cm separating from the uterus and pushing the uterus to the left side of the pelvis. The giant mass filled the whole abdominal and pelvic cavity which extended to the area 2 cm lower than the xiphisternum, and stretched down to the pubic symphysis and the left. The right bound of the mass reached the anterior axillary line. Her liver, pancreas, spleen, and adrenal glands were normal and no ascites or lymphadenopathy was detected (Fig. 1).

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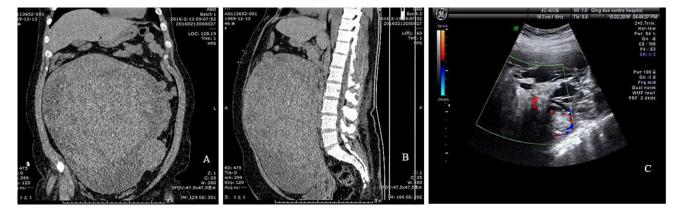


Fig. 1. (A–B) Abdominopelvic computed tomography scan revealed a well-defined, heterogeneous, and mixed density mass of  $35 \times 30$  cm separating from the uterus and pushing the uterus to the left side of the pelvis. (C) Transvaginal ultrasound showed a giant cyst-solid complex mass, and color flow imaging displayed abundant blood flow signals.

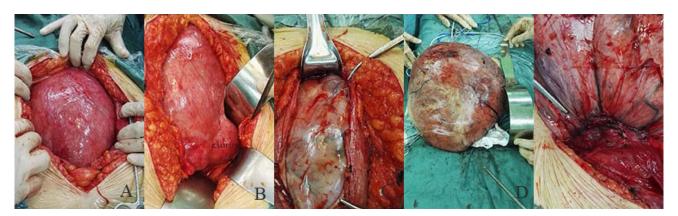


Fig. 2. (A–B) Intraoperative findings revealed a normal uterus, bilateral fallopian tubes, bilateral ovaries, and a giant broad ligament leiomyoma. (C–D) The surface of broad ligament leiomyoma showed solid and cystic areas. The solid foci had a brown fleshy homogeneous appearance and the cyst contained colorless and transparent fluid. (E) A large number of prominently dilated nutrient vessels were identified in the root of the broad ligament leiomyoma.

The transvaginal ultrasound on 15-02-2016 showed a giant cyst-solid complex mass and color flow imaging displayed abundant blood flow signals (Fig. 1). Furthermore, in the transvaginal ultrasound, we could found the normal uterus and left ovary, but the right ovary couldn't be detected. So the provisional diagnosis of an ovarian tumor was made given the elevated CA125 level and the cyst-solid nature of the mass. Then the patient was planned for abdominal unilateral salpingo-oophorectomy.

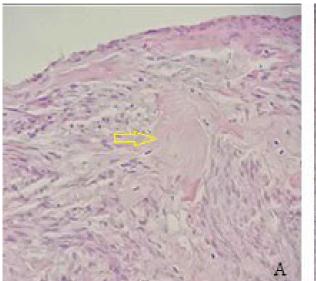
Exploratory laparotomy was done on 17-02-2016 which revealed a huge leiomyoma in the right broad ligament. A giant cyst-solid mass was identified below the abdominal incision intraoperatively (Fig. 2). Firstly, we aspirated the colorless and transparent fluid contained in the multilocular cystic areas of the mass surface to reduce the mass volume and enlarge the operative space. Then the mass which measured about 38 cm  $\times$  29 cm  $\times$  20 cm in size was put upperside, a normal size uterus, normal size ovaries, and fallopian tubes were found. The giant cyst-solid mass lay separately from the right ovary, within the right broad ligament. The right

fallopian tube, right ovary, and right infundibulopelvic ligament were stretched over its upper surface. A large number of prominently dilated nutrient vessels were identified in the root of the broad ligament leiomyoma. The right ureter below the lateral side of the giant mass. So we resected the giant broad ligament leiomyoma and the specimen was sent for frozen section. The solid foci of the mass had a brown fleshy homogeneous appearance. The frozen section presented myoma of the uterus without malignancy. The final histopathological examination suggested leiomyoma of the uterus with extensive cystic changes and partial cells intensely proliferated but without histologic signs of malignancy (Fig. 3). Finally, the patient was discharged 5 days after surgery with no complications.

# 3. Discussion

Leiomyoma (also called uterine fibroids) are most often benign, monoclonal, hormone-dependant tumors composed of uterine smooth muscle interlaced with connective tissue, and it's size range from a few millimeters to tens of centimeters. Approximately 20%–30% of women of reproductive age

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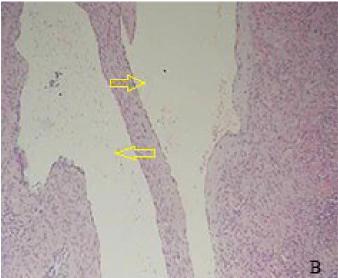


Fig. 3. (A) Hematoxylin and eosin stain at  $\times$ 100 magnification showing mature smooth muscle cells and intermingled hyaline degeneration (arrow). (B) Photomicrograph shows histomorphology of leiomyoma with cystic degeneration (arrow). Cystic spaces surrounded by smooth muscle cells are seen (Hematoxylin and eosin  $\times$ 40).

