

# Rare Case of Ovarian Cystic Lymphangioma Managed at Laparoscopy

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## ABSTRACT

Lymphangiomas are rare, usually benign tumors of the lymphatic system. Lymphangiomas may arise in any part of the body. Lymphangioma of the ovary is very rare tumor, which is usually slow-growing, remains asymptomatic for a long time, and identified incidentally at histopathological examination after excision. It is advisable to excise the lesion with microscopically clear margins. Many times, diagnosis becomes difficult because of confusion with malignant ovarian mass, especially in post-menopausal woman. A 72-year-old post-menopausal woman had a symptomatic pelvic mass. Imaging studies demonstrated a complex right ovarian cyst. Laparoscopic total hysterectomy with bilateral salpingo-oophorectomy was performed successfully. Cystic Lymphangioma should be included in the differential diagnosis of an ovarian cystic mass, and laparoscopic excision may be the method of treatment.

**Key words:** Laparoscopy, lymphangioma, ovarian cyst, ovary

## INTRODUCTION

Lymphangiomas are rare tumors of the lymphatic system comprised of multiple cystic spaces lined by endothelium. They are generally benign tumors. Lymphangiomas can be classified as capillary, cystic, or cavernous. They contain serous or chylous fluid.<sup>[1]</sup> Lymphangiomas may arise anywhere in the body. In children, they were more commonly arising in the head, neck, and axilla. In adults, most often they are superficial cutaneous or intraabdominal.<sup>[2]</sup>

Lymphangioma of the ovary is rare and we have identified only few reports in literature survey (PubMed).<sup>[3-10]</sup> Typically, lymphangiomas are slow-growing tumors. They may vary in their clinical presentation and may remain asymptomatic for a long time. They are most often the incidental findings during abdominal or pelvic imaging studies or at surgery or autopsy.<sup>[8,9]</sup> Occasionally, they grow large and cause mass effect and compression of adjacent organs.<sup>[8,9]</sup> We present a case report of a cystic lymphangiomas arising from the

ovary in a 72-year-old post-menopausal woman that was successfully removed at laparoscopy.

## CASE REPORT

A post-menopausal 72-year-old woman was referred to a gynecologic laparoscopic surgeon for a consultation after an ultrasonography and computer tomography scan finding of a right ovarian complex cyst measuring 6.3 cm × 4.4 cm × 3.2 cm. Initially, the patient had lower abdominal pain, bloating, constipation, and urinary frequency. She is hypertensive and on regular anti-hypertensive drugs. Pelvic examination revealed mildly enlarged, non-tender, mobile right adnexa. The remainder of the physical examination yielded unremarkable findings. The serum CA 125 concentration was within normal limits at 9.7 U/mL.

She was planned for total laparoscopic hysterectomy with bilateral salpingo-oophorectomy. Exploratory laparoscopy was performed and a 10 mm trocar was placed in the

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umbilicus and three 5-mm secondary trocars were placed under direct visualization, suprapubically and in the left and right lateral position, respectively. Exploration revealed an enlarged right ovary with a cystic mass, with no excrescences or implantation on the peritoneum [Figure 1]. The uterus was small in size and a small amount of chylous milky ascites observed in pelvis. Peritoneal fluid was obtained for cytological analysis. The both-sided infundibulopelvic and tubo-ovarian ligaments were coagulated and cut and bilateral salpingo-oophorectomy was performed [Figures 2-4]. The adnexa were removed intact at colpotomy using an Endobag. Multiple peritoneal biopsies were taken from pelvic peritoneum, peritoneum overlying right, and left paracolic gutter and sub-diaphragmatic peritoneum [Figures 5 and 6]. On frozen section of the right ovarian, mass showed a gray and white multiple lobulated mass containing multiple cystic areas filled with milky fluid. The diagnosis was benign mesenchymal

mass with marked myxoid degeneration. The surgery was completed with assuring complete hemostasis and suturing of colpotomy laparoscopically [Figure 7]. Final histologic analysis confirmed the diagnosis of cystic lymphangioma of the right ovary.

