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Case report

OSSIFYING RETROPERITONEAL CYSTIC LYMPHANGIOMA IN A PREGNANT WOMAN

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ABSTRACT

Retroperitoneal cystic lymphangiomas are very rare lesions and may be misdiagnosed. Longstanding lymphangiomas may show secondary changes like inflammation, hemorrhage, fibrosis and rarely ossification. Treatment is complete surgical excision. We are reporting a rare case of ossifying retroperitoneal lymphangioma in a pregnant woman which was misdiagnosed clinically as ovarian tumor. Our diagnosis was confirmed by IHC-CD-31 and D2-40 positivity. Postoperative follow up for 3 years, patient is fine and she is full term pregnant now.

Keywords: Retroperitoneum, Cystic lymphangiomas, Ossification, Pregnant woman, IHC D2-40 and CD-31.

INTRODUCTION

Lymphangiomas are rare benign cystic neoplasms of the lymphatic system, common in children but rare in adults ^[1]. They are thought to occur due to obstruction of the local lymph flow. Approximately 95% of the lymphangiomas are found in head neck and axilla, and 5% appear in other regions like the lungs, pleura, pericardium, esophagus, stomach, jejunum, colon, pancreas, liver, gallbladder, kidney and mesentery ^[2]. 1% of all lymphangiomas are located in retroperitoneum and nearly 186 cases have been reported ^[3]. The most common presentation of lymphangioma is that of a soft, fluctuant mass that enlarges, remains static, or waxes and wanes during the period of clinical observation ^[4]. We report a case of ossifying retroperitoneal cystic lymphangioma in a pregnant woman.

CASE REPORT

A 28 years old woman, gravida 2, para 1, presented with history of 3 months amenorrhea and mass in the left side of the lower abdomen since two years, but painful since three months. Clinically it was thought to be an ovarian tumor. USG showed a

retroperitoneal cystic mass. The mass was enucleated and pregnancy was terminated. The mass was sent for histopathological examination to the department of pathology KBNIMS, Gulbarga. Grossly it was a large globular mass, grey white to grey brown in colour, measuring 15x12x10 cms, well circumscribed. Cut surface showed multiple cysts of varying sizes (0.3 to 2.5cms), filled with cloudy fluid and showed hemorrhagic areas (Fig-1).

Microscopy showed multiple cystic spaces, these cystic spaces were lined by flattened endothelial cells (Fig-2). Most of the cystic spaces were filled with eosinophilic material mixed with lymphocytes (Fig-3). Stroma showed dense lymphocytic infiltration, areas of hemorrhage, fibrosis and ossification (Fig-4). Immuno- histochemistry was done showed positive for D2-40 (Fig-5) and CD-31 (Fig-6). Our final diagnosis was ossifying retroperitoneal cystic lymphangioma. Postoperatively patient is fine. After one year she conceived and at present she is full term and healthy.

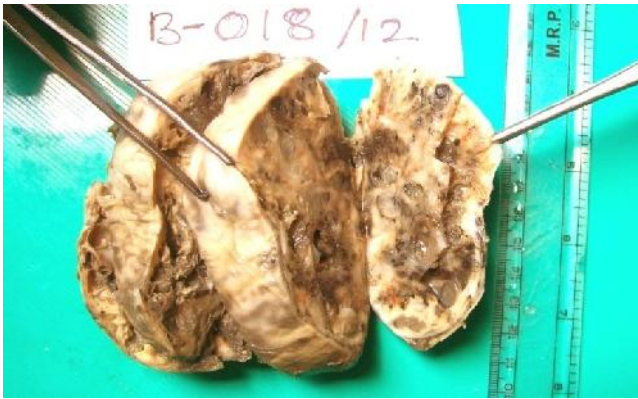


Fig 1: Cut surface of large grey-white mass showing multiple cystic spaces and grey-brown hemorrhagic areas.

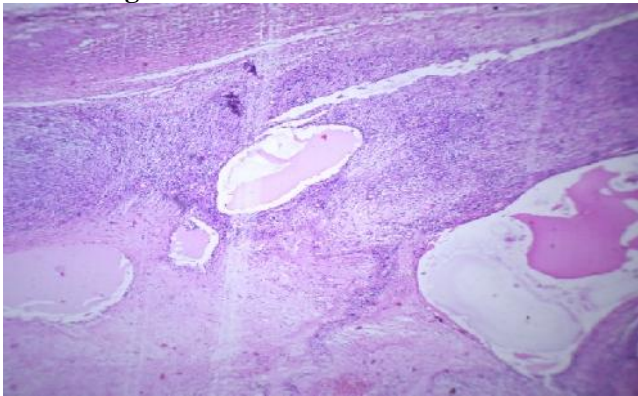


Fig 2: Microscopy showing multiple large cystic spaces filled with eosinophilic fluid, separated by areas of fibrosis. 5x10

