

Bilateral Lymphangioma of Ovary-A Rare Case Report

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Abstract: Lymphangiomas are rare benign tumor of lymphatic system arise in any part of the body. Lymphangioma of the ovary is an extremely rare lesion and usually unilateral and asymptomatic. Ultrasound report was suggestive of bilateral complex cysts of ovary possibly tuberculosis. Milky fluid obtained from the pouch of douglas. Histopathological examination revealed bilateral benign lymphangioma of ovary with multiple epidermoid cysts. Immunohistochemistry examination showed positive for CD31 and CD34 and negative for Cytokeratin. Differential diagnosis of adenomatoid tumor and hemangioma was ruled out. No evidence of reactive and inflammatory process suggestive of hamartomatous etiology.

Keywords: Lymphangioma, Bilateral, Immunohistochemistry.

INTRODUCTION

Lymphangioma is benign lesion of the lymphatic system characterised by multiple cystic spaces lined by endothelium and contains serous or chylous fluid [1]. It mainly arise in head, neck, axilla, superficial cutaneous or intraabdominal [2]. Lymphangioma of the ovary is extremely rare and which is usually unilateral. Bilateral lymphangioma has been reported very rarely in literature [3-5]. Ovarian lymphangioma are mainly asymptomatic and usually an incidental finding during routine gynecological procedure [6]. Earlier reports suggested it to be neoplastic but now it is believed to be hamartomatous [7]. We present a rare case of bilateral lymphangiomas arising in a 37 year old lady.

CASE REPORT

A 37 year old female presented with complaint of abdominal pain and dysfunctional uterine bleeding in the gynecology OPD. She had two previous normal deliveries. There was no difficulty and complications during and after the delivery. Hematological and biochemical findings were normal. Ultrasonography was done by the gynecologist which was suggestive of bilateral ovarian complex cyst with solid and cystic component favoring tuberculosis. Serum CA 125 was within normal limit. She was planned for total hysterectomy with bilateral salpingoophorectomy. During surgery bilateral multiloculated cyst filled with cheesy material was seen with multicystic ovaries. During surgery chylous fluid was seen in the pouch of douglas.

Hysterectomy specimen was sent to us for the histopathological examination. Uterus and cervix with

bilateral salpingoophorectomy weighing 120 gm with uterus and cervix measuring 6.5x4.5x2cm and one ovary measuring 3.5x3x2.5cm and the other measuring 4.5x3x3cm. External surface of both ovaries showing multiple nodules filled with thick chylous fluid (Fig 1).



Fig-1: Hysterectomy with bilateral ovaries showing multiple cystic lesions

Histopathological examination revealed cystically dilated channels lined by endothelial cells and are filled with pink eosinophilic material and few lymphocytes and occasional red blood cells. Stroma is showing fibrous tissue (Fig 2). No obstructive pathology or no evidence of inflammation of infection found in the fallopian tube.

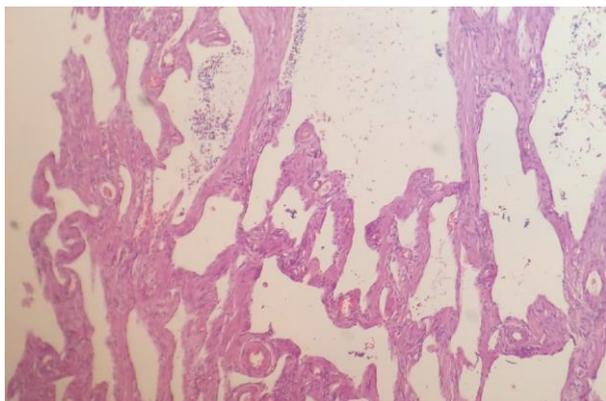


Fig-2: Multiple cystically dilated channels lined by endothelial cells filled with pink eosinophilic material with lymphocytes and occasional red blood cells

Immunohistochemical examination showed positivity for CD31 (Fig 3), CD34 is positive and Cytokeratin was negative (Fig 4) suggestive of lymphangioma. Adenomatoid tumor and possibility of hemangioma was ruled out.