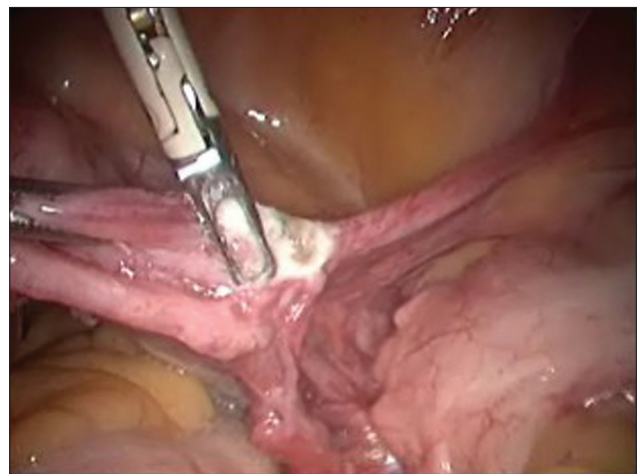
Cytologic analysis of pelvic washings was significant for mesothelial cells, histiocytes, and lymphocytes without evidence of malignancy. The post-operative course was unremarkable.

## DISCUSSION

Lymphangiomas are rare, usually benign lesions of the lymphatic system.<sup>[1]</sup> It is still uncertain whether they represent true neoplasms, hamartomas, or lymphangiectasis.<sup>[10]</sup> Their exact etiology and true incidence is unknown. In children, they more commonly arise in the head, neck, and axilla where the lymphatic sacs fail to communicate with the



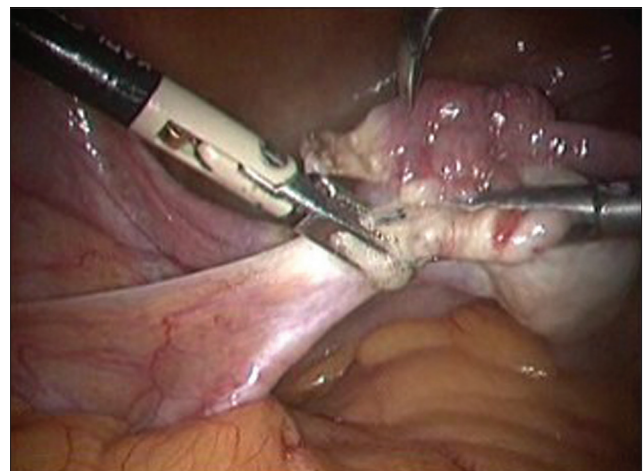
**Figure 1:** Enlarged right ovary without any excrescences or implantation on the peritoneum



