Case Report

A case report of bilateral ovarian luteinized thecoma with sclerosing peritonitis masquerading as a malignant mass

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ABSTRACT

Luteinized thecoma with sclerosing peritonitis (LTSP) is a very uncommon syndrome, characterized by the presence of single or bilateral ovarian thecomas and peritoneal fibrotic lesions. The disease occurs in young women, and it can lead to peritoneal fibrosis and bowel obstruction. We present the case of an ovarian mass in a 20-year-old woman. Radiologically, the patient had bilateral large, well-defined, heterogeneously enhancing solid mass lesions in the bilateral adnexa. The lesion extends into the lower abdomen, abutting the compressed fat planes. Her serum carcinoembryonic antigen (1.26 ng/mL) was within normal limits, but the cancer antigen 125 (CA125) (882/mL) level was high. Total abdominal hysterectomy with bilateral adnexal mass, along with part of the omentum, was removed. Histopathological diagnosis was LTSP and was confirmed by immunohistochemistry. Postoperatively, the patient was managed conservatively and was kept in follow-up.

Key words: Ovarian tumor, Peritoneum, Surgery, Thecoma

Luteinized thecoma with sclerosing peritonitis (LTSP) is a very rare disease with a prevalence of <0.01% of all ovarian cancers [1]. Less than 50 examples of LTSP have been histopathologically described in the literature to date, making it an uncommon condition. The condition was first described in 1994 [2]. LTSP is a rare ovarian tumor; despite its benign histology, it can lead to severe complications due to extensive peritoneal fibrosis. The condition often mimics malignancy, making an accurate diagnosis essential to avoid overtreatment. It is believed that fibrogenic substances released by luteinized cells may trigger the sclerosing response. Reporting such cases is vital to improve clinical recognition and guide appropriate management strategies [3]. The etiology is yet unknown. According to a few theories, neoplastic theca cells can produce substances that promote fibroblastic proliferation when released into the peritoneal cavity. In addition, it has been proposed that luteinized theca cells release progesterone and estrogen, two steroid hormones that target hormonally sensitive submesothelial fibroblasts. Cytokines, specifically transforming growth factor beta, are among the compounds generated. These cause a fibrotic response in the peritoneum and, in rare cases, even in distant anatomical locations, mimicking a metastatic state [4]. Clinically, the patient may experience discomfort in the abdomen and ascites. Acute or subacute signs of small intestinal obstruction may also be seen in a small percentage of patients. No indications of hormone imbalance can be seen [5]. In imaging, it may present as a predominantly solid and sometimes cystic mass with thickened contrast material enhancement in the peritoneal membrane [6]. Histologically, sheets of bland spindle cell proliferation arranged in short fascicles can be appreciated. Mitotic activity is usually prominent, and luteinized cells are arranged singly or in small nests with small to moderate amounts of clear or eosinophilic cytoplasm. Edema with extravasated erythrocytes, prominent small vessels, and microcystic areas is present, and spindled cells with fibrosis are seen in peritoneal/omental lesions [7]. Although surgery is the mainstay of treatment, resection by itself may not usually provide total disease control. While there have been reports of medical treatment attempts in recent years, a true standard therapy has not yet been established [4]. Numerous therapeutic options have been tried, including high-dose steroids, luteinizing hormone-releasing hormone agonists, and antiestrogens. Despite

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having potentially fatal consequences from sclerosing peritonitis, the disease entity is still classified as a benign entity [1,8].

CASE REPORT

A 20-year-old woman presented to us with abdominal swelling and mild pain with a history of spontaneous abortion 4 months back. The pain was constant throughout the duration and had no aggravating factors.

On general examination, the patient had no complaints of generalized weakness. On local examination, the patient had a firm palpable mass in the lower abdomen. The mass was not ballotable.

