

Received: 2020.12.06

Accepted: 2021.05.12

Available online: 2021.05.24

Published: 2021.06.30

Cystic Ovarian Leiomyoma in a Patient with Progressive Abdominal Pain

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Conflict of interest: None declared

Patient: Female, 45-year-old
Final Diagnosis: Leiomyoma with massive cystic hydropic degeneration
Symptoms: Abdominal distention and pain
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Rare disease

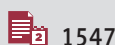
Background: Ovarian leiomyomas are rare, benign, smooth muscle, solid tumors that occur in women aged 20 to 65 years. Because their histology is benign, the prognosis for patients is good. We report the case of a patient with a huge ovarian leiomyoma who presented to the General Surgery Clinic with chronic abdominal pain and progressive abdominal distention.

Case Report: A 45-year-old woman with a history of multiple myomectomies and a total abdominal hysterectomy with right oophorectomy presented to the General Surgery Clinic with abdominal pain and a 4-year history of progressive distention. After being examined, the patient underwent an exploratory laparotomy, during which a pelvic cyst was found that measured 39×30.2 cm, was filled with serous fluid (10.5 L), and occupied most of the abdominal space. The surgery went smoothly and there were no complications during or after the procedure. The patient was discharged home 7 days later in stable condition. Postoperative pathology using hematoxylin and eosin staining and immunohistochemistry with desmin and alpha-smooth muscle actin resulted in a diagnosis of leiomyoma with cystic degeneration. When the patient was seen in the outpatient clinic 2 weeks and 3 and 6 months after surgery, her tumor markers were within normal limits. Abdominal and pelvic computed tomography scans performed at the 6-month visit showed resolution of the loculated intraperitoneal fluid and no gross local recurrence of the tumor.

Conclusions: Ovarian leiomyomas are difficult to diagnose preoperatively. Suspicion for one should be high, however, in patients who present with a large cystic mass in adnexal tissues, especially if they have a history of hysterectomy and oophorectomy.

Keywords: Adnexal Diseases • Leiomyoma • Uterine Myomectomy

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/930299>



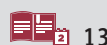
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Background

Leiomyomas of the ovary are rare, benign, solid tumors of smooth muscle. They typically are seen in women of childbearing age and less frequently in women who are postmenopausal [1,2]. Ovarian leiomyomas can present unilaterally or bilaterally; the majority of bilateral cases occur in young patients [2]. Most patients with ovarian leiomyomas have no symptoms because of the small size of the tumors and the incidental nature of their discovery. Some patients, however, present with abdominal pain with or without distention [3]. Most women with ovarian leiomyomas have a history of uterine leiomyoma and hysterectomy [2]. Here, we report our experience with and considerations about diagnosis and treatment of an unusually large cystic ovarian leiomyoma.

Case Report

A 45-year-old woman with well-controlled diabetes mellitus and hypertension was admitted through the General Surgery Clinic for investigation of abdominal pain and a 4-year history of progressive abdominal distension. Her medical history revealed 3 myomectomies, the first done at age 30 years and the other 2 successively for uterine leiomyomas. At age 40, the patient underwent a total abdominal hysterectomy with right oophorectomy.

On physical examination, the patient's vital signs were stable. Her abdominal examination revealed a distended abdomen with mild tenderness in the lower portion. Preoperative tumor marker levels were 1.86 for carcinoembryonic antigen (CEA) and 14.62 for human carbohydrate antigen 125 (Ca-125), both within normal limits. Pelvic ultrasonography done outside our hospital revealed a massive intraperitoneal collection of thick fluid with layering and septations in the right and left upper abdomen and the pelvic cavity, with a 15×7-cm heterogeneous mass in the right lower abdomen, with cystic areas and hypervascularity. Also, it showed a 5.7×5-cm left adnexal cystic lesion. Preoperative computed tomography (CT) of the abdomen and pelvis revealed an irregular heterogeneous-complex lesion on the right side of the pelvic cavity that measured 14×11 cm and was compressing but not invading the bladder. A large amount of loculated fluid was seen in the anterior aspect of the abdominal cavity, which measured 40×24×32 cm and had a thin enhancing wall. A 5.3×3.5-cm cyst also was seen in the left ovary (**Figure 1**).

