CASE REPORT

Endometrial Stromal Nodule: A Rarity and Diagnostic Challenge

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Abstract

Background: Endometrial stromal tumors (ESTs) are rare tumors arising from the uterus. The incidence of EST is 2 per million women. Endometrial stromal nodules (ESNs) are rare subtype of ESTs and they are benign tumors. We describe a case of ESN since it is a rare tumor and the clinical presentation was different.

Case description: A 54-year-old perimenopausal woman, para 2, live 2 presented with complaints of lower abdomen pain for 2 months. Clinical features, ultrasound, and contrast-enhanced computed tomography (CECT) abdomen report were suggestive of malignant ovarian tumor. Tumor markers were within normal limits. Staging laparotomy was done. Solid mass of 9 × 6 × 6 cm arising from uterine fundus above which large cystic lesion measuring 21 × 18 × 13 cm was seen. Both side fallopian tubes and ovaries were normal. The morphological, microscopic, and immunohistochemical features of that solid with cystic mass were consistent with ESN. Preoperatively suspected malignant ovarian tumor case later turned out to be ESN.

Conclusion: There is no definitive test available to confirm ESN before surgery. Usually, it is diagnosed postoperatively and the clinical presentation could be variable like in our case.

Clinical significance: High-grade endometrial stromal sarcoma (HGESS) can be differentiated from low-grade endometrial stromal sarcoma (LGESS) by gross appearance, microscopic features, mitotic state, and immunohistochemistry.

Keywords: Endometrial stromal nodule, Endometrial stromal tumors, Low-grade endometrial stromal sarcoma.

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Introduction

Among the uterine neoplasms, 3% are endometrial stromal tumors.[1] In 2014, the World Health Organization (WHO) classified EST based on their histological appearance as follows: (1) endometrial stromal nodule, (2) low-grade endometrial stromal sarcoma (LGESS), (3) high-grade endometrial stromal sarcoma, and (4) undifferentiated uterine sarcoma (USS). Endometrial stromal nodule is a rare subtype of EST. Endometrial stromal nodule and LGESS are commonly seen in premenopausal age-group. High-grade endometrial stromal sarcoma and USS usually present in postmenopausal age-group. Endometrial stromal nodule commonly presents as abdominal pain, abnormal uterine bleeding, and pressure symptoms. Here, we present a case of Endometrial stromal nodule not only for its rarity, but it was also a diagnostic challenge since the clinical presentation was different.

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Case Report

A 54-year-old woman was presented with complaints of lower abdomen pain for 2 months. There were no menstrual complaints. She attained menarche at 11 years of age. Menstrual cycles were regular once in 30 days. Flow was moderate, lasting for 2 days. There was no dysmenorrhea. There was no loss of appetite and no loss of weight. No history of bowel or bladder disturbances. She was para 2 live 2, both were normal deliveries; sterilization was done 23 years back. No significant medical or surgical illness occurred. No history of malignancy in the family was found. On examination, she was obese with BMI 37. There was a large abdomino pelvic mass of 28-week gravid uterus size, which was firm in consistency, non-tender, and mobile sideways. The same mass was felt through right, anterior, and left fornices on bimanual examination. Ultrasound examination revealed cystic mass lesion in right adnexa extending into right lumbo-iliac region suggestive of ovarian tumor, hence proceeded with CECT abdomen, which showed a large well-defined complex cystic pelvic-abdominal lesion with multiple blood fluid levels, thick and thin enhancing septations and enhancing solid components in the inferior aspect. Left ovary was not visualized separately. Left ovarian vessels appeared prominent and enlarged. Contrast-enhanced computed tomograph abdomen report was suggestive of malignant ovarian tumor arising from left ovary. Hence, tumor markers were done which were within normal limits. In view of suspected malignant ovarian tumor, we proceeded with staging laparotomy. Ascites around 30 mL was present; it was sent for cytology. Both side fallopian tubes and ovaries were normal. Solid mass of 9 × 6 × 6 cm arising from uterine fundus above which large cystic lesion measuring 21 × 18 × 13 cm was

