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Case Report

A Diagnostic Dilemma between Ovarian Cyst and Ascites or Lymphangioma of Small Intestine; A Case Report and Literature Review

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Abstract

Lymphangioma or lymphatic spaces proliferation as a benign tumor is so rare when presented in the small-bowel mesentery (account for less than 1% of all lymphangiomas).

A 24 year old woman with an abdominal huge cyst that presented at the time of diagnosis with abnormal unwanted Weight Gain about 4 kg in one month and abdominal distention. The ultra-sonographic diagnosis was a large mass of adnexa uteri, but intraoperative findings was a huge, yellowish cystic mass that was originated from the small bowel mesentery, containing a milky white fluid with surgical spill, it was resected completely without any small bowel injury and the final histological diagnosis was lymphangioma of small mesentery.

Our case demonstrates the low accuracy of imaging before surgery for this diagnosis and requirement of surgery for definite diagnosis in addition to possibility of mass excision without any need to small bowel resection.

Keywords: Lymphangioma, Intestine, Small, Adnexa Uteri

Introduction

Lymphangioma is not a true lymphatic tumor, it is a benign condition due to lymphatic space enlargement and consider as a congenital lymphatic vessels malformation [1]. The common hypothesis is "blind sac" that means dilation and proliferation of lymphatic vessel is due to lack of lymphatic connection [2]. Clinical presentation in most of the cases is abdominal pain, nausea, diarrhea or constipation. The soft and compressible mass is the common finding on physical examination [2]. Lymphangioma is an important diagnosis due to its potency for heroic complications such as bowel obstruction, volvulus or perforation [3].

Imaging modality has valuable role for diagnosing these cystic lesions, especially computed tomography (CT) scan and magnetic resonance imaging (MRI) providing more detailed information about lesion size and location versus ultrasonography [4].

Although Conservative management could be considered in selected patient due to 10% chance of spontaneous regression of lymphangioma in asymptomatic cases, resection is recommended to avoid complications related to cyst growth. Based on previous study, the standard treatment for lymphangioma is surgical excision [1-4].

Case Report

A 24 year old nuli-gravida female referred to our center with unexplained distention of abdomen for 40 days ago. On physical exam she was pale with stable vital sign, abdomen was distending but had no tenderness. Laboratory tests were normal and only the Hemoglobin level was 9 mg/dl.

Ultrasound examination of the abdominal cavity detected a 120 x 200 mm multilocular fluid-filled cyst from upper abdominal

region downward to the pelvis which suggested an adnexal huge mass with ascites. Further diagnostic evaluation by the use of CT scan had performed for her; a 200 x 100 mm fluid-structure lesion in the abdominal region, well circumferential external outline, with adipose tissue compartments at the pelvis up to upper abdomen was detected. The lesion had compressed the adjacent bowel and showed air-fluid levels which suggested a chylous mesenteric cyst in regard (Figure1).