Because a peritoneal malignancy was suspected, the Surgical Oncology team recommended resection of the mass and possible hyperthermic intraperitoneal chemotherapy (HIPEC), depending on the results from an intraoperative frozen section. Assessment of the patient by the Anesthesia team showed

that she had good functional capacity, her metabolic equivalent of task was >4, and she had no respiratory symptoms or signs of reduced pulmonary function. Her airway assessment was good (normal mouth opening, thyromental distance, temporomandibular joint movement, and a Mallampati score of 1). The plan was for general anesthesia (inhaled), invasive monitoring with an arterial line, and regional analgesia in the form of an epidural.

Under general anesthesia and through a laparotomy incision, we found a large pelvic cyst filled with 10.5 L of serous fluid, which was occupying most of the abdominal space and adherent to the pelvic wall. There were no peritoneal deposits or mucinous masses. The intraoperative frozen section specimen showed fibrous inflammation with giant cells, with no clear evidence of mucin. We then dissected the cyst from the pelvic wall, bladder, and rectal mesentery and performed a left oophorectomy and omentectomy without HIPEC (**Figure 2**). After surgery, the mass was dissected and described as a deflated cyst measuring 39×30.2 cm. The wall was approximately 1.5 cm thick. The outer surface was irregular with a single white deposit and was otherwise smooth, glistening, and intact. The inner surface also was smooth.

Postoperative pathology using hematoxylin and eosin staining and immunohistochemistry with desmin and alpha-smooth muscle actin resulted in a diagnosis of leiomyoma with cystic degeneration (**Figure 3**). The surgery took 6 h and there were no intraoperative or postoperative complications. The patient's vital signs were stable throughout the procedure. During surgery, she received 5 L of crystalloids and 2 units of packed red blood cells. Her urine output was approximately 450 mL. The approximate fluid balance was 1.1 L. After successful extubation, the patient was transferred to the Surgical Intensive Care Unit for postoperative care and management. Two days later, she was moved to the non-intensive care unit. Her epidural catheter was removed 48 h after surgery. The patient was discharged home 7 days after surgery in stable condition. When she was seen in the outpatient clinic 2 weeks and 3 and 6 months later, her tumor markers within normal limits. During the 6-month visit, abdominal and pelvic CT showed resolution of the loculated intraperitoneal fluid and no gross local recurrence.

Discussion

Ovarian leiomyomas have been described as rare benign tumors. They reportedly account for 1% of all ovarian tumors. Most ovarian leiomyomas have benign histology but can range from benign to borderline to malignant [3,4]. In a few cases, estrogen and progesterone have been reported to play a role in the development and progression of ovarian leiomyomas.

