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CASE REPORT

Sclerosing stromal tumor of the ovary: Two case reports and literature review



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KEYWORDS

Sclerosing stromal tumor;
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Abstract

Introduction: Sclerosing stromal tumors (SSTs) are rare benign ovarian tumors. They represent 6% of sex cord stromal tumors. Its preoperative diagnosis is often a challenge due to its similarity to malignant tumors on ultrasound imaging. We present two cases of SSTs to emphasize the consideration of this type of tumors in the differential diagnosis of solid adnexal masses in young women. A review of the literature on the typical ultrasound features, clinical presentation, and management of SSTs was performed.

Main symptoms and/or clinical findings: Pelvic pain was the main symptom in both cases. In the first case, transvaginal ultrasound revealed an unilocular solid adnexal mass of 59 mm × 44 mm × 45 mm with cystic areas and marked peripheral and central vascularization. MRI (magnetic resonance imaging) revealed a 50 mm × 50 mm heterogeneous adnexal mass with a solid peripheral component and a cystic-necrotic center. In the second case, pelvic ultrasound showed a solid cystic adnexal mass of 103 mm × 77 mm with marked peripheral vascularity.

Main diagnoses: Postoperative anatomopathological diagnosis in both cases was an ovarian SST. **Therapeutic interventions and results:** Unilateral laparoscopic salpingo-oophorectomy and oophorectomy, respectively, was performed without incidents. There has been no recurrence during follow-up.

Conclusion: It is important to consider SSTs in the differential diagnosis of young women with a unilateral solid-cystic adnexal mass with a high degree of peripheral and central vascularization. Laparoscopic approach together with fertility-sparing techniques should be considered the treatment of choice.

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PALABRAS CLAVE

Tumor estromal
esclerosante;
Neoplasia de ovario;
Descripción de un
caso

Tumor estromal esclerosante de ovario: reporte de 2 casos clínicos y revisión de la literatura**Resumen**

Introducción: Los tumores esclerosantes del estroma (SST) son tumores benignos raros del ovario. Representan un 6% de los tumores del estroma de los cordones sexuales. Su diagnóstico preoperatorio suele ser un desafío por su similitud ecográfica con los tumores malignos. Presentamos 2 casos de SST para enfatizar la consideración de este tipo de tumores en el diagnóstico diferencial de masas anexiales sólidas en mujeres jóvenes. Se realizó una revisión de la literatura sobre las características ecográficas típicas, la presentación clínica y el manejo de los SST.

Principales síntomas y/o hallazgos clínicos: El dolor pélvico fue el síntoma principal en ambos casos. En el primer caso, la ecografía transvaginal reveló una masa anexial unilocular sólida de $59 \times 44 \times 45$ mm con áreas quísticas y marcada vascularización periférica y central. La resonancia magnética nuclear reveló una masa anexial heterogénea de 50×50 mm con componente sólido periférico y un centro quístico-necrótico. En el segundo caso, la ecografía pélvica mostró una masa anexial sólido quística de 103×77 mm con marcada vascularización periférica.

Diagnósticos principales: El diagnóstico anatomopatológico postoperatorio en ambos casos fue de un SST de ovario.

Intervenciones terapéuticas y resultados: Se realizó ooforectomía y salpingooforectomía unilateral laparoscópica, respectivamente, sin incidencias. No se ha producido recidiva durante el seguimiento.

Conclusión: Es importante considerar los SST en el diagnóstico diferencial ante mujeres jóvenes con una masa anexial sólido-quística unilateral con un alto grado de vascularización periférica y central. El abordaje laparoscópico junto con técnicas preservadoras de fertilidad deben ser consideradas el tratamiento de elección.

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Introduction

Sclerosing stromal tumors (SST) of the ovary are a rare benign neoplasm that account for approximately 8% of all primary ovarian neoplasms.¹ They are a subtype of ovarian stromal neoplasms of the sex chord-stromal category. This entity was first reported in 1973 by Chalvardjan and Scully.² Unfortunately, lack of familiarity with this entity makes a preoperative diagnosis challenging.

