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Symplastic leiomyoma with cystic degeneration masquerading as an endometriotic cyst on ultrasound

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Abstract:

Symplastic leiomyoma is a rare type of leiomyoma diagnosed histologically by the presence of bizarre cells and nuclear atypia. It is difficult to diagnose preoperatively due to its overlapping clinical presentation and ultrasound features with typical leiomyomas. This paper presents a case of a 52-year-old woman who consulted due to right lower quadrant pain and abdominal enlargement. On transvaginal ultrasound and computed tomography scan, she was diagnosed with right ovarian new growth measuring 9 cm × 8 cm × 10 cm with features suggestive of an endometriotic cyst and a 16.2% risk of malignancy by the International Ovarian Tumor Analysis ADNEX model. Tumor markers CA-125 and the premenopausal ROMA were elevated. The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy. Intraoperatively, the right ovary appeared normal and a 10 cm retroperitoneal cystic mass was seen adherent to the right posterolateral aspect of the uterus and extending towards the right broad ligament, which exuded yellow to brown serous fluid on cut section. Histopathology revealed an intraligamentary symplastic leiomyoma with cystic degeneration. The patient was discharged stable after 3 days.

Keywords:

Cystic degeneration, endometrial cyst, hysterectomy, international ovarian tumor analysis, ovarian new growth, symplastic myoma

Introduction

Leiomyoma is the most common benign tumor of the uterus, affecting mostly reproductive-age women between 30 and 50 years old. Myomas appear on ultrasound as well-defined, round, solid, hypoechoic, heterogeneous masses in the uterus exhibiting posterior acoustic shadowing. However, there are leiomyomas, specifically those with degeneration, which can mimic the ultrasound appearance of a cyst. This paper presents a case of a symplastic leiomyoma with cystic degeneration mimicking an endometrial cyst on ultrasound.

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Case Report

The patient is a 52-year-old Gravida 3 Para 2 (2-0-1-2), with 2 previous spontaneous vaginal deliveries, who consulted for intermittent right lower quadrant pain and an enlarging lower abdomen. The last menstrual period was 3 months before admission. Physical examination showed a normal body mass index of 22.5. The abdomen was soft, nontender, with a palpable cystic mass on the right lower quadrant. The internal exam revealed a slightly movable, nontender, cystic mass in the right adnexa measuring 10 cm × 8 cm.

Three months before the surgery, the patient consulted a different doctor who requested an abdominal computed tomography

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scan with contrast was done which revealed a right adnexal mass measuring 8.5 cm × 8.6 cm × 11.2 cm (volume: 426 ml) described to have heterogeneously enhancing, irregularly thickened walls, displacing the uterus to the left, compressing the right iliac vein and pushing the sigmoid colon to the left. She was advised transvaginal and transabdominal ultrasound which showed a unilocular-solid cyst in the right adnexa, probably the right ovary, measuring 8.93 cm × 7.79 cm × 10.38 cm (vol. 378.10 ml), with ground glass echo pattern and a solid component within measuring 6.80 cm × 5.19 cm × 1.72 cm (volume: 31.79 mL), casting posterior acoustic shadows and exhibiting

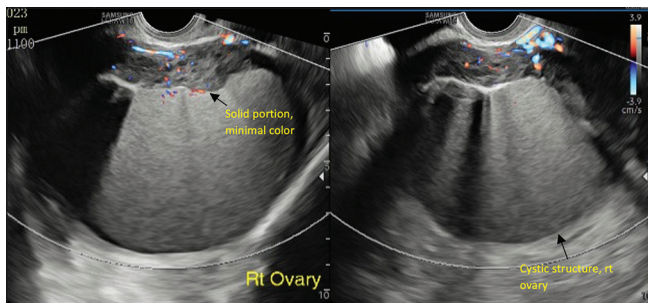


Figure 1: Transvaginal ultrasound showing a unilocular cyst in the right adnexa, thought to be the right ovary, containing low-level internal echoes, described as “ground glass,” characteristic of an endometrial cyst (seen in 95% of cases). Within the cyst is a solid component with posterior acoustic shadowing and minimal color on flow mapping

minimal color on flow mapping [Figure 1]. This mass was noted to be adherent to the right lateral aspect of the uterus. The left ovary was converted into a unilocular cyst measuring 3.13 cm × 2.91 cm × 2.35 cm (vol. 11.18 ml), with low and medium level echoes, with no color on flow mapping, suggestive of an endometriotic cyst. Serum tumor markers showed a slightly elevated CA-125 (35.77), normal HE4 (50.48), and an elevated Premenopausal ROMA (7.99). The (International Ovarian Tumor Analysis [IOTA]) ADNEX model was used to assess if the right ovarian cyst was benign or malignant, and the results showed a 10.7% risk of malignancy.

