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Case Report

Lymphangioma of ovary: A case report

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ABSTRACT

Ovarian lymphangioma are unusual benign tumours characterized by proliferation of lymphatic channels. The exact etiology and the true occurrence is not well understood. We here in discuss a case of ovarian lymphangioma in 45 yr old female who presented with chief complains of abdominal pain. USG revealed features suggestive of right complex ovarian cyst, that was then excised and send to pathology department for histological analysis. Histopathological examination revealed an ovarian lymphangioma which was further confirmed by IHC marker CD31 that highlighted the endothelial nature of lymphatic channels. Patient was disease free 6 months post surgery and is on regular follow up till date.

1. Introduction

Lymphangiomas is a congenital malformation that is characterized by proliferation of lymphatic channels,[1] composed of cystic spaces lined by single layer of endothelial cells which is separated by fibrocollagenous septa. Most common site of lymphangiomas in childrens being the head and neck, while in adults its commoner in superficial skin and intraabdominal site.[2] Majority of ovarian lymphangiomas are slow growing tumours that remain asymptomatic for a long time, while the tumour being detected incidentally during histopathological examination after excision. The incidence of visceral lymphangiomas are however infrequent with ovary being one of the uncommon sites. Extensive search in Pubmed data base reveals 23 cases of ovarian lymphangioma been reported till date..

2. Case Summary

A 58 yr old female, presented to OBG outpatient department with abdominal pain on and off since one month. Her physical examination showed no visible lump and her past history was also not significant. Ultrasound examination showed a multiloculated cystic mass inclined to the right adnexa measuring 12.5cmx9 cm x2 cm. There was no vascularity within the lesion. Features were suggestive of right ovarian complex cyst, probably neoplastic. Serum tumor markers CA125, beta HCG and alphafetoprotein were within normal limits. Other systemic examination was within normal limits with no enlarged pelvic or aortic lymphnodes. The patient underwent abdominal hysterectomy and the tissue was sent to our department for histopathological analysis. Grossly we received two labelled containers. Container 1- labeled as uterus and cervix with unilateral appendage(left ovary and tube) measuring 8x5x2 cm. (Figure 1 A) Left side ovary measured 5x4x1cm (Figure 1 B) Endometrial thickness was 1mm, myometrium grossly appeared normal. Sections were given from cervix, endometrium, myometrium and attached left tube and ovary that appeared normal on gross. Container 2- labeled as right

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ovarian mass – grossly was cut opened cystic structure measuring 11x9x2 cm, that was multiloculated containing approximately 5 ml of serous fluid with variable wall thickness. Three sections were given from different areas. Histopathologically sections from cervix showed features of non specific cervicitis, left tube showed features suggestive of non specific salpingitis and endometrium was reported as non secretory with unremarkable myometrium. Sections from right ovary revealed dialated cystic spaces lined by flattened endothelial cells containing lymphocytes and pale eosinophilic material in the lumen (Figure 2 A, B, C, D). IHC markers CD31 was done that was positive highlighting the endothelial nature of lymphatic channels and thus confirmed the diagnosis of lymphangioma of right ovary.(Figure 3 A, B, C) Sections from left ovary showed features consistent with serous cystadenoma.

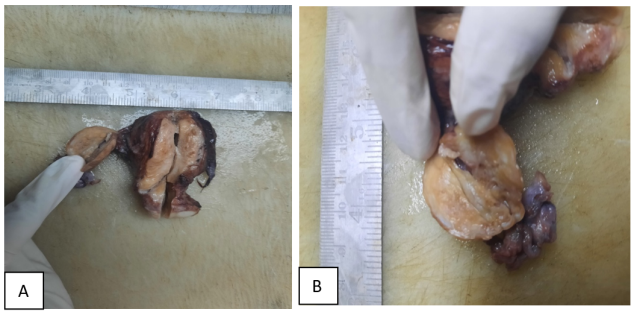

Figure 1: A): Gross pic-uterus and cervix along with left adnexa measuring 8x5x2 cm; **B**): Gross pic-left ovary measuring 5x4x1 cm