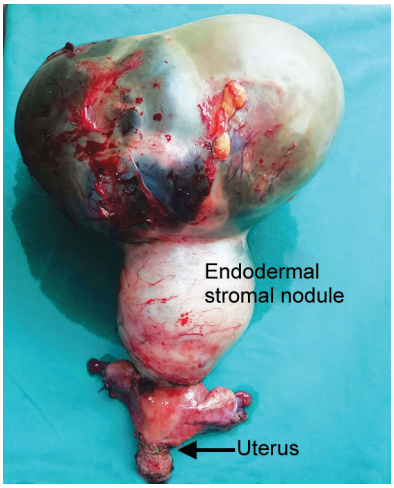

Fig. 1: Gross appearance of the tumor

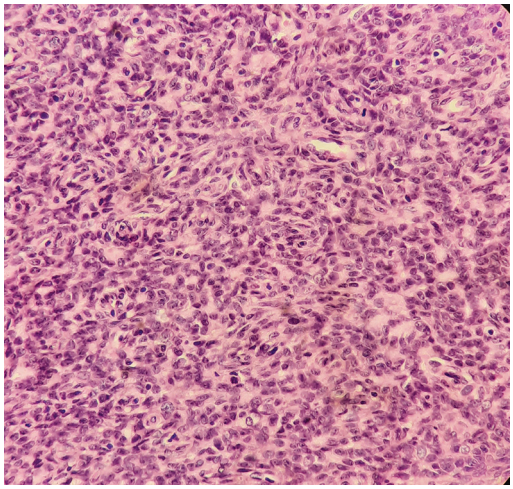

Fig. 2: Microscopic appearance of the tumor

seen (Fig. 1). Both solid and cystic masses were removed. Staging laparotomy was done (total abdominal hysterectomy + bilateral salpingo-oophorectomy + pelvic lymphadenectomy + infracolic omentectomy). Postoperative period was uneventful. Ascitic fluid cytology was negative for malignant cells. Uterus, cervix, bilateral tubes, bilateral ovaries, bilateral pelvic lymph nodes, and infracolic omentum showed no evidence of tumor cells. On gross examination—a large cystic and solid mass of size 21 × 18 × 13 cm with smooth and congested external surface was found. A multiloculated cyst filled with brownish fluid and a solid area measuring 9 × 6 × 6 cm was seen on cut section. Cyst wall thickness was 0.1–0.5 cm. The solid area was firm and grayish-white. On microscopic examination, sections from cystic and solid mass showed tumor cells that are round to polygonal cells with minimal cytoplasm, elongated nuclei with nuclear grooves which morphologically resembled EST (Fig. 2). The cells were positive for estrogen receptor (ER), progesterone receptor (PR), cluster of differentiation (CD 10), cluster of differentiation (CD 99), smooth muscle actin (SMA), and desmin. Since there was a suspicion of a malignant ovarian tumor, it was a solid and cystic mass, further analyzed with calretinin and PanCK immunohistochemistry markers. The tumor was negative for PanCK and calretinin which ruled out epithelial ovarian tumor and granulosa cell tumor, respectively. All features were consistent with endometrial stromal sarcoma (ESS). There was no vascular or lymphatic invasion that ruled out LGESS. Hence, overall morphological, microscopic, and immunohistochemical features were consistent with ESN. Preoperatively suspected malignant ovarian tumor case later turned out to be ESN.

Discussion

ESTs are rare tumors arising from the uterus. The incidence of EST is 2 per million women. WHO classified ESTs into four categories as follows: (1) ESN, (2) LGESS, (3) HGESS, and (4) USS. Endometrial stromal nodule are rare subtype of ESTs.[2]

Endometrial stromal nodule usually have absent or minimal myometrial invasion (≤3 mm and <3 protrusions) without any vascular invasion.[3]

Endometrial stromal nodule are benign tumors commonly seen in premenopausal age-group. The clinical presentations are abnormal uterine bleeding, pressure symptoms, and pelvic pain. There is no definitive test available to diagnose the tumor before surgery. Diagnosis is usually made postoperatively by histopathology and immunohistochemistry.

Endometrial stromal nodule occur commonly in the uterus than in cervix. They are well-circumscribed masses with soft consistency and characteristic yellow to orange color. In contrast, LGESS typically shows multinodular growth within the endometrium and myometrium. Cyst formation, ischemic necrosis, and hemorrhage may also be present. In this case, the cystic change was very large measuring 21 × 18 × 13 cm that made impression of ovarian tumor in preoperative imaging. They composed of endometrial cells with scant cytoplasm, uniform dark staining small round or oval nuclei, occasional mitosis, and fine granular chromatin. Lymphatic and vascular invasion are absent in ESNs. The most important criterion to diagnose ESN is non-infiltrating border of the tumor. But in the same case, there may be infiltrating irregular margins like lobulated or fingerlike projections into myometrium which are not ≥3 mm and are not >3 in number.[4]

Clinical Significance

It is difficult to diagnose ESN and LGESS based on endometrial curettage specimens. But it is important to differentiate the two types, since treatment and prognosis are different. The two main features that help us to differentiate ESN and LGESS are myometrial and vascular invasion. Hence, extensive sampling of the tumor margin is necessary. High-grade endometrial stromal sarcomaS can be differentiated from LGESS by gross appearance, microscopic features, mitotic state, and immunohistochemistry.

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