The patient had a normal routine blood profile, with a normal carcinoembryonic antigen (CEA) (1.26 ng/mL) and cancer antigen 19.9 (20.4 U/mL). However, the lactate dehydrogenase and CEA levels were raised with values of 521 U/L and 882 U/mL, respectively. B-human chorionic gonadotropin and alpha-fetoprotein were within normal limits with values of 2.39 U/mL and 6.22 IU/mL, respectively. The antinuclear antibody (ANA) was positive with a point titer of 1:320.

Contrast-enhanced computed tomography abdomen showed that the patient had bilateral large, well-defined, heterogeneously enhancing solid mass lesions in the bilateral adnexa, approximately measuring 94×90×83mm and 84×72×67 mm on the right and left adnexa, respectively. The lesion extends into the lower abdomen, abutting the compressed fat planes. Furthermore, there is the presence of diffuse omeno-peritoneal thickening on imaging. There was the presence of bilateral pleural fluid effusion with pleural thickening or calcification. On imaging, the lesion was suspected to be a case of ovarian malignancy with peritoneal carcinomatosis. However, on ultrasound-guided trucut biopsy from pelvic mass, a diagnosis of spindle cell lesion with a possibility of ovarian fibroma cannot be ruled out and was made outside of the present institute.

The patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy, and part of the thickened omentum was removed.

Grossly, both the ovaries had a lobulated surface with a smooth capsule. On cut section, bilateral ovaries were predominantly solid with solid cerebriform yellowish areas (Fig. 1). There was the presence of uniform thickening of the omentum without any formation of definite nodules.

Microscopically, the tumor comprised fascicles and sheets of plump spindle and oval cells that resembled the cells of theca interna. The cells had bland fusiform nuclei, fine chromatin, and clear vacuolated to pale eosinophilic cytoplasm. Variable number of fibroblastic cells, hyalinized connective tissue are interspersed among the tumor cells (Fig. 2a and b). Omental sections showed proliferations of spindle cells along with a few cells with clear cytoplasm in a whirling fashion (Fig. 2c).

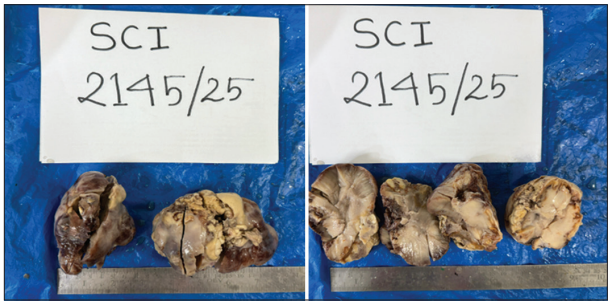

Figure 1: Showing ovarian outer surface and cut surface, respectively

On further immunohistochemistry, the leutenized cells were immunoreactive to calretinin and inhibin immunoreactivity. Further, they were negative for pan-cytokeratin (CK), ruling out any epithelial tumor or metastasizing tumor (Fig. 3).

Postoperatively, the patient was managed conservatively and was kept in follow-up. Regular imaging and clinical monitoring of the disease activity were noted, and the patient is stable to date.

DISCUSSION

Thecomas are relatively rare neoplasms (4% of all ovarian neoplasms) that belong to the sex-cord stromal group of tumors. It is closely related to fibroma, sometimes collectively called the fibrothecoma group. Most commonly occurs in young women (median age of presentation is 28 years) [9]. The tumors are usually bilateral, well-defined, capsulated, and have a lobulated appearance. On cut section, the tumor has predominantly grayish white solid areas with a typical yellow-tan bosselated appearance.

They are often associated with a peculiar condition called sclerosing peritonitis. The process of peritonitis consists of variable cellular proliferation of fibroblastic and myofibroblastic cells separated by collagen and fibrin, mesothelial cell proliferation, and occasionally mononuclear inflammatory cells. The etiology and pathogenesis are not clear [10]. Some proposed fibrosing soluble cytokines secreted by tumor cells to be responsible for the peritoneal manifestations [11]. The tumor cells are shown to have estrogenic activity and are responsible for menstrual irregularities and endometrial hyperplasia, even co-existing carcinomas in 15–30% cases [11].