have leiomyomas [4], commonly presenting with a variety of symptoms, such as menorrhagia, pelvic discomfort, urine frequency, dyspareunia or constipation [5]. Broad ligament leiomyoma which is a type of extra-uterine leiomyoma accounts for 6% to 10% of uterine myomas [6] and it can be divided into two types according to its origin, and one type of leiomyoma which originates from the lateral wall of the uterine corpus or the cervix and invades the broad ligament is called false broad ligament fibroid, while the other type which originates from smooth muscle of round ligament, tubo-ovarian ligament or smooth muscle of uterine artery or ovarian vessels are called true broad ligament fibroid. Its incidence is < 1% [7]. Because a broad ligament leiomyoma grows outside of the uterus, and it doesn't increase the surface area of the endometrium or pressure on venous drainage, and most cases with a broad ligament leiomyoma are asymptomatic. However, the broad ligament leiomyoma usually has the characteristics of remarkably increased vascularity and low growth resistance. When it is neglected for a long time and it reached an enormous size, it may distort the anatomy of the pelvis and push the uterus to the contralateral side, resulting in chronic pelvic pain, compression of adjacent structures like the bladder, ureter and the bowel, such as bladder and bowel dysfunction or hydronephrosis. As the same time, when the leiomyoma outgrows its blood supply, various degenerative changes occur. The type of leiomyoma degeneration includes hyaline or myxoid degeneration, calcification, cystic degeneration, and red degeneration (hemorrhagic infarction). Hyaline degeneration is the most common type of degeneration and it accounts for about 60% of all the degenerations [8], but cystic degeneration is only be found in 4% of all the degeneration [9]. The broad ligament leiomyoma with degenerative changes often leads to clinical and radi-

ological diagnostic challenges. It mimicked ovarian malignancy and affected evaluation of adnexal masses for optimal patient management [10]. Usually, uterine leiomyoma does not present a clinical and radiological diagnostic challenge. During the transvaginal ultrasound examination, the uterine leiomyoma appears solid in echogenicity with hypogenic shadowing. Our case was a false broad ligament leiomyoma with marked cystic degeneration. The pelvic mass is so huge that it pushes the uterus to the opposite side, and occupied the position of the right adnexa. During the abdominopelvic CT and transvaginal ultrasound examination, the pelvic mass appears extensive huge tumor with solid cum cystic areas which were great and with multiple thin septa. In addition, the serum level of CA125 which was often done to discriminate between benign and malignant ovarian tumors [11] was marginally raised. Such variable appearances would mislead the clinicians in making an entirely different diagnosis. In our case, the patient does not have specific menstrual changes that are associated with uterine leiomyomas. Cystic mass or cystsolid mass in female pelvis most often originate in the ovary, then combined with its CT and ultrasound results and the serum level of CA125, the suspicious diagnosis of the ovarian tumor was made. Broad ligament fibroid can also cause a pseudo-Meigs syndrome with elevated CA125 [12].

The giant broad ligament leiomyoma often changed the position of the adjacent organs and vessels during its growth course, and the huge volume of the leiomyoma caused poor exposure during the surgery as well. All those made the surgery more difficult and increase the risk of injury. When working with giant broad ligament leiomyoma, it is necessary to make adequate preparation before surgery, especially to prevent serious bleeding, ureter and bladder injury, and other complications [13]. In the process of surgery, to avoid

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injury to the right ureter we traced its distribution from the position of the right ureter intersecting with common iliac artery. The right ureter was found in the lower right part of the broad ligament leiomyoma. To avoid injury, we first separated the right ureter from the broad ligament leiomyoma. In our case, we also found a large number of nutrient vessels in the root of the broad ligament leiomyoma, which were easy to cause heavy bleeding during the surgery. Bleeding can easily lead to unclear surgical field and in this condition, the action of clamping blood vessels might increase the probability of ureteral injury. Therefore, in the process of dissecting the broad ligament leiomyoma, the pseudo-capsule close to the tumor body was removed gradually. When happened heavy bleeding, the tumor cavity was quickly packed with large gauze to stop bleeding. The large gauze must be pressed tightly and for enough time. It is forbidden to loosen the pressed gauze frequently to observe whether the bleeding has stopped.

## 4. Conclusions

Thus, though uncommon, a giant broad ligament leiomyoma distorting the pelvic anatomy and presenting with atypical ultrasound features should be considered during the evaluation of adnexal masses for optimal patient management. During surgery, we should be meticulous hemostasis and avoid ureter and bladder injury and other complications.

#### **Author contributions**

XQW was responsible for the topic of the manuscript, collecting and sorting clinical information, and submission of the manuscript and contact with the editors of the magazine, and most of the writing of the manuscript and subsequent revisions. XFY was responsible for the first draft, TM was responsible for communication and contact with the patient, and LLL was responsible for sorting and confirmation of pathological pictures.

## Ethics approval and consent to participate

All subjects gave their informed consent for inclusion before they participated in the study. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Ethics Committee of Qingdao central hospital (approval number: [Y]KY202009001).

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## **Conflict of interest**

The authors declare no conflict of interest.

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