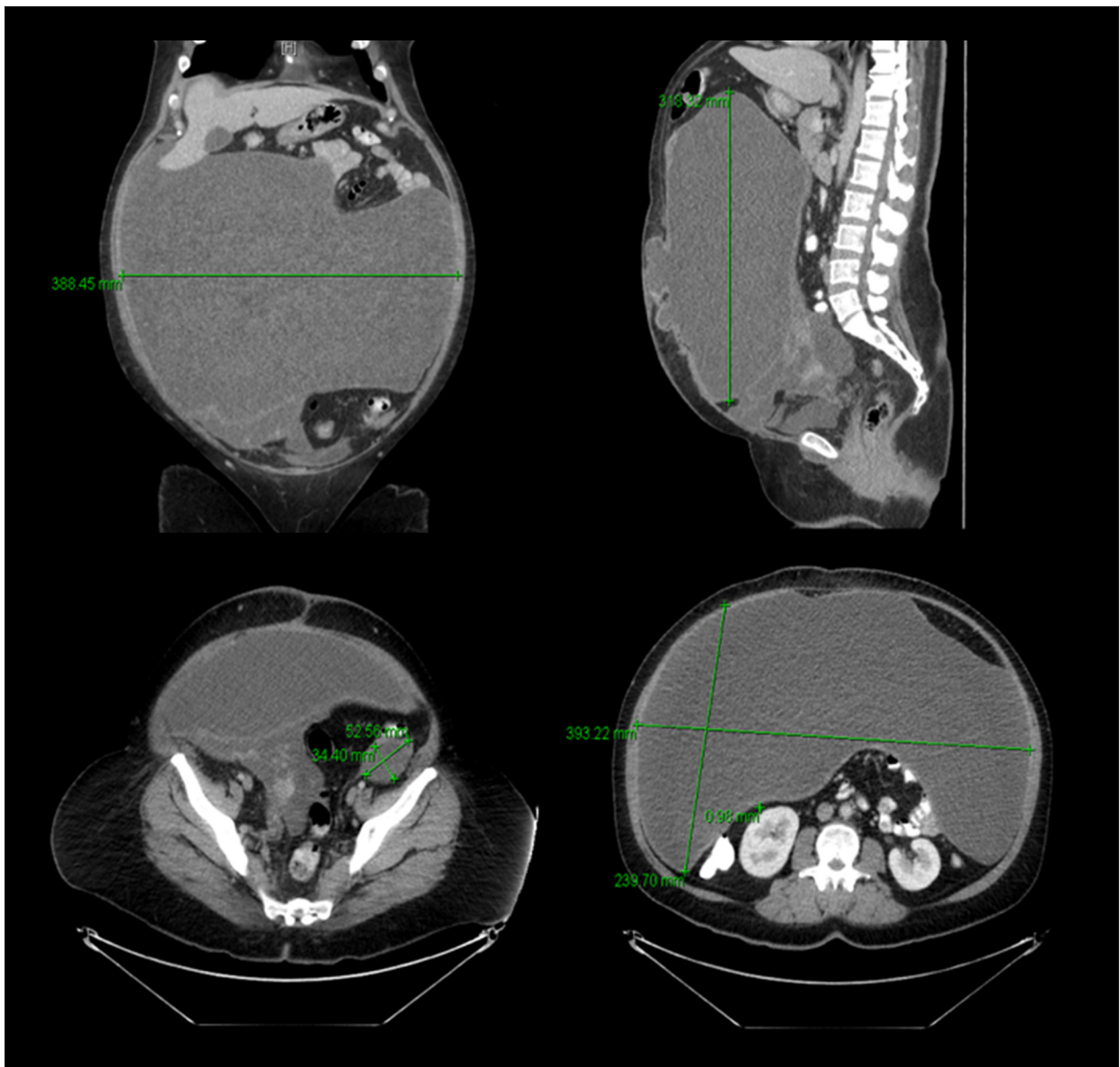


Figure 1. Computed tomography shows an irregular, heterogeneous, complex lesion in the right side of pelvis measuring approximately 14×11 cm, a 5.3×3.5-cm cyst in the left ovary, and a large, loculated fluid collection occupying the anterior aspect of the abdominal cavity that measures 40×24×32 cm in transverse, anterior posterior, and craniocaudal dimensions, respectively, with a thin enhancing wall.

In some patients, the tumors have been noted to increase in size with pregnancy and menstruation [5,6]. Patients with ovarian leiomyomas usually have no symptoms. When the tumors are sizable, however, they can present with non-painful abdominal swelling or, less often, minimal abdominal pain. On rare occasions, the tumors can cause compression, resulting in symptoms of urinary frequency and constipation [1-3]. Moreover, ovarian leiomyomas can be primary or secondary, based on the time of presentation. Primary ovarian leiomyomas are thought to arise from smooth muscle cells in vessel walls of the ovaries. Some scholars theorize that they arise

from smooth muscle cells in the ovarian ligament or stroma. Others believe that the tumors develop from undifferentiated reproductive cells and cortical smooth muscle cells, or from smooth muscle cells in the form of metaplasia or ovarian endometriosis [3]. Ovarian leiomyomas that develop from tissues external to the ovaries and come into contact with the ovaries are referred to as secondary. As in the patient in our case, who had undergone a hysterectomy and right oophorectomy due to multiple myomas, some tumor cells can remain in or fall from the uterus, attach to the ovary, and grow over time [5-7].



Figure 2. The large pelvic cyst filled with serous fluid after resection.

Given the low incidence and indistinct presentation of ovarian leiomyomas, making the diagnosis before surgery can be extremely challenging. The only method for definitive diagnosis is postoperative pathology. Imaging modalities that have been used to diagnose ovarian leiomyomas include ultrasonography, CT, and magnetic resonance imaging (MRI). MRI is preferable because it has excellent sensitivity and reveals detailed anatomical features in multiple planes [6,8].

