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Primary Broad Ligament Endometroid Adenocarcinoma: A Case Report and Review of Literature

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Introduction

Since the broad ligament is closely related to other structures, such as the uterus, fallopian tubes, and ovaries, secondary involvement of it due to primary malignancy elsewhere is common. Primary broad ligament malignancies are rare. Only 29 cases of primary broad ligament malignancies have been reported in the literature, out of which only 5 cases of endometrioid adenocarcinoma [1]. Generally, the primary broad ligament tumor diagnosis is made retrospectively, intraoperatively, or on histopathological examination, due to its rarity.

I have encountered a case where laparotomy and myomectomy were planned with a preoperative diagnosis of large subserous leiomyoma and intramural leiomyoma. Per operatively large friable mass was present between 2 leaves of the broad ligament on the right side, and a large adenomyoma was present in the posterior wall of the uterus. Subsequently, adenomyomectomy and excision of the broad ligament mass was done. On histopathology, the broad ligament mass was diagnosed as endometroid adenocarcinoma, and the uterine mass as an adenomyoma.

Case report

A 43-year-old nulliparous female presented to the clinic with complaints of amenorrhea, infertility, and mild pain in the lower abdomen for three months. She has been evaluated clinically, and on examination, a pelvic mass corresponding to 20 weeks of gestation was palpated. On further evaluation by ultrasonography and MRI pelvis: a large subserous fibroid from the uterus measuring approximately 10.6cm x 8.3 cm x 14 cm and an intramural fibroid of 7 cm x 6.1 cm indenting endometrium is seen. The right ovary was seen anterior to the subserous fibroid. The

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left ovary is seen in the pouch of Douglas. The cervix appears normal, and no enlarged lymph node is seen. Mild free fluid was noted in the pelvis. The patient was nulliparous and willing for future pregnancy, so the decision to perform laparotomy and myomectomy was taken. Per operatively, mild hemorrhagic ascites was present. A friable mass protruding from the posterior leaf of the broad ligament on the right side and an adenomyoma in the posterior wall of the uterus was present. Broad ligament mass was excised, and adenomyomectomy of uterine mass was done. Broad ligament mass was not attached to the uterus, right side ovary, or fallopian tube though they looked edematous. The left side tube and ovary were normal. As the mass was suspected to be malignant due to physical character omental biopsy was taken. On histopathology, the uterine mass was confirmed to be adenomyoma, but the broad ligament mass was diagnosed as endometrioid adenocarcinoma. Omentum was negative for any specified disease. She was referred to the oncosurgeon. Computerized Tomography (CT) chest, abdomen, and pelvis were performed further to rule out local and distant metastasis. In the CT chest -no metastatic foci were seen, and CT abdomen pelvis shows no liver metastasis, ascites, or abdominal lymph adenopathy. A large hypodense lesion, approximately 6.3 cm x 4.8 cm seen in the right parametrium and right adnexal region with soft tissue swelling noted, which was suspicious of residual disease. The surgical oncology team did total abdominal hysterectomy, bilateral salpingo-opherectomy, pelvic lymphadenectomy, and omental biopsy. On histopathology, foci of atypical endometriosis in the background of endometriosis in the right ovary and atypical glandular hyperplasia in uterine adenomyotic foci without any evidence of invasion were seen. Left ovarian endometriosis was present. Pelvic lymph nodes were negative without any evidence of malignancy in the sent specimens. The final diagnosis of primary broad ligament endometrioid adenocarcinoma was made. She received postoperative chemotherapy (carboplatin + paclitaxel). She has been doing well to date without recurrence for more than one year.

Discussion

Primary broad ligament tumors are defined by the proposed criteria given by Gardner et al., Primary location within or on the surface of the broad ligament but completely separated from the uterus, ipsilateral ovary, and fallopian tube [2].

Malignant tumors of the broad ligament are extremely rare; therefore, their etiopathogenesis, optimum management, follow-up strategy, and prognosis are still uncertain.

