Case Report

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Primary ovarian leiomyosarcoma in a 72-year-old nulligravid and the 2022 provisional diagnostic criteria for primary ovarian leiomyosarcoma

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Abstract:

Primary ovarian leiomyosarcoma (POLMS) is an exceedingly rare neoplasm accounting for only 0.1% of all ovarian malignancies and most commonly occurring in postmenopausal women. Prognosis is poor with only a 20% 5-year survival rate. Surgery remains to be its mainstay treatment. Discussed here is a 72-year-old nulligravid with hypogastric pain. Transrectal ultrasound showed a right ovarian new growth, probably malignant on International Ovarian Tumor Analysis (IOTA) simple rules, with a 79.2% risk of malignancy by IOTA ADNEX and an unremarkable uterus. Other workups were normal. She underwent primary cytoreductive surgery. Histopathologic diagnosis was ovarian leiomyosarcoma with positive immunohistochemical staining for desmin, S-100 protein, smooth muscle actin, and epithelial membrane antigen. She refused adjuvant chemotherapy postoperatively. The latest published literature on POLMS was also reviewed to develop the provisional criteria for its prompt diagnosis, thereby decreasing the heterogeneity of the diagnostic approach as well as supporting future researches on manifestations, clinical courses, and therapeutic plans.

Keywords:

Ovarian leiomyosarcoma, ovary, primary, prognosis, surgery

Introduction

Primary ovarian leiomyosarcoma (POLMS) is highly uncommon and has been reported in only 0.1% of ovarian malignancies.[1] It is frequently diagnosed in peri- and postmenopausal women with a mean age at diagnosis of 45–60 years of age.[1,2] Patients typically present with abdominal symptoms including pain and altered bladder and bowel habits. The diagnosis is made by direct histopathologic analysis along with further immunohistochemical staining. Surgery remains to be the mainstay treatment. However, recurrences and metastases have been reported in some patients who did not undergo postoperative adjuvant chemotherapy.[2,3] Moreover, the rarity of this disease entity has limited the data available to establish the optimal course of postoperative monitoring and treatment. Prognosis is poor with only a 20% 5-year survival rate.[1] This report aims to propose diagnostic criteria for POLMS that encompass the clinical, surgical, histopathologic, and immunohistochemical aspects of the disease, derived from the most recent published papers on POLMS.

Case Report

This patient is a 72 year-old nulligravid who presented with hypogastric pain. She has no known comorbidities and has no family history of ovarian cancer.

Six weeks prior, she noted the onset of dull hypogastric pain, with a pain score of 8 out of 10, aggravated by movement and urination. There was no associated

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vaginal discharge, fever, or urinary symptoms. The patient sought outpatient consultation and transrectal ultrasound showed a normal-sized uterus with a slightly thickened endometrium of 0.52 cm. There was a was a unilocular heterogeneous mass with solid components and irregular borders measuring 12.24 cm × 11.83 cm × 9.89 cm in the right adnexa and anterior to the uterus. The mass had a color score of 3, Sonologic impression was to consider an ovarian new growth probably malignant based on the International Ovarian Tumor Analysis (IOTA) simple rules with a 79.2% risk of malignancy by the IOTA ADNEX [Figure 1]. No free fluid in the cul-de-sac. Complete blood count showed mild anemia, leukocytosis with neutrophilic predominance, and thrombocytosis. Urinalysis was normal. Impaired fasting glucose was noted at 106 mg/dL but hemoglobin A1c was within normal limits at 5.4%. Serum albumin level was low at 2.8 g/dL. Her chest X-ray and 12-L *electrocardiogram* findings were unremarkable. CA-125 was requested but the patient did not comply due to financial constraints. She was then scheduled for surgery. Upon admission, the patient was conscious, coherent, and not in respiratory distress, with stable vital signs. On abdominal examination, a 6 cm × 4 cm mass was palpated on the hypogastric region with smooth borders and was fixed and nontender. A digital rectal examination showed no palpable masses, no tenderness, and no fullness in the cul-de-sac.