Performing frozen section histopathology intraoperatively in cases of suspected ovarian leiomyoma is important, as the results provide a better understanding of the type of tumor and help guide surgical decisions. In our patient's case, intraoperative chemotherapy had been planned but was not given because the frozen section results showed fibrous inflammation with giant cells and no evidence of malignancy [3]. The postoperative pathology report resulted in a diagnosis of leiomyoma

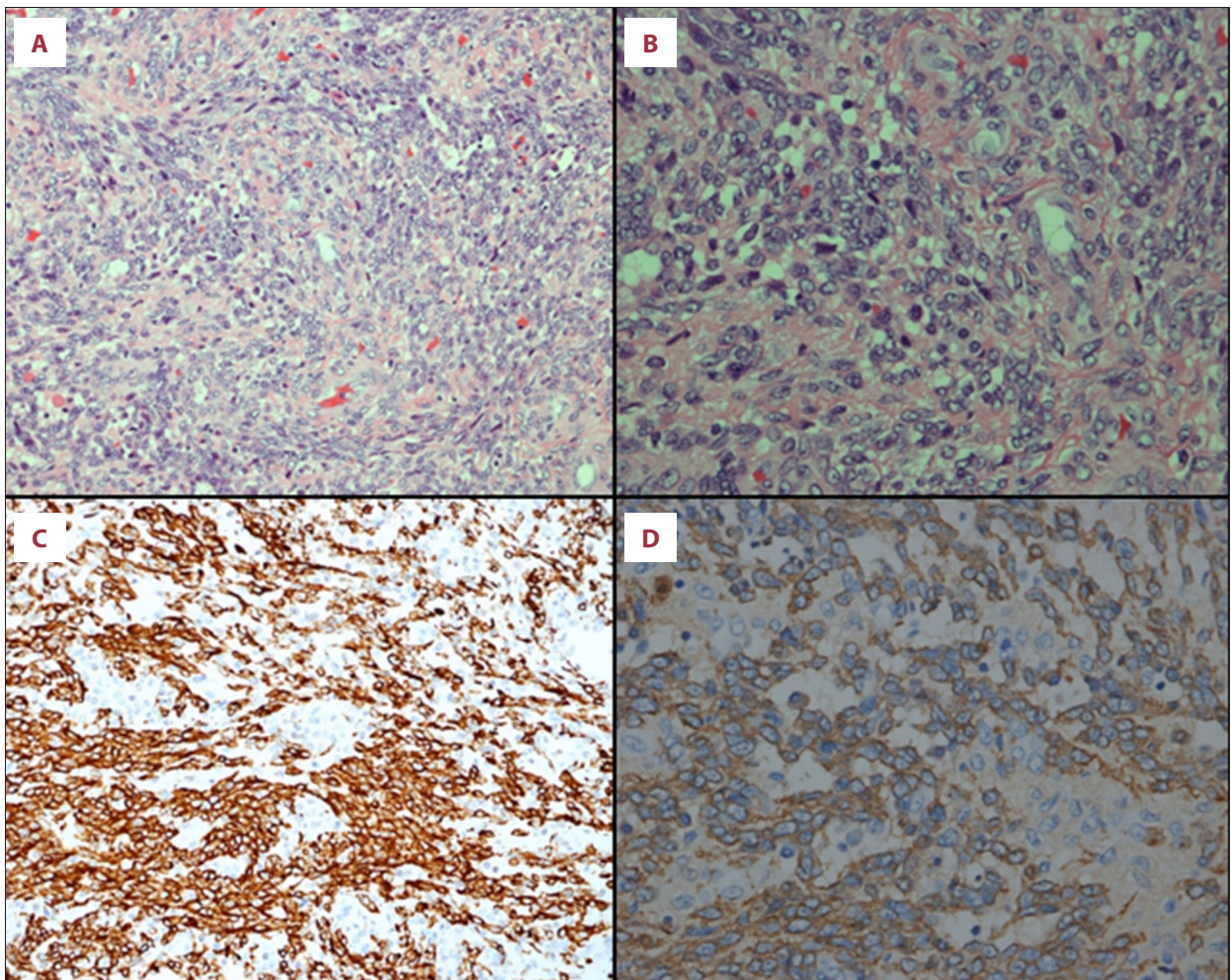


Figure 3. (A, B) Hematoxylin and eosin staining shows spindle-shaped tumor cells with no mitoses (100× and 200×, respectively). (C) Immunohistochemistry of the tumor for desmin. (D) Immunohistochemistry for alpha-smooth muscle actin.

with cystic degeneration. Reports in the literature about ovarian leiomyoma describe tumors with an average size of 5.5 cm, the largest documented case measuring 25 cm in diameter [1,3,6,9]. Different histopathological types of leiomyoma have been reported, including typical, cellular, and mitotically active leiomyomas; leiomyomas with bizarre nuclei; and myxoid leiomyomas with all of those types sharing a solid component. When leiomyomas grow, different types of degeneration can occur as they expand beyond their blood supply, including include cystic, hyaline, myxoid, and dystrophic calcification. Cystic degeneration, which was similar to our case, is rare and is observed in about 4% of leiomyomas; only a few cases have been reported in the literature [10-12].