Patients usually present with vague complaints, pain or heaviness in the lower abdomen or a lump abdomen. Sometimes it may be silent and detected incidentally during evaluation for other problems. In our case, the patient came with complaints of amenorrhea, infertility, and mild pain in the lower abdomen. Initially, the decision for laparotomy and myomectomy was taken due to her willingness for future fertility.

Primary endometrioid carcinoma of the broad ligament is rare, and only 5 cases have been reported. Most of the epithelial malignancies of the broad ligament are derived from the Mullerian remnants, including (i) serous, (ii) mucinous, (iii) clear cell, (iv) endometrioid, (v) borderline tumors and (vi) endometrial stromal sarcomas. Among all the reported carcinomas, serous adenocarcinoma, clear cell adenocarcinoma, and endometrioid adenocarcinoma are the most common histologic variants [2]. Other rare malignancies reported to arise from the broad ligaments are (i) Sex cord-stromal tumors, (ii) Neuroendocrine carcinomas (small cell and large cell), (iii) Sarcomas and (iv) Female Adnexal Tumor of probable Wolffian Origin (FATWO) [1]. In our case, evidence of endometriosis in both ovaries with foci of atypical changes and adenomyosis with atypical changes have been seen. The development of primary broad ligament endometrioid carcinoma could be hypothetically explained by the malignant transformation of long-standing endometriosis of the broad ligament, as endometriotic involvement of the broad ligament is common. Malignant transformation occurs in 0.7-1% of patients with endometriosis, particularly in the ovaries [3]. The most frequent sites of extragonadal involvement are the rectovaginal septum, the colon, and the vagina [4]. ovary is the primary site in 79 percent of cases, and extragonadal sites are identified in 21 percent [5]. Two cases of endometrioid carcinoma and a case of clear cell carcinoma developed in the diaphragm were reported. Both patients had a history of endometriosis/adenomyosis [6]. A rare endometrioid carcinoma developed from deep infiltrating endometriosis in the uterosacral ligament six years after treatment for an atypical proliferative endometrioid tumor of the ovary in a 48-year-old woman reported [7].

Management and prognosis of broad ligament endometrioid adenocarcinoma are still uncertain. Most of the cases patients were managed by total abdominal hysterectomy with bilateral salpingectomy with pelvic lymphadenectomy and/or peritoneal and omental biopsies. The role of Postoperative chemotherapy/radiotherapy is still not established due to the paucity of cases. As histopathologically they have similarities with ovarian endometrioid carcinoma, most authors advise adjuvant platinum-based chemotherapy (paclitaxel and carboplatin) [1]. Our patient received a similar regimen. Most cases of broad ligament cancer were assumed to be in the early stage (PT1) and have a favorable prognosis; 5-year survival rates of 80-90% were reported [8] aggressive progression is defrayed, and metastasis is uncommon because they lack their blood supply, and lymphatic drainage in contrast to ovarian cancers [9].

Primary broad ligament malignancy should be a differential diagnosis of uterine, adnexal masses. Both clinicians and radiologists require a high index of suspicion to diagnose broad ligament malignancies pre-operatively. In a few cases, CA 125 was found, but this is a non-specific marker raised in several benign and malignant gynecological and non-gynecological conditions. There is no specific biomarker has been established for endometrioid adenocarcinoma (ovarian or nonovarian origin). Several epidemiological studies observed the close association of endometriosis with particular histology of endometriosis-associated ovarian cancer, endometrioid, and clear cell carcinoma, especially when the diagnosis of endometriosis was more evidence-based, such as through surgical-pathological diagnosis [10]. The overall rate of malignant transformation in endometriosis has been estimated to be 0.3-0.8%, with a relative risk ranging from 1.3 to 1.9 [11] and should be suspected if a suspicious mass is found even at the extragonadal site in a patient with longstanding endometriosis or with symptoms suggestive of endometriosis, so that timely diagnosis ensures expeditious and efficient management and favorable prognosis.

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