They usually appear in young women and common clinical presentations of SST include menstrual irregularities, pelvic pain, or nonspecific symptoms associated with pelvic mass.³ Imaging findings for SSTs can mimic those of borderline or malignant ovarian tumors. Transvaginal ultrasound is usually the first step. Gray-scale ultrasound⁴ and color Doppler findings have been described in previous publications.^{5,6}

To contribute to the proper clinical, ultrasound and pathological evaluation of SST and highlight the importance of considering SST in the differential diagnosis in young women, we describe two cases of SST.

Patient information

In our first case, a 21-year-old healthy nulliparous woman without medical or surgical history of concern, was presented in the outpatient department of gynecology with pelvic pain during the last 3 months.

In the second case, a 19-year-old caucasian woman without medical or surgical history of interest presented to the emergency department with sudden onset of abdominal pain. A urine pregnancy test was negative. The patient had been diagnosed of an ovarian mass a few months ago and was being followed up in an outpatient clinic and waiting for surgery.

Clinical findings

In the first case, a vaginal examination revealed a large, palpable mass in the pouch of Douglas. There were no unusual symptoms such as hypermenorrhea, menstrual irregularities or virilization. In the second case, on abdominal palpation, a firm mass in the hypogastric region was noted.

Timeline and diagnostic assessment

In the first patient, ultrasound examination showed a normal uterus and right ovary (Fig. 1a). On the left site there was an unilocular solid mass of $59 \text{ mm} \times 44 \text{ mm} \times 45 \text{ mm}$ with cystic areas and peripheral and central vascularization with doppler score 3. Minim free fluid in the pouch of Douglas was found. Ultrasound evaluation of the mass was GIRADS-4 with suspicion of a non-epithelial tumor. Laboratory tests, including tumor markers (Ca-125, Ca-19.9, alpha-FP), were normal. MRI was performed and revealed a heterogeneous left adnexal mass of $5 \text{ cm} \times 5 \text{ cm}$ with peripheral solid