3. Discussion

Lymphangiomas are rare benign tumours of the lymphatic system comprised of multiple closely packed thin walled vascular spaces lined by endothelial cells containing lymphocytes and pale eosinophilic material in the lumen called lymph. The stroma contains lymphoid aggregates. Based on the size of lymphatic vessels they are classified as capillary, cystic or cavernous and contain either serous or chylous fluid.[3]

Visceral lymphangiomas are rare, and if they occur they are mostly in intestine and mesentry. Lymphangioma of ovary is an extremely rare entity that was described by Kroemer in 1908.[4] Due to its low occurrence, its pathogenesis is poorly understood. Few authors suggested that it is a hamartomatous process due to secretion of lymphatic tissue during embryonic development while others believed it that they are true neoplasm. The cause of acquired lymphangiectasia leading to lymphangioma in adults could be due to proliferation of lymphoid nests after inflammation, genetic predisposition, degeneration of lymphnodes, end result of mechanical pressure, trauma, radiation therapy or disorders of lymphatic vasculature.[5] These etiologies were identified as precipitating factors in various case study while analysing the literature review. In a case study, the lymphatic drainage disorder was the cause of ovarian lymphangioma who had chronic follicular salpingitis.[6] In another study, radiation due to wilms tumor was the cause of lymphangioma in children.[7] Deteriotion of lymphatic channels due to a 35cm leiomyoma was the cause of lymphangioma in a post menopausal women as described in the study done by Akyildiz et al.[8] In our case no such precipitating factors was there that ruled out the cause of reactive process as the etiology but rather favoured a neoplastic cause.

Lymphangiomas of ovary are usually unilateral, however few cases of bilaterality were reported in literature.[9] They are most commonly seen in adult women but can rarely occur in children. Ovarian lymphangiomas are located on the surface of ovary or in the parenchyma, and occasionally grows large and cause mass effect leading to compression of adjacent organs and resulting in abdominal pain and distension as a presenting complaint. Sometimes they are very large in size, where it mimicks a malignant ovarian mass leading in unwarranted radical surgery or over treatment.

The differential diagnosis to be considered are hemangioma, teratoma with prominent vascular component and adenomatoid tumor. Histologic appearance of lymphangioma is similar to hemangioma but latter has more red cells within the vascular lumen, lined by continous endothelial cells and lacks lymphocytes within the wall. The absence of other elements in the present case ruled out the possibility of teratoma. Adenomatoid tumor is a solid tumor of mesothelial origin characterised by cystic spaces lined by continous cuboidal to flattened mesothelial cells and do not contain lymphocytes in the stroma. IHC shows negativity for endothelial markers, where as lymphangioma shows positivity for endothelial markers. Even though malignant transformation in lymphangioma is rare, few case studies conducted by Rice et al.[10] and Aristabal et al[11] have described lymphangioma with malignant counterparts. The role of IHC is optional however it highlights the endothelial nature of lymphatic channels. IHC that was positive in our case was CD31.

4. Conclusion

The case is reported to highlight its rarity and to consider lymphangioma in the list of differential diagnosis of multicystic lesions of ovary. Diagnostic accuracy solely depends on histopathology and IHC confirmation. Complete wide excision with clear margins and a close follow up is essential to prevent possibility of recurrence in the future. Our patient is on regular follow up since last 6 months and is disease free without any fresh complaints.

