Case Report

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Ovarian Lymphangioma with Mature Cystic Teratoma

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ABSTRACT

Lymphangiomas are slow-growing tumors that remain asymptomatic for a long time, with the tumor being identified incidentally during histopathological examination after excision. Mature cystic teratoma is benign tumor consisting of mature tissue derived from two or three germ layers. We have 47-year-old woman who underwent total abdominal hysterectomy for right adnexal mass. As her ultrasound report revealed a right adnexal mass with solid and cystic components. The histological analysis along with immunohistochemistry (D2-40) maker confirmed the diagnosis of lymphangioma of the ovary coexisting with mature cystic teratoma. There is paucity of reported case of co-existing these two tumors in same tissue.

Keywords: D2-40; lymphangioma; mature cystic teratoma; ovary.

INTRODUCTION

Lymphangioma of the ovary is a rare lesion. It was first described in 1908[1] and there are only 19 cases reports in 50 year literature survey.[2] Mature cystic teratoma (MCT) is a germ cell neoplasm composed of well-differentiated derivations from two of the three germ cell layers. Mature cystic teratomas account for 15% of all ovarian neoplasm.[3] MCT tends to be identified in young and middle-aged females.[4] These tumors are usually benign and unilateral but can be bilateral in 10-15% of cases.[5]

A collision of tumor means coexistence of two tumors adjacently, but histological different features in the same tissue. Here, we have reported an extremely rare presentation of collision tumor in the ovary showing histology features of lymphangioma and mature cystic teratoma.

CASE REPORT

A 47 years woman presented to the gynecology outpatient department with complains of pain abdomen for 6 months and per-vaginal discharge for 1 week. All blood parameters were normal. Vaginal examination revealed 6-8 weeks uterus size. USG report revealed fibroid uterus; right adnexal mass with solid and cystic areas. Then, the patient underwent total abdominal hysterectomy with bilateral salphingo-oophorectomy. The postoperative course was uneventful.

Macroscopically, uterus showed intramural fibroid with unremarkable cervix and normal tubes. Right ovary showed solid, firm and cystic structure, measured 9.5x4.5x3cm, cystic part measured 5.0x4.0x3cm, cut-section showed unilocular cavity containing sebaceous materials and hair. The solid, firm part measured 4.5x3.0x2.5cm, cut-section showed multiple small variably size cystic spaces measuring between 0.2cm-0.4cm in diameter containing serous like fluids (Figure 1). The left ovary measured 3x2x0.9cm, with a smooth outer surface and on cut section, showed a single tiny cystic cavity, measuring 0.5cm in diameter, containing clear fluids.

Figure 1. Gross picture of ovarian tumor showing solid and cystic component.

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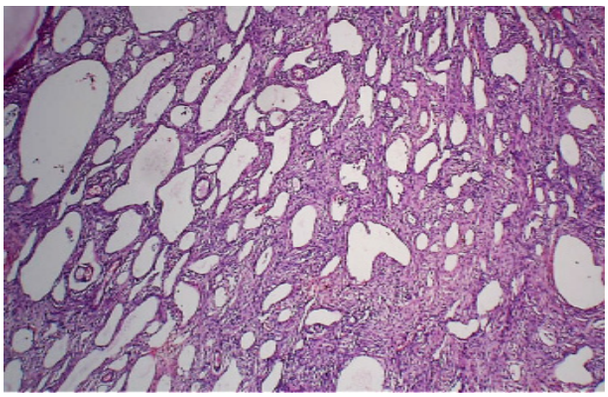

Figure 2. H & E stain: 400x shows thin walled multiple vascular space of varying sized lined by flattened endothelial cells which are separated by fibrocollagenous septa.

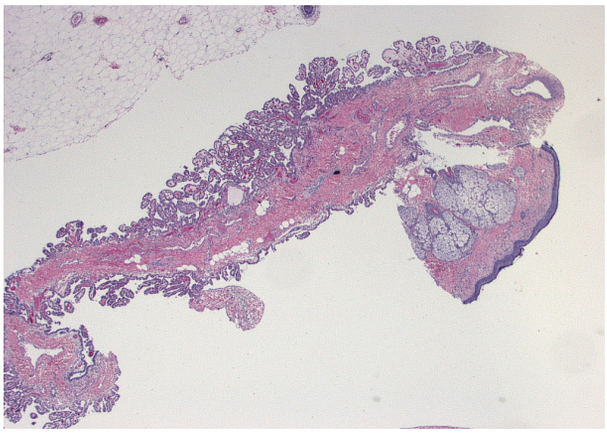

Figure 3. H&E stain 400x cystic structure lined by stratified squamous epithelium and dermal appendages.