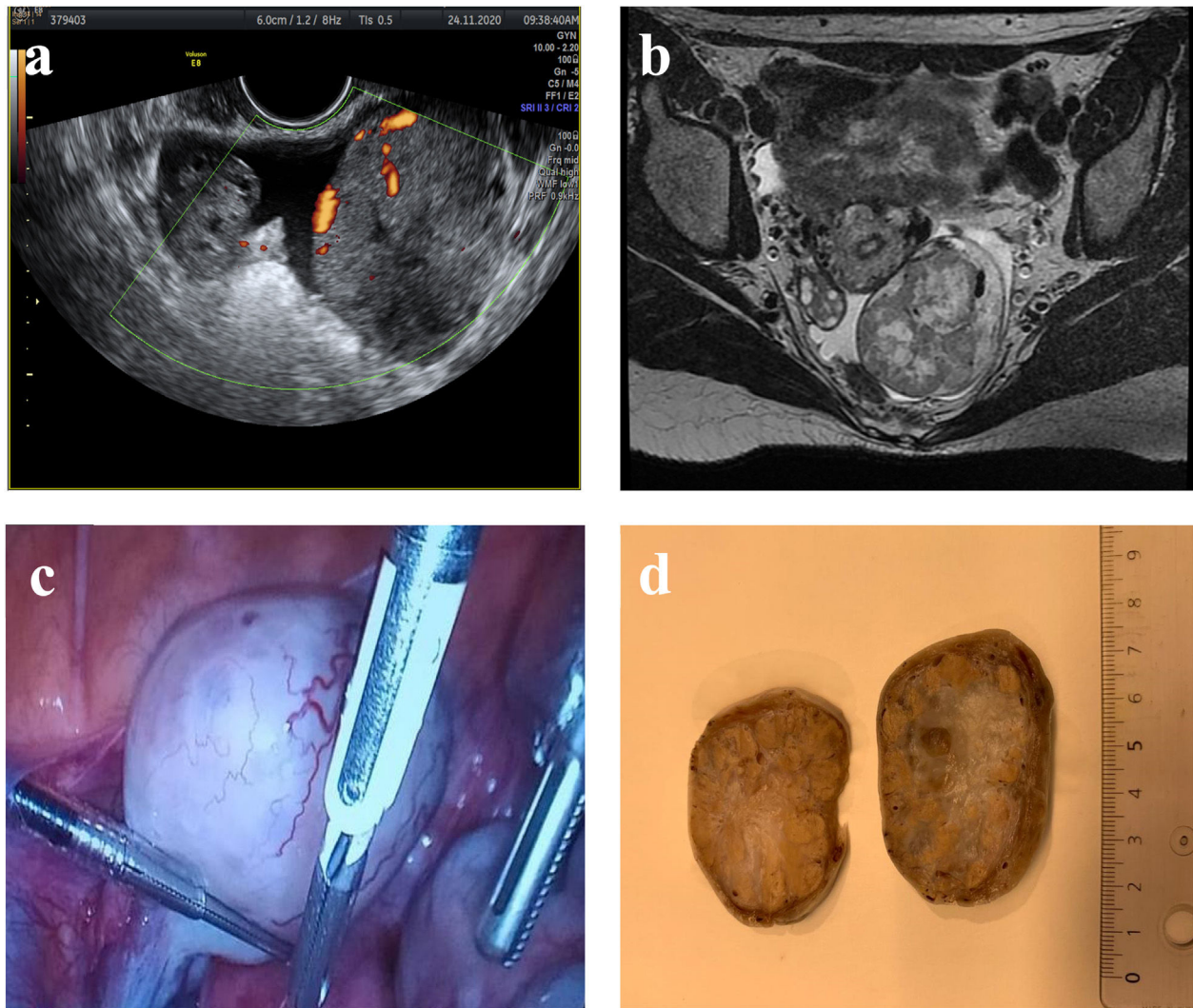


Figure 1 (a) Transvaginal ultrasound revealed a solid hypoechoic tumor with cystic areas and regular borders with multiple vessels in the periphery and central part. The right ovary was normal. A small amount of free fluid was present. (b) MRI revealed an ovoid solid tumor with regular contours. (c) On laparoscopy a solid lobulated mass with a high vascularized capsule depending of the left ovary was found. (d) Macroscopic examination revealed a capsulated tumor with firm consistency and yellowish nodular peripheral components and an oedematous central part.

component and a cystic-necrotic center showing a low signal on T1 and heterogeneous on T2, with enhancement of solid component. The right ovary, uterus and cervix were without alterations (Fig. 1b).

In the second case, pelvic ultrasonography showed a right solid-cystic adnexal mass of 103 mm × 77 mm with marked peripheral vascularization. The left ovary and uterus were normal.

Therapeutic intervention

The first patient underwent laparoscopic surgery with left oophorectomy as there was no normal ovarian tissue present. Surgery revealed a normal uterus and right ovary with the left ovary replaced by a solid mass with marked superficial vascularization (Fig. 1c). On gross inspection, the removed left ovarian mass measured

64 mm × 55 mm × 40 mm. The mass was gray-white in color and had a smooth and well-encapsulated surface with prominent vascularization. The cut surface was a mostly solid with a medium-elastic consistency and slightly edematous in the center with a peripheral multinodular orange border (Fig. 1d).

The second patient also underwent laparoscopy, which revealed the presence of a large solid adnexal mass which was torsioned. Unilateral salpingo-oophorectomy was conducted.

The histological diagnosis in both cases was a SST of the ovary.

