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Medicine Science International Medical Journal

CASE REPORT

Medicine Science 2018;7(2):418-20

Huge colo-mezenteric cystic lympgangioma: A case report

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Received 05 August 2017; Accepted 30 October 2017
Available online 15.01.2018 with doi: 10.5455/medscience.2017.06.8726

Abstract

A 29-year old female patient was presented to the our department with mild intermittent lower abdominal pain. Otherwise she was healthy and she did not report previous exposure to toxic chemical agents. After physical examination, laboratory investigation and imaging study there was abdominal mass. Surgery was done the resected mass sent for histological examination that confirm the diagnosis lymphangioma. Lymphangiomas are congenital malformations of the lymphatic system that account for about 5% of all benign tumors in infants and children. The most common sites are the neck and axilla, which account for 95% of cases. Lymphangiomas in the peritoneal cavity are extremely rare, particularly in adults. Most commonly arising from the mesentery followed by the omentum, mesocolon and retroperitoneum. Preoperative diagnosis is often difficult due to the frequent silent clinical course. Radiological investigations are a useful diagnostic tool, but definitive diagnosis is confirmed by histopathology after a complete surgical resection.Keywords: Granuloma annulare, non-annular, female

Keywords: Cyst, lymphangioma, congenital, lymphangiomas, retroperitonium

Introduction

A 29-year old female patient presented due to mild intermittent lower abdominal pain. Otherwise she was healthy, fter physical examination, laboratoty investigation and imaging study there were abdominal mass. Surgery was done the resected mass sent for histological examination which confirm the diagnosis lymphangioma. The most common sites are the neck and axilla, which account for 95% of cases. Lymphangiomas in the peritoneal cavity are extremely rare, particularly in adults. Most commonly arising from the mesentery followed by the omentum, mesocolon and retroperitoneum. Radiological investigations are a useful diagnostic tool, but definitive diagnosis is confirmed by histopathology after a complete surgical resection.

Case Report

A 29-year old female patient, gravida 4, para 4(G4 P4)referred to us from a gynecologist clinic due to mild intermittent lower abdominal pain for 2-week ago. She has been experiencing menses irregularity since one year.Otherwise she was healthy and she did not report previous exposure to toxic chemical agents. The blood creatinine level was 0.8 mg per deciliter (normal range, 0.6 to 1.5 mg per deciliter (53 to 133 µmol per liter), blood urea nitrogen level was 29 mg per deciliter (normal range, 8 to 25 mg per deciliter) and fasting blood glucose level 85 mg/dl. Thyroid stimulating hormone (T.S.H) prolactine, alkaline phosphatase(ALP), AST and ALT were normal see table [1].

Table 1. Hormonal and liver function tests

Test	Result	Normal range
T.S.H	2.5	(0.5-5)uI/ml
Prolactin	7.2	(1.9-25.9)ng/ml
Testosterone	0.3	(0.1-1.2)ng/ml
ALP	118	(0-258)u/ml
AST	30	(0-42)u/ml
ALT	28	(0-42)u/ml

Blood investigations including complete blood count (CBC), were within normal except for moderate microcytic hypochromic anemia see table [2].

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Table 1. Complete blood count(CBC).

Test	Result	Normal range
WBC	4.9 k/ul	4.1-10
Lymph	2	0.6-4.1
MID	0.8	0.0-1.8
Gran	4.8	2-7.8
RBC	4.12 M/ml	4.2-6.3
HGB	9.9 g/dl	11-13 g/dl
HCT	29%	37-51%
MCV	67.9fl	80-97
MCH	21.3pg/u	26-37
MCHC	31.3 g/dl	31-36
PLT	235 k/ul	140-440

On examination, the temperature was 37.6°C, pulse 73 beats per minute, blood pressure 110/85 mm Hg, respiratory rate 16 breaths per minute, and the oxygen saturation 95%. She was stable and well nourished.Breath sounds were normal vesicular of both lungs on auscultation. The abdomen was soft and flat except left sided abdominal fullness but no palpable masses or tenderness.; the remainder of the examination was normal.

Imaging study including ultrasonography(US) for abdomen and pelvis showed a multi-loculated cystic lesion seen in the left hypochonderium extending downward to Left lower abdomen. There were no organ enlargement or lymph nodes or any other pathology.