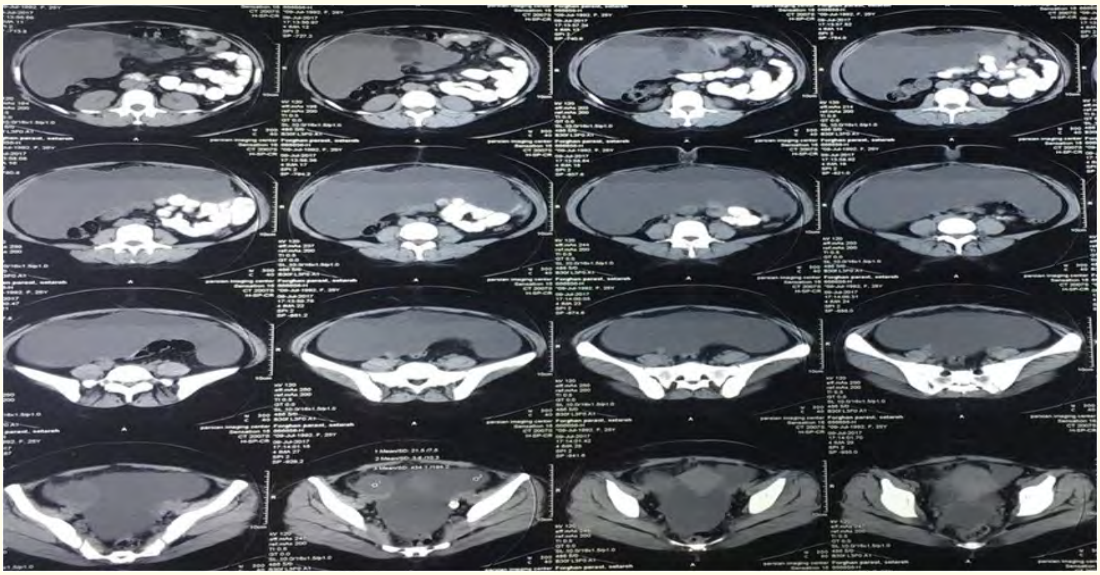

Figure 1: Computed tomography scan reveals A huge multilocular fluid-filled cyst from upper abdominal region downward to the pelvis.

Laparotomy was planned for her. Intra operation, a huge and yellowish pink mesenteric mass was found adjacent to small bowel (in the jejunal region of mesentery), cystic and fulfilled with milky white fluid in about 3000 mL representing lymphatic fluid (Figure 2). Histopathological examination showed cystic walls with various sizes of small lymphatic spaces lined by a flat endothelium which containing small lymphoid cells suggested the diagnosis of lymphangioma mesentery (Figure 3).

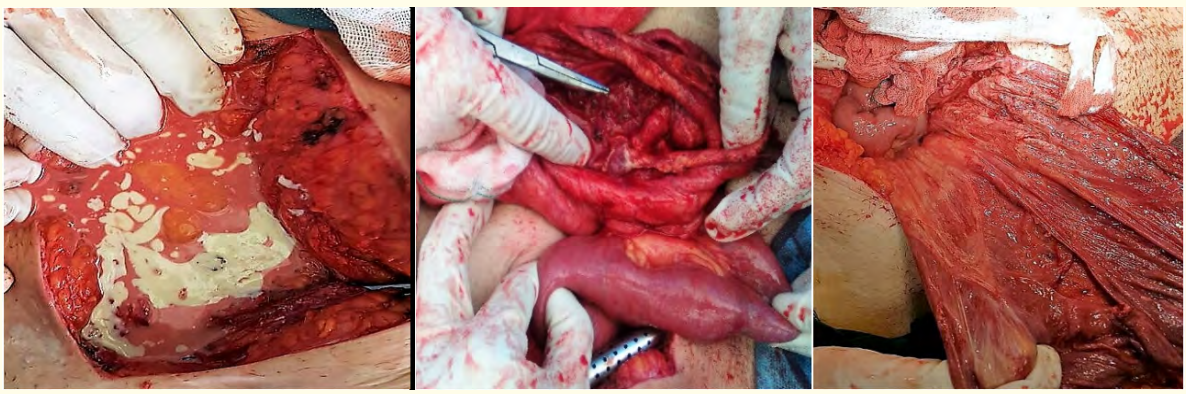

Figure 2: A Cystic lesion fulfilled with milky white fluid in jejunal region.

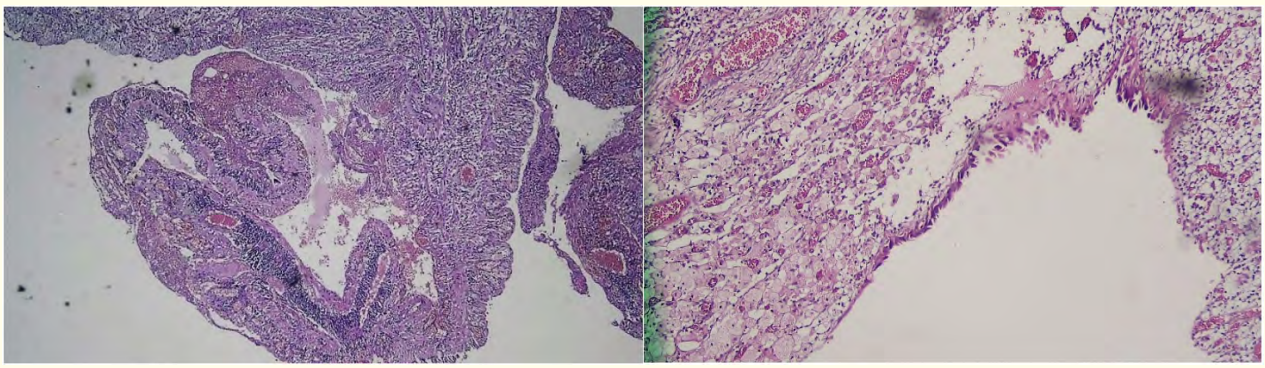

Figure 3: Pathology (H&E 100×-400×) shows dilated lymphatic spaces in various size in correlation with smooth-muscle cells.