Follow-up and outcomes

The first patient's postoperative course was uneventful. She was discharged on day 2 after surgery. No recurrence has

Table 1 Overview of case reports on SST between 2014 and 2022.

Case report	Age (years)	Clinical presentation	Tumor markers	Tumor size (cm)	Ultrasound findings	Surgical treatment
Ahuja et al., 2022. (1 case)	13	Abdominal pain	No data	9.7-cm	Solid, heterogenous left ovarian mass	Laparoscopic cystectomy
Ashrafganjoei et al., 2016 (1 case)	23	Abdominal pain	α -Fetoprotein: <0.5 U/ml CA-125: 17.2 ml ⁻¹ β -HCG: 0 miu/ml	5 × 5cm	Solid ovarian mass with heterogeneous echotexture.	Laparotomic excision
Atram et al., 2014. (3 cases)	Case 1: 15 Case 2: 24 Case 3: 25	Case 1: pelvic pain and menstrual irregularity Case 2: vaginal bleeding Case 3: abdominal distension	No data	Case 1: 7 × 6 × 4 cm Case 2: 5.3 × 4 × 3.9 cm Case 3: 9 × 8 × 4 cm	Case 1: ovarian mass of 7 × 6 × 4 cm of variegated consistency with an irregular surface. Case 2: well-defined solid-cystic echogenic mass Case 3: no data	Case 1: excision Case 2: salpingo-oophorectomy Case 3: laparoscopic excision
Bairwa et al., 2017 (1 case)	24	Lower abdominal pain.	Tumor markers were within normal limits.	10 × 10 × 8 cm	Solid cystic mass	Salpingo-oophorectomy
Bennett et al., 2015 (8 cases)	20–37	4 incidental findings at cesarean section and 4 were found at prenatal visits at 15, 16, 20, and 32wk of gestation.	No data	Tumors ranged from 3.1 to 21 cm (mean = 8.8 cm)	No data	Salpingo-oophorectomy (7), cystectomy (1).
Chaurasia et al., 2014 (1 case)	17	Precocious puberty	β -HCG: 2.5 miu/ml CEA: 1.5 ng/ml α -fetoprotein: 3.5 ng/ml	16 × 12.5 × 10 cm	Well-defined, heterogeneous solid and cystic mass	Salpingo-oophorectomy
Chen et al., 2022. (1 case)	17	Virilization. Meig's syndrome	No data	27.1 cm × 20.0 cm × 11.0 cm	No data	Laparotomic right salpingo-oophorectomy
Furukawa et al., 2015. (1 case)	18	Menstrual irregularities	CA125, CEA, CA19-9, AFP normal	6 cm	Echogenic mass separated from the uterus. Color Doppler showed peripheral hypervascularity.	Total, laparoscopic cystectomy

Table 1 (Continued)