The patient had cardiovascular pulmonary clearance for surgery. Thereafter, she underwent exploratory laparotomy. Upon opening the peritoneum, the large bowel was firmly adherent to the peritoneum. A solid, irregularly nodular contoured mass of approximately 9.5 cm × 9.0 cm × 5.0 cm was noted attached to the right adnexa with firm adhesions to the omentum and bowels. Peritoneal fluid sampling, enterolysis, excision of right ovarian tumor, frozen section, abdominal hysterectomy, and bilateral salpingo-oophorectomy were done. During enterolysis, a surgical spill of approximately 100 ml of pus-like, foul-smelling fluid occurred. On the cut section, the mass was partially cystic with tan, soft to slightly firm, and dull solid areas [Figure 2]. The uterus with cervix measured 7.0 cm × 5.0 cm × 3.0 cm and the serosa was extensively covered with adhesions. The uterus appeared grossly normal on sectioning. Both the right and left adnexa were extensively engulfed in adhesions [Figure 2]. Segments of the ileum and rectosigmoid sustained lacerations during further enterolysis, hence segmental ileal resection with end-to-end anastomosis, primary repair of the ileal and rectosigmoid perforation, lavage, and Jackson Pratt (JP) drain insertion was done. The frozen section of the ovarian tumor showed a malignant mesenchymal (mesodermal) sarcoma favoring neural origin.

Postoperatively, the patient underwent a blood transfusion. She was discharged after 4 days.

On follow-up, the final histopathologic report of the right ovarian tumor showed a neoplastic process without any recognizable normal tissue, with multifocal areas of necrosis, characterized by a proliferation of clumped spindle cells demonstrating a prominent whorl arrangement, enlarged nuclei with abnormal mitoses [Figure 3], and consistent with leiomyosarcoma. Further immunohistochemical staining of the ovarian tumor showed strong positivity for desmin, weak to strong positivity for S-100 protein, moderate-to-strong positivity for smooth muscle actin (SMA), and moderate-to-strong positivity for epithelial membrane antigen (EMA) [Figure 4]. The right ovary showed multiple serous cysts and endometriosis. Both fallopian tubes, the left ovary, the uterus, segments of the ileum along with the attached fatty mesentery, and peritoneal fluid were all unremarkable.

The patient was diagnosed with leiomyosarcoma of the right ovary, Stage IIB. The plan was to give adjuvant

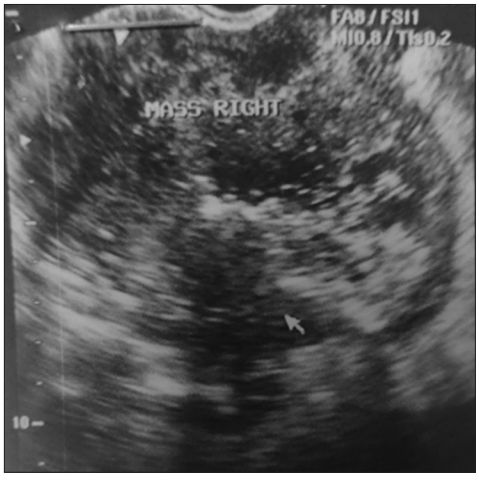

Figure 1: Ultrasonographic image showing a unilocular solid heterogenous mass in the right adnexa

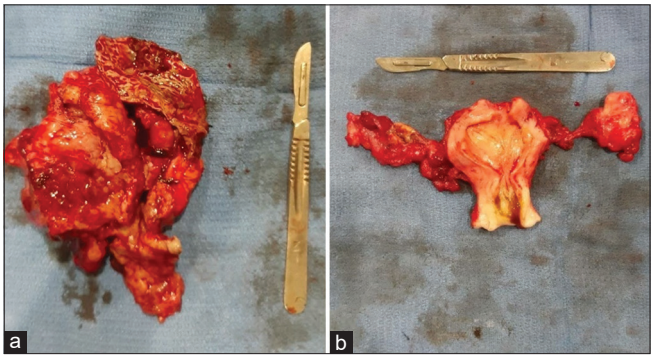

Figure 2: Gross specimen: (a) right ovarian tumor. (b) The grossly normal-looking uterus and both the right and left adnexa are extensively engulfed in adhesions obliterating the tubo-ovarian anatomical architecture