CT of the abdomen and pelvis, performed with the administration of contrast materialrevealed multiple cystic changes seen on the left side of the abdomen primary affecting the mesentery from left hypochondrial area to left pelvic area, with closed relation to descending colon and no line of cleavage figure [1].

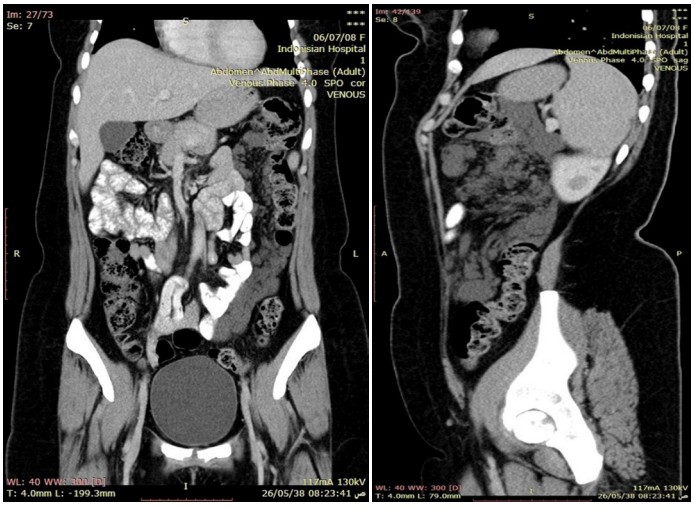

Figure 1. C T of the abdomen and pelvis shows Hypo attenuated multi-cystic lesion arising from the splenic flexure at the level of T12 till the pelvic inlet

Tumor markers includingcarcinoembryonic antigen (CEA and CA125), CA125 level was 35 u/ml[normal range 0-37 u/ml].

Ultrasound guided fine needle aspiration (FNA) was done which revealed 85% lymphocytes.The patient was counseled for surgical decision. A diagnostic test result was received, and the decision was surgical excision as treatment . Surgical consent, prophylactic antibiotic andI.V fluid started andbowel preparation were done.

During the operation, a multicystic tumor that appeared to engulf the colon from the distal transverse colon down to the sigmoid was noted (30cm x5cmx8cm). The distal part was well encapsulated and the proximal part was contained within the leaflets of the grossly distorted mesentery(variable sized cysts ranging from few mm to 6cm) .The transition to normal colon mesentery proximally and mesosigmoid distally was clear suggesting a benign etiology figure [2].

There was no evidence of either carcinomatosis or suspicious metastatic tumor deposits. The fluctuant mass and the involved colon were completely resected followed by a primary colo-colic anastomosis.After an uneventful recovery, the patient was discharged on postoperative day five. Outpatient clinic follow up weekly for two weeks then monthly for three months shows the patient is doing well.

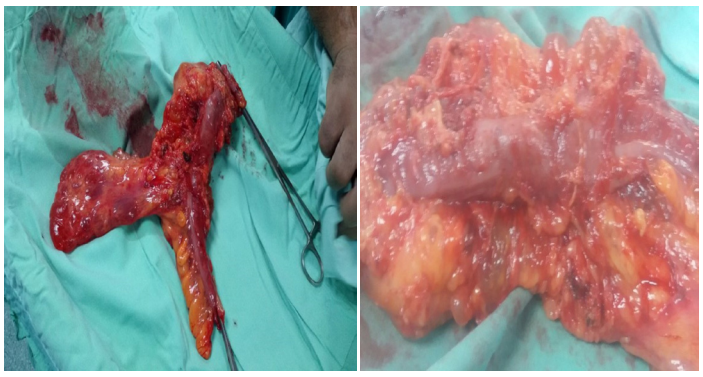

Figure 2. The resected cystic mass associated with attached colonic segment

Differential Diagnosis

In this 29-year old female patient referred to us from a gynecologist clinic due to mild intermittent lower abdominal pain for 2-weeks ago. She has been experiencing menses irregularity since one year. Otherwise she was healthy and she did not report previous exposure to toxic chemical agents. Blood investigations including KFT, CBC, LFTs, were within normal except for moderate microcytic hypochromic anemia.