Case report	Age (years)	Clinical presentation	Tumor markers	Tumor size (cm)	Ultrasound findings	Surgical treatment
Grechi et al., 2015 (1 case)	24	Secondary amenorrhea	CA125: 21 U/ml	10 × 9 × 10.5 cm	Multiloculated cystic lesion in the region of the left adnexa with multiple solid vascular- ized areas (color score 3); no residual ovarian tissue.	Salpingo-oophorectomy
Hirakawa et al., 2016. (2 cases)	Case 1: 25 Case 2: 24	Case 1: irregular and frequent menstruation Case 2: irregular menstruation	Case 1: CA-125, CA19-9, β-HCG normal limits. Case 2: CA-125: 79.5 U/ml.	Case 1: 10 cm Case 2: 8 cm	Case 1: pelvic mass Case 2: pelvic mass with a solid part	Case 1: salpingo-oophorectomy Case 2: salpingo-oophorectomy
Karabulut et al., 2014. (1 case)	20	Intermittent abdominal pain and menstrual irregularity.	CA19-9: 66.3 IU/ml Other tumor markers were within normal limits.	5 × 6 cm	Semisolid mass	Laparotomic salpingo-oophorectomy.
Kim et al., 2014. (1 case)	80	Postmeno-pausal bleeding	AFP: 11.6 ng/ml CEA: 2.28 ng/ml Ca19-9: 6.8 U/ml Ca-125: 114.8 U/ml β-HCG: 1.8 mIU/ml	9 × 10.5 × 10 cm	Pelvic mass	Laparotomic total hysterectomy and bilateral salpingo-oophorectomy
Lee et al., 2015 (2 cases)	Case 1: 63 Case 2: 59	Case 1: lower abdominal discomfort Case 2: abdominal pain with dysuria	Case 1: tumor markers negative. Case 2: CA-12,537.9 U/ml	Case 1: 17.5 × 14.5 × 9.0 cm Case 2: 13.2 × 10.0 × 5.5 cm	Case 1: well defined heterogeneous, predominantly cystic pelvic mass with solid portions. Case 2: solid mass with a cystic component	Case 1: total abdominal hysterectomy with salpingo-oophorectomy, left external iliac lymph node sampling and washing cytology. Case 2: total abdominal hysterectomy, salpingo-oophorectomy and omental biopsy.
Liu et al., 2015 (2 cases)	Case 1: 29 Case 2: 28	Case 1: abnormal vaginal bleeding associated with lower abdominal pain. Ectopic pregnancy. Case 2: ectopic pregnancy.	No data	Case 1: no data Case 2: 5.0 × 5.0-cm	Case 1: well-defined solid mass Case 2: heterogeneous solid mass	Case 1: Laparoscopic enucleation Case 2: laparoscopic tumor enucleation

Table 1 (Continued)

Case report	Age (years)	Clinical presentation	Tumor markers	Tumor size (cm)	Ultrasound findings	Surgical treatment
Liu et al., 2017. (1 case)	46	No data	Elevated CA-125.	No data	Cystic-solid left pelvic mass and ascitis	No data
Marinho et al., 2021. (1 case)	18	Abdominal pain	Tumor markers were within normal limits.	7.7 × 6.9 × 9.8 cm	Cystic and solid mass with vascularization.	Laparoscopic salpingo-oophorectomy
Matsutani et al., 2018 (1 case)	17	Abdominal pain	AFP, CEA and CA19-9 was all within normal limits CA-125: 76.3 U/ml	15 cm	Multiseptated cystic mass	Oophorectomy and omentectomy.
Mensah et al., 2017. (1 case)	32	Menstrual irregularity and secondary infertility. Primary mesenteric extra gonadal sclerosing stromal tumor.	No data	8.2 × 9.1 × 9.4 cm	No data	Laparotomic excision.
Mokhtari et al., 2014. (1 case)	45	Hypermenorrhea and abdominal pain	LDH, CEA 1.9, AFP CA-19-9, CA-125 normal	5 × 3.5 × 3 cm	Solid echogenic mass	Laparotomic excision
Naidu et al., 2015 (1 case)	14	Virilization	β-HCG: <1 miu/ml α-Fetoprotein: 2.9 ng/ml, CEA: 1.3 ng/ml	12 cm and 5 cm	Large bilateral solid pelvic masses. Normal ovaries were not clearly identified	Laparotomic left salpingo-oophorectomy and right cystectomy.
Özdemir et al., 2016. (1 case)	Postmenopausal	Virilization	No data	10 × 9 × 5 cm	Encapsulated ovarian mass without any capsular breach	Salpingo-oophorectomy
Palmeiro et al., 2016 (1 case)	20	Pelvic pain.	AFP, CA 125, β-HCG within normal limits	7.8 cm	Solid hypoechoic tumor with regular borders, Color Doppler: multiple vessels inside the lesion, more numerous at the periphery and extending to its central portion	Salpingo-oophorectomy

Table 1 (Continued)

