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Case Report

Rare Case of Ovarian Cystic Lymphangioma

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ABSTRACT Lymphangiomas are rare, generally benign tumors of the lymphatic system comprised of multiple cystic spaces lined with endothelium. Lymphangiomas may arise in any part of the body. Lymphangioma of the ovary is rare; we have identified only 13 reports in a 50-year literature survey (PubMed 1959–2009). Typically, lymphangiomas are slow-growing tumors that remain asymptomatic for a long time. They are most often found incidentally in abdominal or pelvic imaging studies or at surgery or autopsy. Wide excision of the lesion with microscopically clear margins is the best approach when feasible. A postmenopausal woman had a symptomatic pelvic mass. Imaging studies demonstrated a complex left ovarian cyst. Complete removal of a cystic lymphangioma was successfully performed at laparoscopy. Cystic lymphangiomas should be included in the differential diagnosis of an ovarian cystic mass, and laparoscopic excision may be the method of treatment. Journal of Minimally Invasive Gynecology (2010) 17, 97–99 © 2010 AAGL. All rights reserved.

Keywords: Lymphangioma; Ovary; Laparoscopy

Lymphangiomas are rare, generally benign tumors of the lymphatic system comprised of multiple cystic spaces lined by endothelium. They are classified as capillary, cystic, or cavernous and contain serous or chylous fluid [1]. Lymphangiomas may arise anywhere in the body. In younger children, they are preferentially located in the head, neck, and axilla. In adults, they most often are superficial cutaneous or intraabdominal [2].

Lymphangioma of the ovary is rare. We have identified only 13 reports in a 50-year literature survey (PubMed 1959–2009) [3–15]. Its clinical signs and symptoms are variable and may be misleading. Typically, lymphangiomas are slow-growing tumors that remain asymptomatic for a long time. They are most often found incidentally during abdominal or pelvic imaging studies or at surgery or autopsy [16,17]. Occasionally, they grow large enough to become symptomatic due to mass effect and compression of adjacent organs [16,17]. We present a case report of a cystic lymphangioma arising from the ovary in a postmenopausal woman that was successfully removed at laparoscopy.

Case Report

A postmenopausal 52-year-old woman was referred to a gynecologic laparoscopic surgeon for a consultation after an ultrasonographic finding of an enlarged left ovary measuring 5.6 × 4.1 × 8.2 cm. Initially, the patient had lower abdominal pain, hematuria, and urinary frequency. She denied nausea, vomiting or diarrhea, weight loss, constipation, or bloody stools. She had mitral valve prolapse and a known uterine myoma. Her family history was significant for an aunt who had ovarian cancer in her 70s. Pelvic examination revealed a mildly enlarged, nontender, mobile left adnexa. The remainder of the physical examination yielded unremarkable findings other than a systolic murmur. The patient was sent by her internist for sonographic evaluation and subsequently was referred for consultation.

Laboratory results confirmed microscopic hematuria. The CA 125 concentration was within normal limits at 7.1 U/mL. Renal ultrasonography revealed normal kidneys. Magnetic resonance imaging of the abdomen and pelvis confirmed the presence of an enlarged myomatous uterus measuring 8.0 × 7.3 cm. In addition, a cystic mass was noted in the left lower quadrant that measured 6.1 × 6.7 cm (Fig.1).

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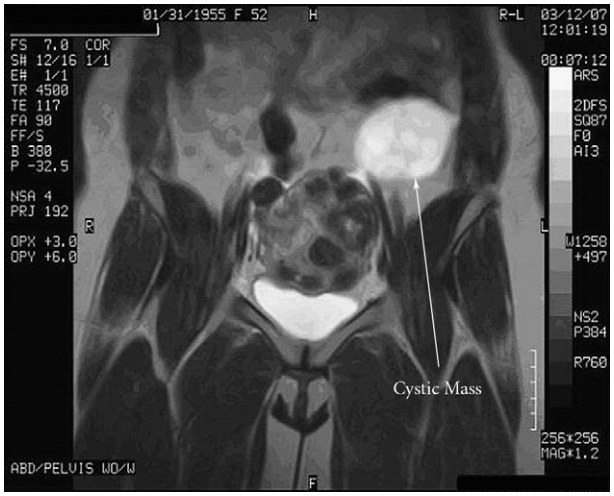

Fig. 1. Pelvic magnetic resonance image demonstrates left adnexal mass measuring 6.1 × 6.7 cm.

Exploratory laparoscopy was performed. A 10-mm trocar was placed in the umbilicus, and under direct visualization, two 5-mm trocars were placed suprapubically and in the left lateral position, respectively. Exploration revealed an enlarged left ovary with a cystic mass, with no excrescences or implantation on the peritoneum. In addition, an enlarged myomatous uterus, a small amount of chylous milky ascites, and perihepatic adhesions were observed (Fig. 2). Peritoneal fluid was obtained for cytologic analysis. The left infundibulopelvic and tubo-ovarian ligaments were clamped, cut, and tied using laparoscopic suture technique, and left-sided salpingo-oophorectomy was performed. The adnexa was removed intact at colpotomy using an Endobag. Frozen section of the left ovarian mass showed a tan, gray, and white slightly lobulated mass measuring 6.5 × 4.2 × 3.9 cm. Sectioning revealed a tan-pink to tan-gray slightly spongy cut surface. Multiple cystic areas measuring from 0.5 cm to 2.1 cm in greatest dimension were identified and were filled with tan-white slightly milky or opalescent fluid. On the external surface there was a small 1.5-cm well-circumscribed rubbery nodule. Sectioning of this nodule revealed a tan-white whorled rubbery cut surface. The diagnosis was benign mesenchymal mass with marked myxoid degeneration and a small leiomyoma. The surgery was completed with a right-sided salpingo-oophorectomy. The colpotomy was sutured intracorporally. Final histologic analysis confirmed by immunohistochemistry, established the diagnosis of cystic lymphangioma originating from the left ovary (Fig. 3). Cytologic analysis of pelvic washings was significant for mesothelial cells, histiocytes, and lymphocytes without evidence of malignancy. The postoperative course was unremarkable.