At 11 months follow up after operation the patient did not manifest any postoperative complications. Written consent was obtained from the patient for reporting the case.

Discussion

Lymphangioma mesenteri is a cystic tumor which is extremely rare especially in small intestine and either in adult (less than 1% of all lymphangiomas) [1]. Lymphangioma is mostly manifested in head and neck and child boy [1,3]. This case is third case of mesenteric small bowel lymphangioma that presented with retroperitoneal mass to date [3,5].

Studies revealed different cause for this lymphatic vessel abnormality formation such as inflammation, abdominal trauma, abdominal surgery, radiation or lymphatic obstruction [1].

Abdominal distension, a palpable abdominal mass as present case or acute intestinal obstruction and volvulus are the most common manifestation of Intra-abdominal lymphangioma so could presented with acute abdominal pain [1,3,6].

Severe anemia and abdominal mass can be a rare manifestation for lymphangioma by lymphatic vessels bleeding explanation, in this case anemia was one of her accidental laboratories finding [6].

The differential diagnosis of a fluid-filled cystic lesion on the imaging approach are pseudocysts, dermoid cysts, enteric duplication cysts, lymphocele, mesothelioma, lipoma and various types of sarcoma which could be distinguished from small intestine lymphangioma [4]. Due to the rarity and nonspecific symptom and imaging finding of this disease, definite diagnosis is difficult before opening the abdomen. In the other hand intermittent or persistent pain of the patient could be due to lymphangiomas critical complication; intestinal obstruction or perforation even mesenteric ischemia due to involvement of the main branch of the mesenteric arteries which require emergent surgery [3,4].

Studies support early surgery as the optimal therapeutic procedure, which reduce the possibility of complications and ruled out the more important differential diagnosis.

Based on previous study, the standard treatment is surgical excision of the tumor completely and with the surrounding organs that may be potentially invaded, due to previous study recurrence rate may increase from 10% up to 100% depending on adequacy of surgery. This extended surgery has potency of important complication such as infection, fistula and hemorrhage [1-3].

So, the most efficient treatment method is excision of the lesion completely even by expert laparoscopist but not easily in urgent cases [4]. There are case reports of sclerotherapeutic agents used into the cyst [3].

Although partial resection of small intestine is necessary for exclude malignancy in previous study [7] but our patient had full recovery whit no recurrence by close follow up after 11 months in spite of local resection of the mass without segmental resection of intestine. In this case this limited method was associated with a good prognosis.

The milky white fluid cyst supported lymphoid cells of lymphangioma, must be differentiated from lymphangiomyoma and benign multicyclic mesothelioma; the first one shows proliferation of smooth-muscle cell around the lymphatic spaces with pericytic differentiation in microscopic view and HMB-45 immunostaining positivity but the second one consists of mesothelial cells and Calretinin is its immunohistochemistry marker [1].

The characteristic histologic view of cystic lymphangioma is dilated lymphatic spaces in various size in correlation with smooth-muscle cells. The immunohistochemical marker of lymphatic endothelial marker is D2-40. In the other hand lymphoid follicles and lymphoid infiltrates in the stroma supported the diagnosis as what had been showed in present patient [1,8,9].

There are three type for lymphangioma: Capillary (or small size, the commonest in face which could cause deformity), Cavernous (cyst of different sized cavities that connected with normal lymphatic vessels) and Cystic one which is the least common type which built of large cysts and are common in abdomen similar to this case histology [3].

Conclusion

In the differential diagnosis of a cystic abdominal mass even in adult, lymphangioma must be keep in mind which is rare and with difficult preoperative accurate diagnosis. Lymphangioma could cause fatal complications such as volvulus. Our case demonstrated the possibility of resection of lymphangioma without small bowel mesentery injury with no increase in recurrence after near one year of follow up.

Conflict of Interest

None declared.

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