Case report	Age (years)	Clinical presentation	Tumor markers	Tumor size (cm)	Ultrasound findings	Surgical treatment
Squillarro et al., 2018 (1 case)	10 months	Precocious puberty	Gonadotropin, estradiol, and androgen levels were normal as well as ovarian tumor markers	2.7 × 2.5 × 1.7 cm	Solid, slightly heterogeneous lesion with associated color flow anterior to a normal-appearing uterus without clear visualization of either ovary.	Laparotomic salpingo-oophorectomy
Tomimatsu et al., 2016. (1 case)	22	Large abdominal solid mass	No data	No data	No data	Salpingo-oophorectomy
Uner et al., 2017. (1 case)	29 pregnant	Intracesarean incidental finding	No data	30 cm	No data	Salpingo-oophorectomy.
Whang et al., 2022. (1 case)	28	Pelvic mass	Normal limits	4.8 × 3.7 cm	Cystic and solid mass in the left ovary	Laparoscopy
Yen et al., 2014 (1 case)	9	Virilization	B-HCG: 1 mIU/ml α-Fetoprotein: 1.3 ng/ml CA 125: 17.5 U/ml CEA 2 ng/ml	15 × 8.5 × 6 cm	No data	Laparotomic salpingo-oophorectomy
Yesil et al., 2016. (1 case)	17	Abdominal pain.	No data	5 × 4cm	No data	Laparoscopic excision
Zhai et al., 2015. (1 case)	No data	Severe preeclampsia	No data	No data	No data	No data
Zhang et al., 2020. (1 case)	26	Pelvic pain	Ca 125 normal	7 × 7 × 2 cm	Hypoechoic and irregular mass in the left adnexal region. Well circumscribed with sparse-blood flow signals.	Laparotomic salpingo-oophorectomy
Zhang et al., 2020 (1 case)	11	Abdominal pain and menstrual irregularities	α-Fetoprotein: 14.03 μg/L CEA 0.81 μg/L CA 19-9: 10.50 U/ml Ca 125: 10.69 U/ml HE4: 31.24pm Tissue polypeptide antigen: 0.18 ng/ml	7.51 × 5.39 × 7.6 cm	Predominantly solid in echotexture with a multiloculated cystic lesion	Laparoscopic cystectomy
Zhao et al., 2019. (1 case)	70	Postmenopausal bleeding	Serum estradiol: 49.78 ng/L	3 × 2 cm		Laparoscopic hysterectomy and bilateral salpingo-oophorectomy

occurred during 11 months of follow-up. In the second case, post-operative recovery was uneventful, and no recurrence has occurred during follow-up.

Discussion

SST are a rare benign ovarian neoplasm that account for approximately 8% of all primary ovarian neoplasms. They are attributable to theca cell-fibrous tumor subtypes of ovarian sex cord-stromal tumor and account for approximately 6% of them.³

In the literature, reports of ovarian SSTs are rare. Until 2003, 114 cases have been reported by Peng et al.³ Ozdemir et al.⁷ reported a total of 208 cases until 2014. We undertook a MEDLINE® search using keywords “ovarian neoplasms” and “sclerosing stromal tumor” to obtain case reports on this tumor in the English literature. A total of 44 cases from 32 reports have been identified from 2014 up to the writing of this paper in 2022. The clinical reports and their characteristics are listed in Table 1.

Clinically, SST usually present in the 2nd and 3rd decades of life, in contrast to other ovarian stromal tumors which present in the 5th or 6th decade. Most of the cases are unilateral.^{7,8} There are reports for extragonadal localizations.^{9,10}

As in our cases, the most common symptoms are pelvic pain and menstrual irregularities. SST presenting with ovarian torsion were described in two reports.^{11,12} There are publications of SST in pregnant women.^{13,14} Bennett et al.¹³ reported 8 cases of pregnant women with SST and most of the cases were incidental findings during prenatal ultrasound follow up or at cesarean sections.