On examination, the temperature was 36.6°C, the pulse 76 beats per minute, the blood pressure 110/73 mm Hg, the respiratory rate 16 breaths per minute, and the oxygen saturation 95. She was stable and well nourished, Breath sounds were normal vesicular of both lungs on auscultation. The abdomen was soft and flat except left sided abdominal fullness but no palpable masses or tenderness.; the remainder of the examination was normal. The platelet count, red-cell indexes, as were blood levels of glucose, alkaline phosphatase, AST and ALT, the results of other laboratory test were normal. The patient was initially treated for menstrual irregularity and polycystic ovary syndrome, but hormone and US

results deny this diagnosis [1,2]. The impression of the resected mass that it may be benign multicystic peritoneal mesitheloma which is common among child bearing age females in this age group but CT andhistopathologic study reveal it is Mesenteric lymphangioma [3-5].

Lymphangioma is a benign tumor that is rarely seen; it presents in fewer than 1 in 20,000 to 1 in 250,000 hospitalization. Lymphangioma may occur at any age, although it typically develops during childhood[6]. Lymphangioma usually develops in the neck (75% of cases), but it can be seen in the axilla (22%), or rarely in the mediastinum (4–5%) [7].

The etiology of mesenteric lymphangioma is considered to be congenital, with abnormal embryonic development of the lymphatic system causing sequestration of lymphatic tissue [6]. However, other possible causes have been suggested such as: abdominal trauma, lymphatic obstruction, inflammatory process, surgery.Although mesenteric cystic lymphangioma is a benign tumor, it often has life-threatening complications such as secondary infection, rupture with hemorrhage, and volvulus or intestinal obstruction. Clinical manifestations are variable, without characteristic signs and symptoms that can be attributed solely to the mesenteric lymphangioma [8,9]. They may present as nonspecific symptoms such as abdominal pain, vomiting, and constipation. As these symptoms are common to many diseases, creating a wide differential diagnosis, the specific diagnosis of mesenteric lymphangioma is challenging. Assessment by diagnostic imaging, including ultrasonography, CT, and MRI [10].Percutaneous biopsy is not recommended because the diagnostic value is likely to be low owing to the low cellularity index of these tumors. However these studies help to determine, if the tumor is cystic, its size and location, but they are insufficient to establish an accurate preoperative diagnosis. The definitive diagnosis of lymphangioma is based on histopathology and immunochemistry [11].

Discussion

Examination of a core-biopsy specimen of the mass revealed a benign vascular proliferation composed of dilated space filled partially by mature lymphocytes and lined by attenuated thin layer of endothelial cells, surrounded by 1-5 layers of smooth muscle cells figure [3].

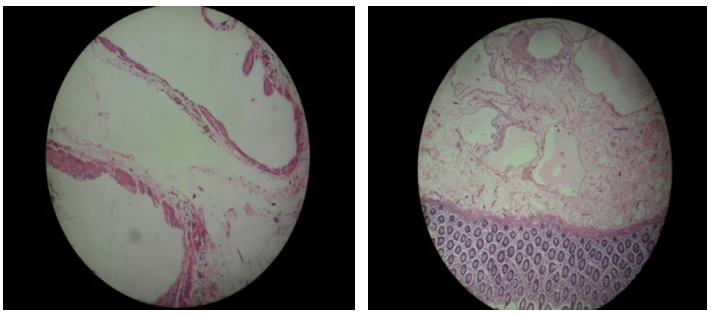

Figure 3. The resected cystic mass associated with attached colonic segment

Mesenteric lymphangioma in adult is a rare disease.It may present as an abdominal swelling or acute intestinal obstruction. Preoperative diagnostic tools are ultrasound and abdomen CT or MR. Complete surgical resection is the ideal modality of treatment for mesenteric lymphangioma [12].

The definitive diagnosis of lymphangioma is based on histopathology and immunochemistry. Few methods are available for the treatment of mesenteric lymphangioma.A complete surgical excision of the mass, that is the treatment of choice for cystic lymphangioma, even if asymptomatic. Drainage has been suggested as a modality of treatment in high – risk patients but is often unsuccessful because of recurrence and for the risk of perforation. OK432, a sclerotherapeutic agent, is the only medication known to be effective may be useful in cases with difficult complete resections [12-14]. There some trials onlaparoscopic surgery but it did not prove to have better outcome. The prognosis after adequate excision of the cystic tumors of the mesentery is considered to be excellent [12].

Competing interests

The authors declare that they have no competing interest.

Financial Disclosure

The financial support for this study was provided by the investigators themselves.

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