Discussion

Lymphangiomas are rare, usually benign lesions of the lymphatic system [1]. It is still uncertain whether they represent true neoplasms, hamartomas, or lymphangiectasis [18].

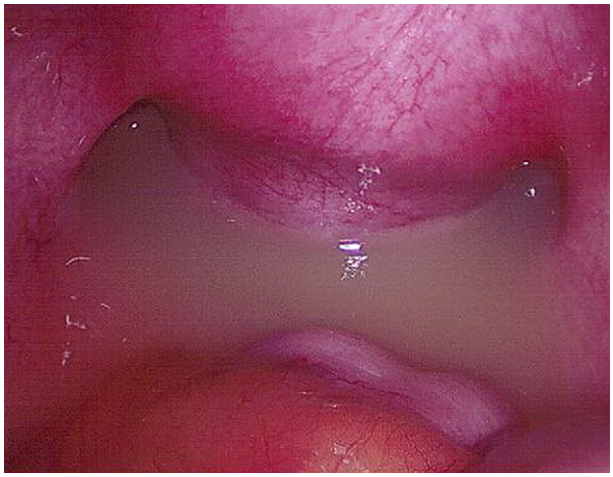

Fig. 2. Laparoscopic image of chylous milky ascites.

Their true incidence is unknown, and their exact etiology is yet to be determined. In children, they generally arise from sequestered lymphatic sacs that fail to communicate with the draining lymphatic channels [18,19]. In adults, their etiology is less clear; most authors favor proliferation of congenital or acquired lymphoid nests after inflammation, fibrotic processes, or genetic predisposition [1,16,17,20]. Other proposed etiologic factors include mechanical pressure, trauma,

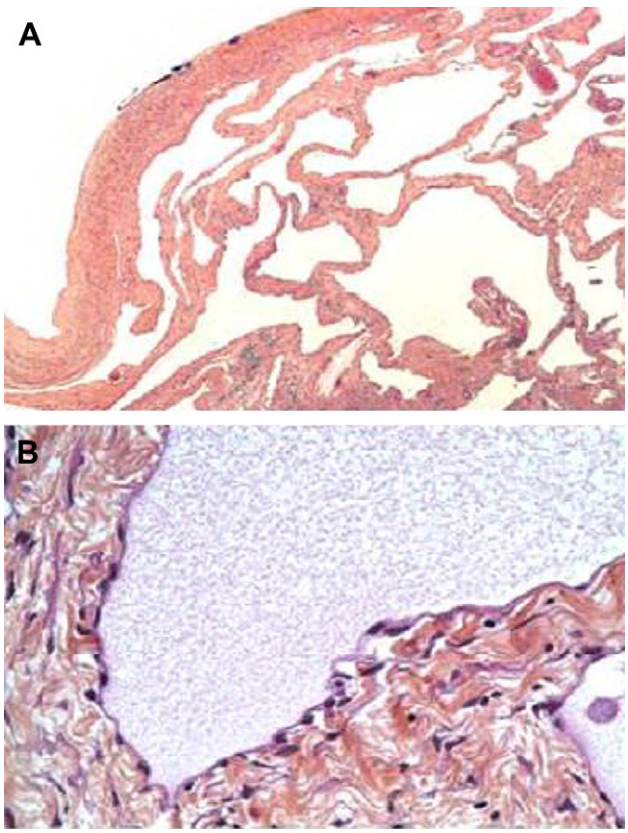

Fig. 3. A, Photomicrograph of ovarian cystic lymphangioma (hematoxylin-eosin, magnification × 20). B, High-power photomicrograph (hematoxylin-eosin, magnification × 100).

degeneration of lymph nodes, and disorders of the endothelial lymphatic vasculature [16,17,20–22].

The cause of the rare ovarian lymphangioma in our case is not clear. There was no known precipitating trauma, infection, or exposure to radiation, no previous surgery, and no adhesions to suggest a reactive process. The lack of evidence of reactivity suggests a neoplastic cause.

There are rare reports in the literature of malignant lymphangiomas. Rice et al [23] reported a histologically benign appearing lymphangioma that was successfully resected at laparotomy, only to have the patient return 6 months later with contralateral ovarian involvement, liver metastasis, and diffuse intraperitoneal dissemination. The patient died, and postmortem examination showed swollen malignant-appearing endothelial cells with hyperchromatic nuclei, hemorrhage, and necrosis [23]. Another report by Aristizabal et al [12] described a benign-appearing lymphangioma that recurred diffusely within the peritoneal cavity on 2 occasions within 2 years of the original open resection. These recurrences had the same benign appearance as the original lesion. Radiation therapy was required to control the disease [12]. 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 [9,12]. Our patient was instructed to 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 and differentiate chylous fluid from blood and pus [7,17].

Laparotomy or laparoscopy are both acceptable methods of treatment. There are several reports in the literature of successful laparoscopic resection of intraabdominal lymphangiomas but only 1 such report describing the laparoscopic treatment of an ovarian lymphangioma [6]. The prognosis with laparoscopic treatment is usually excellent [22,24,25]. 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.

In conclusion, the present case illustrates that lymphangiomas should be included in the differential diagnosis of ovarian cystic masses and that laparoscopic excision is a reasonable mode of treatment.

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