Microscopically, It showed proliferative phase endometrium along with features of adenomyosis, leiomyoma and chronic nonspecific cervicitis. Tubes were normal and cystic follicle in left ovary. Right ovary showed thin walled multiple vascular spaces of varying sizes lined by flattened endothelial cells; separated by fibrocollagenous septa infiltrated by mononuclear cells. Many of these lymphatic spaces contain eosinophilic proteinaceous like materials, lymphocytes and occasionally erythrocytes (Figure 2). On immunohistochemistry examination, the lining epithelium cells of the lymphatic spaces showed positively for D2-40 and negative for CK and HMB45. The sections from adjacent cystic structure showed features of mature cystic teratoma consisting stratified squamous epithelium, dermal appendages and adipose tissue (Figure 3).

With all these features final impression was made as lymphangioma coexisting with mature cystic teratoma in right ovary.

DISCUSSION

The lymphatic system consists of a network of uni-directional vessels that collects excess fluid from the interstitium, to the regional lymph nodes and ultimately drains to the venous system via the thoracic duct.[6] The lymphatic vessels are seen in all organs except in the brain, anterior chamber of the eye and in systems with rich sinusoidal networks such as bone marrow and spleen. Lymphangioma are usually found in the skin, genitalia, or in the head and neck region. Visceral lymphangiomas are infrequent but have been reported in various organs. It has been reported at different sites in the female genital tract i.e. cervix, uterus and fallopian tubes.

Lymphangiomas of the ovary are very rare and usually encountered as incidental findings, mostly located on the surface of the ovary or in the parenchyma. They are usually unilateral; occasionally bilateral can be seen and measured up to 6cm in diameter. And in our case its diameter was 4.5 cm.

Pathogenesis of lymphangiomas is uncertain. Some authors thought they are true neoplasms. According to well-established theory, sequestration of lymphatic tissue during embryonic development can cause lymphangiomas. But it can be seen in acquired lesion, like in lymphatic obstruction occurs due to infection, surgery or radiation. Since, in our case there was no evidence of infection and no obstructive pathology noted in the surrounding lymphatics, we believe the lesion might be of hamartomatous origin.

The main differential diagnosis of lymphangioma is adenomatoid tumor. Adenomatoid tumor is a benign solid tumor of mesothelial origin, which on microscopic features shows glandular structures lined by cuboidal epithelium and positive staining for low molecular weight cytokeratin and is negative for endothelial marker (CD-34). In our case cytokeratin was negative on immunohistochemistry examination showing D2-40 positive. Another differential diagnosis includes hemangioma, which unlike lymphangioma, contains RBCs within the vascular spaces. Such features are also absent in our case.

In our case, there was a coexistent of two types of tumors in same ovary. One is mature cystic teratoma and another one is Lymphangioma which is very rare findings of coexisting tumor. Bostanci MS.[7] had also reported rare

histological combinations of two tumors in their articles.

Teratomas, called dermoid cyst, predominantly occur in young women. They account for 10-20% of all ovarian tumors and are bilateral in 10 to 15% of cases.[8] They arise in the ovary but can be located at the midline and in paraxial regions of the body, rarely in lungs or ilea.[9] In our case it was present in ovary.

Pathologically, MCT are composed of tissues derived from one or more of the three primitive germ layers. Typically it contains mature tissues of ectodermal, mesodermal (muscle, fat) and endodermal origin.[10] The initial biological event that leads to teratoma is not yet understood. Stenens et al. [11] postulated that teratomas were derived from oocytes that undergo maturation and spontaneous parthenogenic activation followed by embryonic development within the ovarian follicles. MCT is usually asymptomatic and doesn't have any specific symptoms. Malignant transformation occurs in 1% of cases.[12]

As both tumors are benign and completely excised; with no postoperative complication and patient was discharged with expectation of no adverse consequences in future.

CONCLUSIONS

Ovarian lymphangioma is rare lesions of ovary and even rare is collision of this rare variant of tumor with other tumor. Here, we demonstrated coexistence of lymphangioma with mature cystic teratoma in same tissue. Both tumors have low malignant potential. One should keep in mind that; collision tumor can occur when there is solid and cystic component in the same tissue.

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