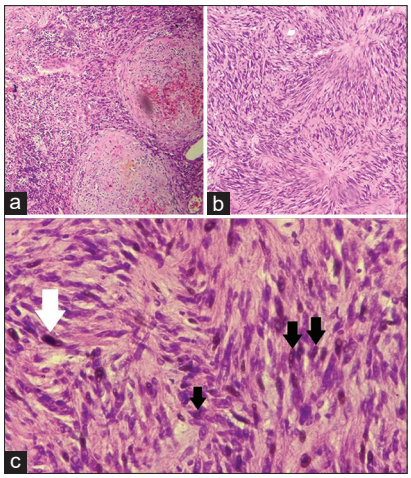

Figure 3: Histopathologic assessment revealed an ovarian tumor, which contained a neoplastic process with multifocal areas of necrosis. (a) The proliferation of clumped spindle cells demonstrates a prominent whorl arrangement (H and E, ×40). (b) Spindle cells with enlarged nuclei (white arrow) with abnormal mitoses (black arrows) (H and E, ×100) (c)

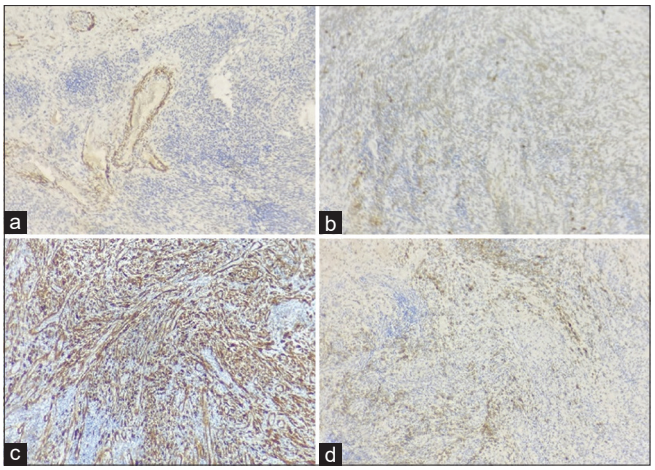

Figure 4: Immunohistochemical staining of the ovarian tumor showing strong positivity for desmin. (a) Weak to strong positivity for S-100 protein. (b) Moderate-to-strong positivity for smooth muscle actin. (c) Moderate-to-strong positivity for epithelial membrane antigen (d)

chemotherapy with gemcitabine and docetaxel every 3 weeks for 6 cycles, but the patient was unable to comply due to financial constraints. At present, she is alive and well and still refuses chemotherapy and surveillance imaging.

Discussion

POLMS is thought to arise from malignant degeneration of an ovarian leiomyoma or of the vascular smooth muscles in the cortical stroma and corpus luteum, muscular attachments of the ovarian ligament, in Wolffian duct remnants, in totipotential cells of ovarian mesenchyme, or arising in a teratoma.[2,4] The mean age at diagnosis is between 45 and 60 years of age.[1,2] However, cases in younger women have also been reported.[2,5] The index case had the oldest age at diagnosis by far, and just like the other reported cases, she also presented with nonspecific symptoms. The others commonly complained of abdominal pain and distension, accompanied in some cases by alteration in urinary and bowel habits. On physical examination, a soft-to-firm mass is usually palpated on the abdomen. Preoperative imaging shows an intrapelvic or intra-abdominal multiloculated, complex mass with solid and cystic areas which may or may not have septa.[2,6] In 90% of cases, the ovarian tumor is unilateral,[1] just like the patient in this report. Preoperative tumor markers such as CA 125, CA 19-9, alpha-fetoprotein, carcinoembryonic antigen, and CA 15-3 are usually within normal limits or only slightly elevated.

Surgery remains to be the recommended treatment for most cases of POLMS and may range from fertility-sparing to systematic surgical staging.[7] As with other ovarian cancers, staging is based on the International Federation of Gynecology and Obstetrics guidelines. For postmenopausal women and for younger women who are no longer desirous for pregnancy, exploratory laparotomy with an attempt to completely resect the tumor must be done. When malignancy is confirmed by frozen section, debulking surgery may then proceed to consist of total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, and excision of all resectable tumors within the pelvis and abdomen.[4] The patient in this paper had advanced age and was postmenopausal hence a primary cytoreductive procedure was performed. On gross examination, the ovarian tumor usually has solid and cystic components, >10 cm in diameter, with areas of hemorrhage and necrosis. The diagnosis of leiomyosarcoma is done through microscopy with evidence of hypercellularity, nuclear atypia, pleomorphism, coagulative necrosis, and high mitotic activity (>5 mitotic figures/10 high-power fields).[4,7,8] Further immunohistochemical staining is generally positive for SMA, desmin, vimentin, and caldesmon, weakly positive to negative for S-100 protein, and negative for inhibin, EMA, and cytokeratin antibodies.[2-5,8,9] Abeler and Nenodovic studied the expression of vimentin, AE1/AE3, SMA, desmin, h-caldesmon, actin, Myf4, CD10, CD31, CD68, CD117, factor VIII, HMB-45, and S-100 protein in 397 uterine sarcomas. They found that SMA was positive in 90% of leiomyosarcomas with increased positivity of 96% and 92% when combined with desmin or h-caldesmon, respectively.[10] Endothelial markers, S-100 protein, and CD117 do not seem to be of any diagnostic value. Both microscopic examination and immunohistochemical staining can differentiate leiomyosarcoma from other soft-tissue tumors but cannot distinguish whether the tumor is of ovarian or uterine origin. Therefore, there is a need to correlate the clinical, histopathological, and immunohistochemical aspects of the disease to arrive at a diagnosis of POLMS.