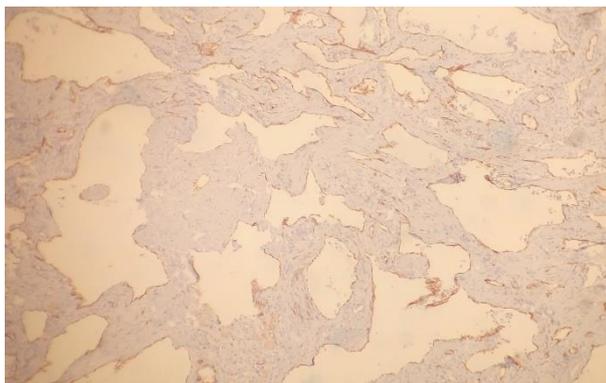


Fig-3: Endothelial linings are positive for CD 31

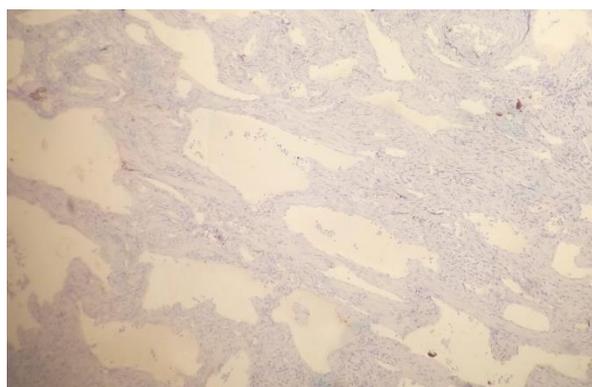


Fig-4: Cytokeratin negative

DISCUSSION

Lymphatic system is formed by network of unidirectional vessels that collects excess fluid from the interstitial tissue which is transported to the regional system and ultimately draining to the venous system through thoracic duct. The lymphatic system present in

all organs except in the brain, anterior chamber of eye, bone marrow and spleen [5].

Lymphangiomas are rare malformation which arises from the sequestration of lymphatic tissue which fails to communicate with lymphatic system [3].

Pathogenesis of lymphangioma is uncertain and matter of discussion. Some authors thought they are true neoplasm [7]. All order to well establish theory, sequestration of lymphatic tissue during embryonic development cause lymphangioma. Various authors thought that impaired regional lymphatic drainage due to chronic salpingitis or radiation therapy may be the cause for cystic lymphangioma of ovary [8]. In this case there is no evidence of chronic salpingitis or any history of radiation therapy. Patient showed chyle accumulation in the pouch of douglas. So in the present case sequestration of lymphatic tissue during embryonic development or neoplastic nature may be the possible pathogenic mechanism.

Intraabdominal cystic lymphangioma can be discovered as an incidental finding during laparotomy for another condition, or they can manifest as a chronic or acute abdomen. Chronic symptoms include progressive abdominal distension and pain. Mimics of ovarian cystic masses include peritoneal inclusion cyst, paraovarian cyst, mucocele of the appendix, obstructed fallopian tube, spinal meningeal cyst, lymphocele, cystic degeneration of lymph nodes, lymphangioliomyomatosis, hematoma, and abscess. Ovarian lymphangioma are asymptomatic and incidental findings and may cause some abnormality in the menstrual cycle. In our case there was a dysmenorrhea and abdominal pain with collection of chyle in the pouch of douglas.

USC is the first step of diagnosis. A cystic lesion with multiple thin septa is the typical USG finding of lymphangioma [8]. In our case also there was complex cyst with thin septa seen in bilateral ovaries. Provisional diagnosis of tuberculosis was suspected after USG.

Histopathologically differential diagnosis of lymphangioma of ovary is a adenomatoid tumor and hemangioma [6]. Adenomatoid tumor is the benign solid tumor of mesothelial origin affecting the female genital tracts. Immunohistochemistry is a diagnostic tool to differentiate these three entities. CD31, CD34 are positive in the hemangioma and lymphangioma . While positivity of Cytokeratin will confidently excludes the diagnosis of the lymphangioma. Adenomatoid tumor is cytokeratin and calretinin positive neoplasm. As this case showed CD31, CD34 positive and Cytokeratin negative, diagnosis of Lymphangioma was confirmed.

CONCLUSION

Ovarian lymphangiomas are rare cystic lesions and should be considered as a differential diagnosis of cystic masses ovary. Clinical symptomatology is polymorphic and not specific. These lesions may be missed diagnosed radiologically hence histopathological and immunohistochemical examination is must for the diagnosis of this lesion. The treatment of choice is surgical and with full resection of the lesion.

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