The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy. Intraoperatively, the uterus was normal-sized and levorotated. Arising from the right posterolateral aspect of the uterus and extending retroperitoneally to the right broad ligament was a thick-walled, cystic mass measuring 9.8 cm × 7.7 cm × 3.6 cm, exuding approximately 300 mL of yellow-brown serous fluid on cut section [Figure 2a and b]. The inner cyst wall had trabeculated areas but no solid components or papillary excrescences [Figure 2b]. The left ovary was cystically enlarged, measuring 3.9 cm × 2.5 cm × 2.4 cm, exuding approximately 10 ml of sebaceous material. The right ovary was found to be grossly normal and located superolateral to the uterine mass.

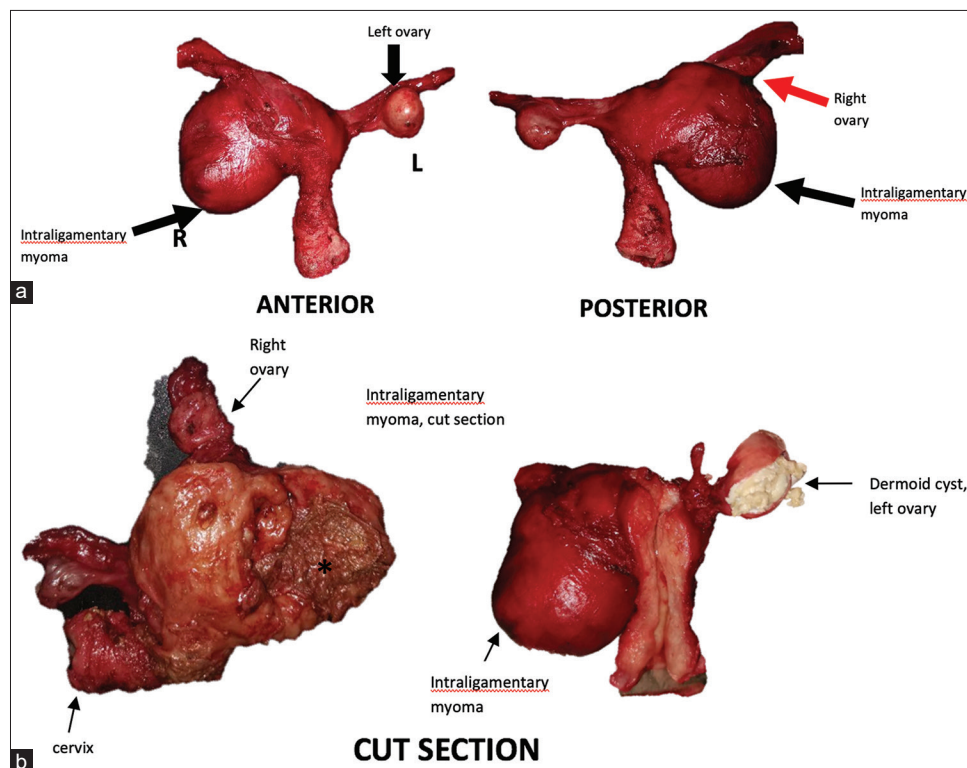


Figure 2: (a) Gross specimen of the patient's uterine corpus, cervix, and bilateral ovaries. The right adnexal mass on ultrasound and computed tomography scan turned out to be an intraligamentary myoma with cystic degeneration. (b) Cut section of the gross specimen. The myoma contained 300 mL of yellow-brown serous fluid (not shown) and the inner cyst wall had trabeculated areas but no solid components or papillary excrescences. Noted the sebaceous material exuding from the left ovary

The patient tolerated the procedure well and was discharged after 3 days. Histopathology revealed a right intraligamentary symplastic leiomyoma with cystic degeneration and dermoid cyst in the left ovary. The right fallopian tube and ovary were normal.