In this report, the patient's gross and microscopic examinations of the tumor along with the strong positivity for both SMA and desmin support the diagnosis of leiomyosarcoma. In addition to this, her clinical presentation of hypogastric pain, a palpable unilateral abdominal mass, ultrasonographic evidence of ovarian new growth, intraoperative findings of a partially cystic mass with the solid areas densely adherent to the right ovary, and the absence of direct connection to and pathology in the uterus and other adjacent organs all validate that the leiomyosarcoma is primarily ovarian.

A review of the literature for the past 10 years has shown that POLMS is difficult to detect preoperatively and there are no existing definite criteria that will lead to its diagnosis. In the summary of cases [Table 1], the common presentation of patients later diagnosed with POLMS is nonspecific abdominal pain with a palpable abdominal mass. Imaging studies, whether by ultrasound, computed tomography, or magnetic resonance imaging, reveal a unilateral pelvic mass with solid and cystic components. Intraoperative findings also reveal the mass to be directly attached to either ovary. No significant pathology was noted in the uterus, where leiomyosarcoma occurs more commonly, as well as in the other adjacent organs. The diagnosis is further supplemented by microscopic examination and immunohistochemical staining of the tumor consistent with leiomyosarcoma. Based on the review of the nine latest reports on POLMS, this paper proposes these provisional criteria for the diagnosis of POLMS [Table 2] to facilitate recognition of the condition by the clinicians, standardize its diagnostic approach, and provide a more homogeneous study population for future treatment researches.

Due to the paucity of data, there is currently no established postoperative course of care for patients diagnosed with POLMS. In cases, where the tumor was completely resected but with no postoperative adjuvant treatment, patients may live recurrence-free for up to 6–31 months.[4,8] However, close surveillance with a computed tomography scan for a span of 10 years may be done as the majority of patients will have a local or distant relapse within the first 2 years.[1] POLMS may metastasize by local invasion, hematogenous, and lymphatic spread. The common sites of metastases are the lungs and liver but it has also been reported to metastasize to the pelvic bones and as a subcutaneous tumor on the shoulder.[3]

Postoperative adjuvant radiotherapy may play a role in local disease control for those with tumors that have not been completely resected.[4] Different courses of adjuvant chemotherapy have been applied [Table 1]. A patient who received four cycles of doxorubicin + ifosfamide + mesna (mycobacterium avium-intracellulare regimen) remained asymptomatic and without signs of disease relapse after 24 months of follow-up.[9] Another case reported receiving vincristine, epirubicin, and cyclophosphamide for six cycles remained disease-free on clinical and radiological examination 6 months after treatment. Furutake *et al*. found that a course of gemcitabine + docetaxel yielded high response rates in uterine leiomyosarcoma and used this in a case of POLMS with liver metastasis and the patient was alive and well 24 months after starting chemotherapy.[3] The largest case series on POLMS published was done by Cojocaru *et al*. in which they found that there is no proven benefit for adjuvant chemotherapy for localized disease. However, in the metastatic setting, long-term tumor control can be achieved by chemotherapy, surgery, radiotherapy, or radiofrequency ablation.[1]

Studies have suggested that the prognosis of POLMS is dismal, especially with residual disease, and depends on the tumor stage, tumor size, and mitotic index on the diagnosis.[2,4] Patients with Stage I disease on diagnosis survived after a mean follow-up period of 41.6 months without any evidence of disease, whereas those diagnosed at a higher stage died after only 14.7 months.[7] In this case, it is too early to tell what the patient's ultimate outcome will be. However, given that she was diagnosed early with no metastatic sites documented, her prognosis may be better compared to the other cases.