Similar to our first case, laboratory findings are nonspecific and tumor markers are often within normal limits, but elevated levels had been reported.^{15,16}

SST are typically nonfunctioning, but there are publications reporting elevated levels of both estrogenic and androgenic hormones. Hormone production can cause irregular menses, amenorrhea, infertility, precocious puberty and virilization.^{17–19} The association with Meigs and McCune-Albright syndrome has been documented.^{20–22} Endometrial hyperplasia and postmenopausal bleeding concomitant with SST have been described.^{23,24}

The etiology of SSTs is unknown. They were thought to arise from pluripotent immature stromal cells of the ovarian cortex.²⁵ Some authors propose that they arise from the perifollicular myoid stromal cells.²⁶ Other theories suggest that SSTs and thecomas are probably closely related entities as they share some morphological features and antigenic determinants.²⁷ They are also thought to be developed from preexisting ovarian fibromas.²⁸

Sclerosing stromal tumors have an edematous section surface with cystic changes. Microscopic examination shows a pseudolobular pattern, admixed spindled and rounded weakly luteinized cells, and prominent typically ectatic sometimes branching staghorn-like blood vessels.²⁹ Histological features include a pseudolobular pattern with focal areas of sclerosis and 2 distinct cell populations of spindled and polygonal cells separated from

each other by hypocellular, edematous, and collagenous stroma with thin-walled blood vessels.³⁰ Immunohistochemical examination shows positivity for inhibin and vimentin, and negativity for S-100 protein and epithelial markers.^{11,31}

As supported by our cases, a preoperative diagnosis of SST is challenging based on clinical and ultrasonographic findings. The differential diagnosis between SST and malignant tumors is difficult due to their ultrasonographic appearance. Differential diagnoses include other sex cord stromal tumors, such as fibroma, thecoma, lipoid cell tumors and metastases.⁸

Pelvic ultrasound, computed tomography and MRI imaging are useful for preoperative diagnosis of SST. Ultrasound is cost-effective and usually the first diagnostic approach to a pelvis mass.⁵ Gray scale ultrasound appearance of SST, described by Joja et al., is mainly a solid or multilocular unilateral cyst with cystic areas and acoustic shadowing.⁴ The importance of color Doppler with a high degree of peripheral and a slight central vascularization was described by Deval et al.⁵ On T2-weighted MRI images, signal intensities of the cystic components are high and those of the solid components are inhomogeneous. Dynamic MRI has a marked early enhancement of the solid components.^{4,32} The contrast enhancement pattern of SST of the ovary on dynamic CT and MRI is characteristic and involves peripheral contrast enhancement in the early phase after contrast material administration followed by centripetal progression in the delayed phases.³³ Ultrasound elastography may be used as a complementary imaging technique to show the stromal component of SST with quantitative strain values.³⁴

The treatment is surgical excision and a laparoscopic fertility sparing approach should be performed whenever it is possible.³⁵ As in our cases, unilateral salpingo-oophorectomy, either via laparoscopy or laparotomy, is the most common surgical technique.

The strengths of our cases include the clinical and ultrasonographic description of a rare case of benign ovarian tumor with few annual publications. Comparison of our findings with review of the existing literature is provided. Limitation of our case reports is due to the retrospective design. Another limitation is the incomplete information in the second case description.

Conclusion

Although SST are rare benign ovarian tumors, our study highlights the importance of consideration SST in the differential diagnosis of a unilateral, solid cystic, ovarian mass with a high degree of peripheral and a slight central vascularization in young female women. A preoperative diagnosis is usually difficult, but the familiarity with typical imaging presentation of SSTs allows an accurate preoperative diagnostic orientation and enables a less invasive surgery. Surgical resection of the tumor is curative, with no reported recurrences. Future studies with more cases will offer better insight into this rare entity.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors declare that no patient data appear in this article and that patients gave informed consent.

Patient consent

Patient gave informed consent.

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Conflict of interests

There is no conflict of interest.

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