Discussion

According to Guo *et al.*, it was the group of W.M. Christopherson who first described symplastic leiomyoma in 1972.^[1] In 1994, Bell *et al.* introduced the term atypical leiomyomas with low risk of recurrence, pertaining to tumors with moderate to severe atypia, no coagulative tumor necrosis, and mitotic index of ≤ 10 mf/10 hpf.^[2] However, subsequent studies showed that tumors with a mitotic index of 6–9 mf/hpf are associated with recurrence and even metastasis. These cases were eventually reclassified as smooth muscle tumors of uncertain malignant potential (STUMP).^[1,3] Hence in the 2020 World Health Organization classification of uterine smooth muscle tumors, symplastic myomas, also known as leiomyomas with bizarre nuclei, were described to have low mitotic count (<5 mitoses per 10 high power field), no tumor cell necrosis, but with bizarre cells in a background of typical leiomyoma.^[1] Currently available studies involve the older definition of atypical leiomyomas; hence, the exact incidence of symplastic leiomyomas is unknown. For example, Travaglini *et al.* did a systematic review and meta-analysis on 12 studies from 1994 to 2019 involving 433 patients diagnosed with atypical leiomyoma.^[3] Six of those studies (188 patients) included patients with STUMP, meaning the number of symplastic myoma cases may be even lower.^[4]

The lesion in our case report is a combination of the rare types of leiomyomas. First, intraligamentary myomas comprise $<1\%$ of cases.^[4,5] Second, cystic degeneration of myomas is only seen in $2\%–4\%$ of cases,^[6,7] which usually happens when large myomas outgrow their blood supply, leading to liquefaction in the central areas, with subsequent tissue degeneration.^[8] Finally, the incidence of symplastic leiomyomas among all cases of myomas is yet to be studied.

There have been cases of intraligamentary myomas and myomas with cystic degeneration being diagnosed as benign or malignant ovarian cysts on ultrasound due to their appearance and location with respect to the uterus.^[9] Myomas with cystic degeneration appear as large, hypoechoic, or heterogeneous masses with cystic spaces, depending on the ratio of connective tissue to smooth muscle and extent of degeneration.^[10] In our patient, the lesion appeared as a unilocular cyst in the right adnexa with homogenous low-level internal echoes similar to the ground glass echopattern of an endometriotic cyst.^[11] The cyst appeared separate but

adherent to the uterus. No normal right ovarian tissue was visualized on ultrasound as well. An ovarian neoplasm was also considered due to the presence of a solid component with posterior acoustic shadowing and minimal color on flow mapping, as well as the slightly elevated Ca-125. Using all these findings, the risk of malignancy was computed through IOTA Adnex model, which was significant (16.2%).

Based on a meta-analysis by Travaglini *et al.*, the recurrence risk of symplastic leiomyomas is 1.9%.^[3] A case report by Cooney *et al.* discussed a case of symplastic leiomyoma recurring as leiomyosarcoma in the vagina after total abdominal hysterectomy and bilateral salpingo-oophorectomy.^[12] Aside from that, the risk of malignant progression of symplastic myomas is unknown.^[3] High-risk patients with large tumor size, infiltrating borders, and atypical mitosis should be closely monitored and advised yearly imaging to check for recurrence or, rarely, new-onset leiomyosarcoma.^[12] Currently, there is no evidence recommending yearly magnetic resonance imaging (MRI) for patients who underwent surgery.

Conclusion

The combination of a symplastic broad ligament leiomyoma with cystic degeneration is a very rare occurrence, with no case reports published yet in literature. A symplastic leiomyoma is an uncommon variant of uterine leiomyomas with a low recurrence rate and rarely may progress to malignant degeneration. Differentiating symplastic from typical leiomyoma is difficult due to the overlapping gross morphologic and ultrasound characteristics, and a histological examination is needed to obtain a definitive diagnosis.^[4] Caution must be exercised when evaluating adnexal masses on ultrasound as myomas with cystic degeneration may look similar to benign and malignant ovarian cysts. Other imaging tools, such as MRI, may be used as an adjunct if ultrasound is inconclusive.^[13]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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