Summary

POLMS is an exceedingly rare malignancy but should still be considered a differential diagnosis in ovarian new growths, especially in postmenopausal women. Prognosis is generally poor but a disease-free survival rate may be improved with early diagnosis and complete tumor resection. Hence, the 2022 provisional diagnostic criteria for POLMS [Table 2] encompassing the clinical, sonographic, gross, microscopic, and immunohistochemical aspects of the disease were proposed in this paper to standardize the diagnostic process and aid clinicians in the early recognition of POLMS to institute treatment promptly. The role of adjuvant chemotherapy and radiation therapy is still not well-established. The course of treatment must be individualized based on existing risk and prognostic factors as well as the availability of the patient's resources.

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Conflicts of interest
There are no conflicts of interest.

Table 1: Summary of case reports

Author/ year	Age	Clinical presentation	Imaging	Surgery	Gross	Microscopy	IHC	Stage at diagnosis	Chemotherapy/ radiation therapy	Outcome
Arslan *et al.* 2010[8]	52	Chronic inguinal pain, soft mass from the uterine fundus up to the abdomen	Well-demarcated lesion at the uterine fundus with occasional hyperechoic areas (suspicious of degenerated leiomyoma)	EL, TAH, BSO, IO, AP, pelvic and para-aortic LND	Yellow-pink, solid with cystic areas and hemorrhagic and necrotic foci; located between the uterine fundus and pouch of Douglas	Hypercellular and necrotic areas; pleomorphic and fusiform cells; 5-7 mitotic figures per 10 HPF	Positive for vimentin and SMA	IA	None	Disease-free after 6 months
Zygouris *et al.* 2012[4]	58	Abdominal pain, palpable pelvic mass	Right adnexal mass	EL, BSO, TO, AP, bilateral LND	Tumor limited to the right ovary without penetrating the serosal surface	Tumor cells with 11 mitotic figures per 10 HPF	Positive for SMA, vimentin, desmin, and ki-67; weakly positive for S-100; negative for EMA, cytokeratin, inhibin, CD 10, and CD 99	IA	None	Disease-free after 21 months
Pankaj *et al.* 2013[2]	27	Right lower abdominal pain, distension, soft-to-firm abdominal mass in the right iliac region with restricted mobility and tenderness	Solid-cystic mass in the right adnexal region	EL, TAH, BSO, TO, pelvic and para-aortic LND	-	Cells with marked atypia, pleomorphic nucleus, prominent nucleoli, >10 mitotic figures per 10 HPF	Positive for desmin and SMA, negative for CD 10 and CD 34	IIB	Gemcitabine + docetaxel	Disease-free after 30 months
Rivas *et al.* 2014[9]	65	Abdominal pain (umbilical and suprapubic region), rectal tenesmus, urinary retention	Rounded pelvic mass compressing the left inferior wall of the bladder, forming a body with the lateral wall of the uterus; normal uterus, and right ovary	TAH, BSO, pelvic radical lymphadenectomy, pelvic lavage	Tumor lesion in the left ovary which compromised the capsule	Fusiform cells set in a random pattern, frequent pleomorphic nuclei, hyperchromatic, prominent nuclei, atypical mitoses >5 in 10 HPF	Positive for SMA, caldesmon, vimentin, desmin, and progesterone receptors	IC	Doxorubicin + Ifosfamide + mesna	Disease-free after 24 months
Kumar, *et al.* 2015[5]	30	Right lower abdominal pain, difficulty in micturition, intermenstrual bleeding, mass palpated in the posterior vaginal fornix	Large mass occupying the whole pelvis just superior to the uterine fundus arising from the right adnexa	EL, TAH, BSO, peritoneal lavage	Ovarian mass with areas of hemorrhage and necrosis	Consistent with ovarian leiomyosarcoma	Positive for vimentin, SMA, and desmin; negative for inhibin	IA	Vincristine + epirubicin + cyclophos-phamide	Disease-free after 6 months

Contd...

Table 1: Contd...