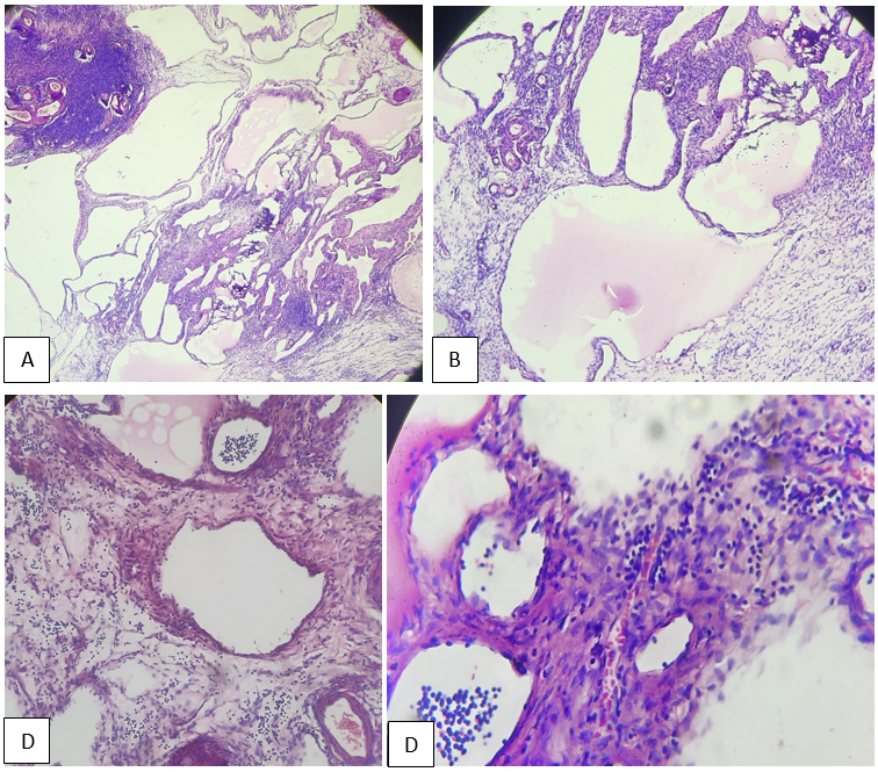

Figure 2: A): Scanner view 40x-Dialated cystic spaces; **B**): Low power view 100x-dialted cystic spaces containing eosinophilic secretions in the lumen; **C**): Low power view 100x-cystic spaces containing lymphocytes; **D):** Highpower 400x- showing lymphocytes within dialated cystic spaces

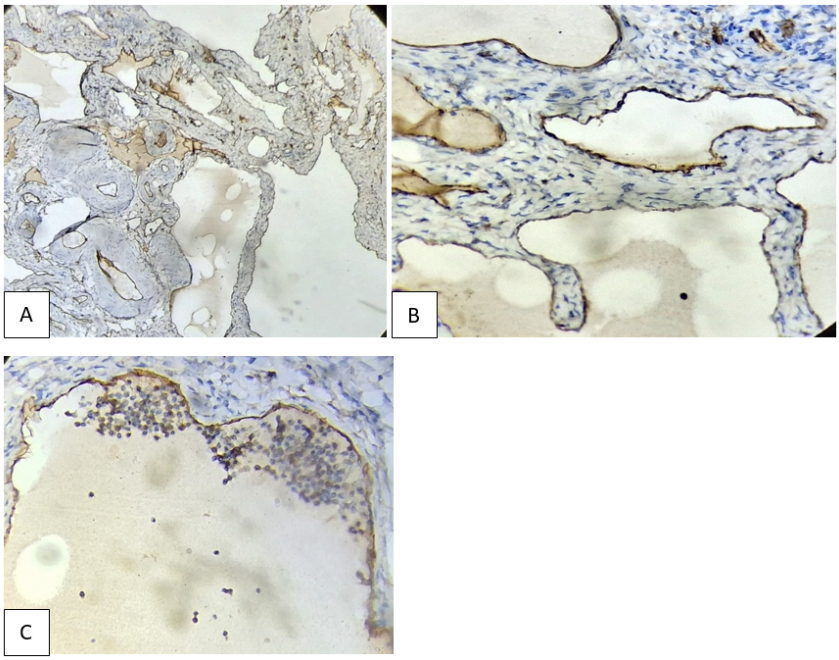

Figure 3: IHC CD31- **A**): Scanner view 40x; **B**): LP 100x, 3; **C**): LP 100X- Highlighting endothelial nature of lymphatic channels

5. Source of Funding

None.

6. Conflict of Interest

None.

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