**Figure 2:** Coagulation of right round ligament

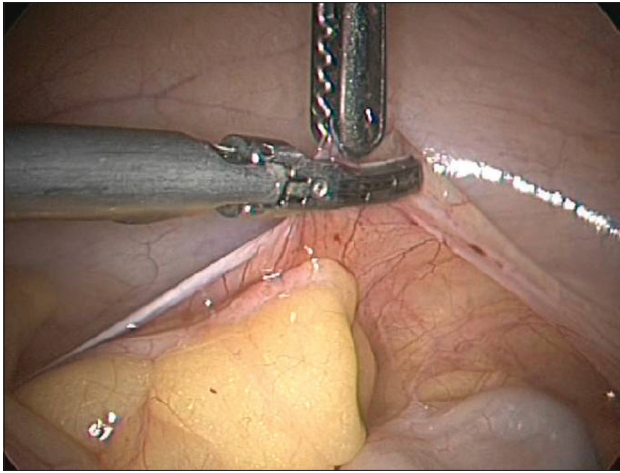


**Figure 3:** Coagulation of right infundibulo-pelvic ligament



**Figure 4:** Coagulation of left infundibulo-pelvic ligament

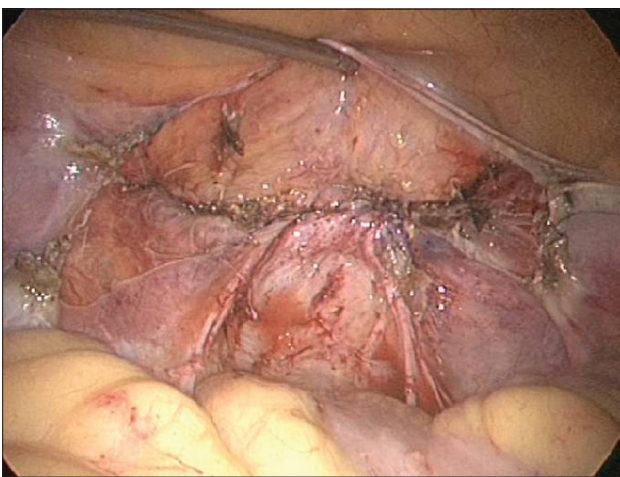




**Figure 5:** Peritoneal biopsy from right paracolic gutter



**Figure 6:** Peritoneal biopsy from left paracolic gutter



**Figure 7:** Pelvis after vault closure

draining lymphatic channels.<sup>[10,11]</sup> In adults, their etiology is less understood, some author suggest that its origin by proliferation of lymphoid nests after inflammation,

fibrosis, or genetic predisposition,<sup>[1,8,9,12]</sup> while, many believe it as an end result of mechanical pressure, trauma, degeneration of lymph nodes, and disorders of the lymphatic vasculature.<sup>[8,9,12-14]</sup> Lymphangiomas most often are superficial cutaneous or intraabdominal.<sup>[2]</sup>

Suspicion of malignancy in our case was high because of age (72 years), post-menopausal stat and complexity of ovarian mass on ultrasonography and computed tomography scan, though, her CA-125 was within normal range. The cause of the rare ovarian lymphangioma in our case is not known. There was no history of precipitating trauma, infection, or exposure to radiation, no previous surgery or adhesions. The lack of evidence of reactive process suggests a neoplastic cause.

The few case reports, malignant Lymphangiomas are available for review. Rice *et al.*,<sup>[15]</sup> reported a histologically benign lymphangioma that was successfully resected at laparotomy, presented with contralateral ovarian involvement, liver metastasis, and diffuse intraperitoneal dissemination 6 months following surgery. The autopsy confirmed malignancy.<sup>[15]</sup> Another case report by Aristizabal *et al.*,<sup>[7]</sup> described recurrence of a benign-appearing lymphangioma diffusely in the peritoneal cavity within 2 years of the open resection. These recurrences had the same benign appearance as the original lesion. Radiation therapy was required to control the disease.<sup>[7]</sup>

These reports suggest that the histologic appearance of ovarian lymphangiomas may not reliably predict their subsequent clinical behavior. They also emphasize the need for complete wide excision with clean margins and for prolonged follow-up for at least 2 years.<sup>[7]</sup> The patients should undergo a pelvic examination and gynecologic sonography every 3 months.

As with other adnexal masses, pelvic ultrasonography is the preferred imaging method. Computed tomography or magnetic resonance imaging may add important preoperative information about anatomical relationships with other structures, differentiate benign from malignant and chylous fluid from blood and pus.<sup>[6,9]</sup>

Laparotomy or laparoscopy both are acceptable routes of surgery for treatment. There are several reports in the literature of successful open resection of intraabdominal lymphangiomas, but only few such report describing the laparoscopic treatment of an ovarian lymphangioma.<sup>[4,5]</sup> The prognosis with laparoscopic treatment is usually excellent.<sup>[14,15]</sup> It has inherent advantages in the form of less intra-operative blood loss, early recovery, less morbidity,

and low complication rate compared with laparotomy. In our case, the choice of laparoscopic surgery enabled us to reach the correct diagnosis and offer the right treatment, adhering to proper oncologic principles, with minimal morbidity. With advances in equipment, instruments, techniques, and training, the endoscopic approach to gynecologic surgery is becoming more and more common, safe, and reliable procedure.

### CONCLUSION

In conclusion, the present case illustrates that lymphangiomas should be included in the differential diagnosis of ovarian cystic masses. Even though, they are being benign in nature majority of the time, wide excision with clear margin and regular follow up is mandatory. The laparoscopic excision is a safe and reliable approach for the treatment.

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