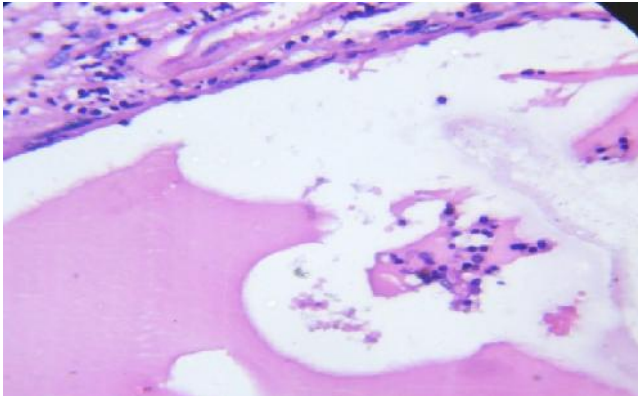


Fig 3: Microscopy showing a large cystic space lined by endothelial cells and filled with eosinophilic fluid containing lymphocytes. 40x10

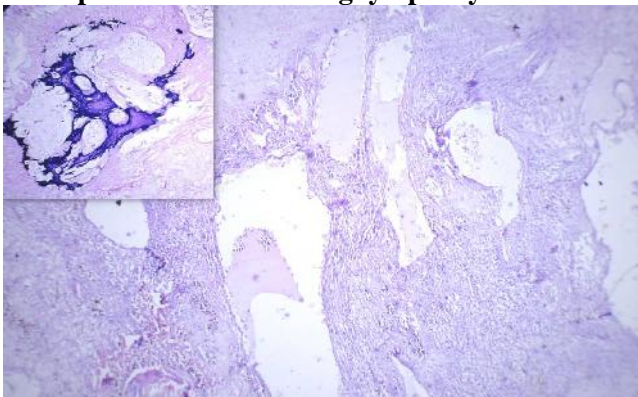


Fig 4: Microscopy showing cystic spaces, areas of fibrosis and inset showing focus of ossification. 10x10

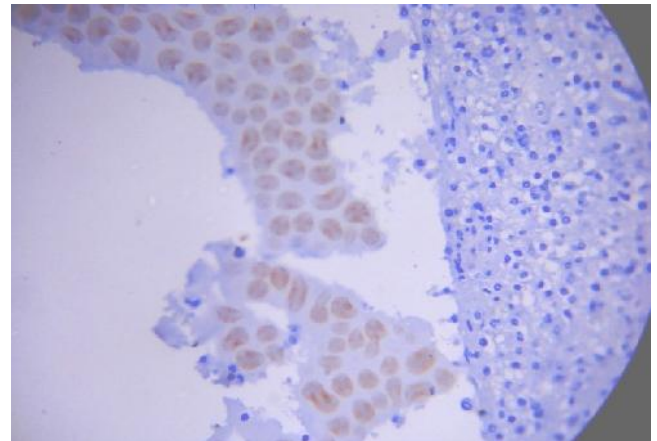


Fig 5: Microscopy showing IHC D2-40 Positivity in the exfoliated endothelial cells. 40x10



Fig 6: Microscopy showing IHC CD-31 Positivity in the lining endothelial cells. 10x10

DISCUSSION

It is often difficult to state whether Lymphangiomas are true neoplasms, hamartomas or lymphangiectasias. They are all benign lesions and symptoms depend on their location and extent ^[1]. Cystic lymphangiomas more commonly occur in the head and neck region and axillary region and very rarely in the retroperitoneum. The most characteristic radiologic finding is a large mass containing fluid with or without septa ^[5]. Lymphangiomas are thought to occur due to obstruction to the local lymph flow ^[2]. Several forms of lymphangiomas –including simple capillary, cavernous and cystic have been described ^[2]. The common presentations of intra-abdominal cystic lymphangiomas are abdominal mass (as in our case) and distension ^[6]. Complications of retroperitoneal lymphangiomas are bleeding, rupture, ascitis, infections, torsion and rarely obstruction of ureter and hematuria ^[3]. Jeong HK, et al, reported a

case of giant retroperitoneal lymphangioma which showed focal bony ossification^[7]. A M Buccoliero and V. Maio reported a case of calcified cystic lymphangioma of the mesentery^[8]. R J I Boskar and E H Eddes reported a case of mesenteric lymphangioma showing calcification and ossification^[9]. Prognosis of lymphangiomas is good because they rarely become malignant^[1]. Complete surgical enucleation is the treatment of choice^[10]. Rarely post-operative recurrence may occur^[11]. Differential diagnosis of a retroperitoneal lymphangioma includes teratoma, ovarian cystadenoma, retroperitoneal hematomas, metastatic lymphadenopathy, fibrosarcoma, leiomyosarcoma and liposarcoma^[12]. A higher index of suspicion and a simple ultrasonography may lead to an earlier correct diagnosis in many of the patients^[13].

CONCLUSION

We report this case of ossifying retroperitoneal cystic lymphangioma in a 28 years old pregnant woman. It has not yet been reported in a pregnant woman.

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Conflict of Interest: Nil

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