The clinical presentation of thecoma varies according to the size of the mass; the larger ones typically present with abdominal swelling, pain, and ascites while smaller ones remain undetected. Those associated with sclerosing peritonitis are typically associated with multiple subocclusive symptoms and need recurrent laparotomy [1].

Microscopically, the lesion comprises fascicles of bland spindle cells with centrally placed nuclei and a moderate amount of pale vacuolated cytoplasm. Intervening tissue may show collagen deposition and hyaline plaque formation. A mitotically active variant has been discussed, which is particularly common in

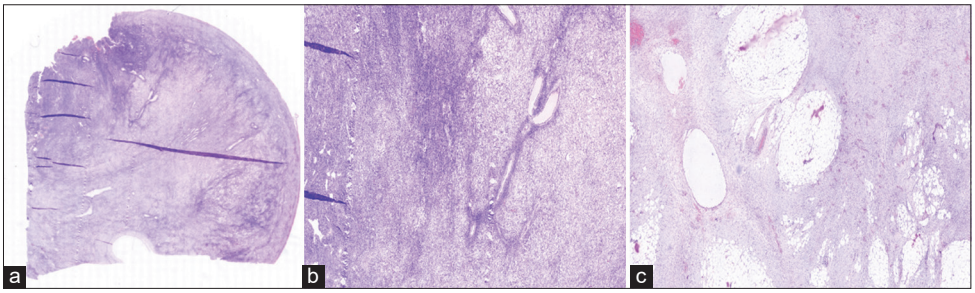

Figure 2: (a and b) Spindle cells with entrapped luteinized cells and presence of brisk mitosis, (c) peritoneal lesion with variable fibrosis

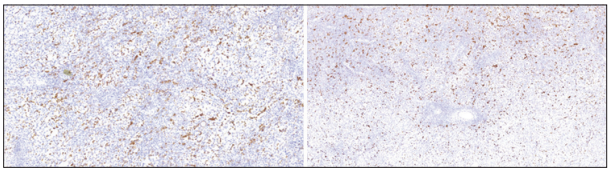

Figure 3: Immunohistochemistry for inhibin and calretinin

sclerosing peritonitis, but the clinical behavior does not seem to be affected by high mitotic count, which is particularly seen in this case [12].

Immunohistochemical studies have shown the tumor cells to be reactive with calretinin, smooth muscle actin, and desmin and variably with alphainhibin, epithelial membrane antigen, beta-catenin, CD34, and transforming growth factor-beta, with focal nuclear positivity for estrogen and progesterone receptor [7]. Our study found that the leutenized cells were immunoreactive for calretinin and inhibin immunostain, whereas they are negative for Pan CK. Differential diagnoses to be considered in this case were fibroma, sclerosing stromal tumor, stromal Leydig cell tumor, and secondary deposits in the peritoneum. In the present study the patient had raised Antinuclear Antibody (ANA) 1:320, and raised ANA can be seen associated with Leutenized thecoma [13].

Although the tumor is benign in behavior, complications due to sclerosing peritonitis may bring down the quality of life. Few studies advocated a conservative approach and followed up the patient for as long as 7 years without any morbidity [1,14]. Leuprolide and tamoxifen have been tried as targeted therapy with success [1]. The current approach is preservation of fertility either with oocyte extraction and preservation [15] or bilateral oophorectomy along with high-dose steroids and hormone suppression [8]. Post-operative complications due to short bowel syndrome and sepsis are frequent and are managed conservatively [1,16].

CONCLUSION

The cases of bilateral luteinized thecoma are seen in young women with a median age of presentation of 20 years. They have been reported to be associated with sclerosing peritonitis. Most often, they may be misdiagnosed as ovarian carcinoma (primary or secondary) with associated peritoneal carcinomatosis. We reviewed the literature and also re-evaluated the immunohistochemical analysis. Surgery represents the cornerstone of treatment, but resection alone does not always allow complete disease control. Attempts at medical treatments have been reported in recent years, but a real standard therapy has not yet been defined.

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