Surgery is of great value for treatment of ovarian leiomyomas and it is important to consider the age of the patient when selecting the type of procedure. For example, in postmenopausal or middle-aged women who are not fertile or do not wish to preserve their fertility, bilateral salpingo-oophorectomy is the best option [3,13]. In our case, the patient had previously undergone a hysterectomy with right oophorectomy and our intervention included a left oophorectomy with excision of the large leiomyoma. After resection, adjuvant chemotherapy was not warranted, given the benign nature of the disease. Furthermore, recurrence of leiomyomas of ovarian origin is uncommon, which makes the prognosis excellent. However, in young women who need to preserve their fertility, surgery to completely eliminate the tumor may not be ideal. Given

the possibility of estrogen and progesterone involvement in the progression of ovarian leiomyomas, antiestrogenic therapy may have a role in the postoperative treatment plan, but more studies are needed. Although more reports about ovarian leiomyomas are being published in the literature, they are still considered rare and additional research is needed to better understand the nature and mechanism of the disease and to help identify the most successful method for preoperative diagnosis and optimum management for women in different age groups.

Conclusions

Ovarian leiomyomas are rare ovarian tumors that are difficult to diagnose before surgery. Suspicion for ovarian leiomyoma with cystic degeneration should be high in patients who present with large, cystic masses in adnexal tissues, especially individuals who have a history of hysterectomy for a fibroid uterus and oophorectomy. Intraoperative frozen section is beneficial and considered an effective approach to help determine how to proceed surgically. A postoperative pathology report should follow to reach a definitive diagnosis.

Conflict of Interest

None.

References:

1. Doss BJ, Wanek SM, Jacques SM, et al. Ovarian leiomyomas: Clinicopathologic features in fifteen cases. *Int J Gynecol Pathol.* 1999;18(1):63-68
2. Vierhout ME, Pijpers L, Tham MN, Chadha-Ajwani S. Leiomyoma of the ovary. *Acta Obstet Gynecol Scand.* 1990;69(5):445-47
3. Wang QM, Zhao Y, Ma Y et al: One case report of giant atypical leiomyoma of the ovary. *Medicine (Baltimore).* 2018;97(40):e12526
4. Van Winter JT, Stanhope CR. Giant ovarian leiomyoma associated with ascites and polymyositis. *Obstet Gynecol.* 1992;80(3 Pt 2):560-63
5. Hsiao CH, Wang HC, Chang SL. Ovarian leiomyoma in a pregnant woman. *Taiwan J Obstet Gynecol.* 2007;46(3):311-13
6. Blue NR, Felix JC, Jaque J. Primary ovarian leiomyoma in a premenarchal adolescent: first reported case. *J Pediatr Adolesc Gynecol.* 2014;27(4):e87-88
7. Lerwill MF, Sung R, Oliva E, Prat J, Young RH. Smooth muscle tumors of the ovary: A clinicopathologic study of 54 cases emphasizing prognostic criteria, histologic variants, and differential diagnosis. *Am J Surg Pathol.* 2004;28(11):1436-51
8. Kawano Y, Takai N, Shimano M, et al. Magnetic resonance imaging findings in leiomyoma of the ovary: A case report. *Arch Gynecol Obstet.* 2006;273(5):298-300
9. van Esch EM, van Wijngaarden SE, Schaafsma HE, et al. The diagnostic and therapeutic approach of a primary bilateral leiomyoma of the ovaries: A case report and a literature review. *Arch Gynecol Obstet.* 2011;283(6):1369-71
10. Low SC, Chong CL. A case of cystic leiomyoma mimicking an ovarian malignancy. *Ann Acad Med Singapore.* 2004;33(3):371-74
11. Hacivelioglu S, Erkanli S. A large pedunculated leiomyoma with two-sided cystic degenerations mimicking a bilateral ovarian malignancy: A case report. *Eur J Gynaecol Oncol.* 2014;35(2):192-94
12. Kim JC, Nam SL, Suh KS. Leiomyoma of the ovary mimicking mucinous cystadenoma. *Clin Imaging.* 2000;24(1):34-37
13. Wei C, Lilic N, Shorter N, Garrow E. Primary ovarian leiomyoma: A rare cause of ovarian tumor in adolescence. *J Pediatr Adolesc Gynecol.* 2008;21(1):33-36