Author/ year	Age	Clinical presentation	Imaging	Surgery	Gross	Microscopy	IHC	Stage at diagnosis	Chemotherapy/ radiation therapy	Outcome
Bacalbasa *et al.* 2016[11]	52	Abdominal pain, soft abdominal mass	Large pelvic mass probably of ovarian origin	EL, TAH, BSO, omentectomy, pelvic and para-aortic LND	Large, ruptured ovarian tumor	Consistent with ovarian leiomyosarcoma	Positive for SMA, negative for inhibin, MNF 116, EMA, and S-100	IC2	Platinum-based chemotherapy	Liver metastasis after 5 years
	57	-	Lesion in the right hepatic lobe with perilesional hematoma	Right hepatectomy	Ruptured liver metastasis in segment VI	Consistent with metastatic ovarian leiomyosarcoma	Positive for SMA, vimentin, caldesmon, desmin, estrogen receptor	IVB	Platinum-based chemotherapy	Disease-free after 2 years
Furutake, *et al.* 2017[3]	40	Abdominal pain	Intrapelvic mass suspicious of a degenerated uterine leiomyoma	EL, left oophorectomy	Misdiagnosed as leiomyoma of the left ovary			IA	None	Metastasis as subcutaneous tumor of the shoulder and liver metastasis after 9 months
	41	Severe back pain, mass on the left shoulder	Left ovarian tumor recurrence, pelvic bone metastasis, multiple liver masses	Biopsy of left shoulder mass	-	Spindle cells arranged in fascicles with partial nuclear atypia and 7 mitotic figures per HPF	Positive for SMA and desmin; negative for S-100, CD 34, and c-kit	IVB	Gemcitabine + docetaxel, RFA	Disease-free after 24 months
Erturk *et al.* 2020[6]	68	Abdominal pain, mobile, hard mass, palpable to the level of the umbilicus	Multilobulated cystic mass with many septa and thick walls posterior to the uterus; origin could not be detected	EL, lavage, adhesiolysis, TAH, BSO	Solid cystic mass arising from the left adnexa adhered to the transverse colon and left pelvic side wall; no evidence of right-sided or upper abdominal disease	Coagulative tumor necrosis, >10 mitoses per 10 HPF, diffuse and severe atypia	None	IA	Adriamycin	-
Present case 2021	72	Hypogastric pain aggravated by movement and micturition, palpable mass in the hypogastric region with smooth borders, fixed and nontender	Mass in the right adnexa, anterior to the uterus, unilocular, heterogeneous with solid components, irregular borders	EL, PFS, enterolysis, excision of right ovarian tumor, TAH, BSO	Solid, irregularly nodular contoured mass attached to the right adnexa with firm adhesions to the omentum and bowels, partially cystic with tan, soft to slightly firm, dull solid areas	Multifocal areas of necrosis, characterized by a proliferation of clumped spindle cells demonstrating a prominent whorl arrangement, enlarged nuclei with abnormal mitoses	strong positivity for desmin, weak to strong positivity for S-100 protein, moderate to strong positivity for SMA, and moderate to strong positivity for EMA	IIB	None	Disease-free after 6 months

EL: Exploratory laparotomy, TAH: Total abdominal hysterectomy, BSO: Bilateral salpingo-oophorectomy, TO: Total omentectomy, LND: Lymph node dissection, IO: Infracolic omentectomy, RFA: Radiofrequency ablation, AP: Appendectomy; PFS: Peritoneal fluid sampling, HPF: High-power field, SMA: Smooth muscle actin, EMA: Epithelial membrane antigen, IHC: Immunohistochemical

Table 2: The 20 22 provisional criteria for the diagnosis of primary ovarian leiomyosarcoma

Clinical presentation
Abdominal or pelvic pain
Palpable abdominal mass
Imaging
Unilateral complex adnexal mass
Malignant features on ultrasound
Gross examination
Complex ovarian tumor with solid and cystic components
With direct attachment to either ovary
Absence of direct attachment to the uterus and other adjacent organs
Absence of tumor lesions on the uterus and other adjacent organs
Microscopic examination
Hypercellularity
Nuclear atypia and pleomorphism
Coagulative necrosis
High mitotic activity (>5 mitotic figures per 10 HPF)
Absence of neoplastic changes in the uterus and other adjacent organs
IHC staining
Positive for smooth muscle actin, desmin, vimentin, and h-caldesmon
Negative for inhibin, CD 10, CD 34, and CD 99

IHC: Immunohistochemical, HPF: High-power field

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