



Rupture of a giant jejunal mesenteric cystic lymphangioma misdiagnosed as ovarian torsion: A case report

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Abstract

BACKGROUND

Cystic lymphangioma is a rare benign tumor that affects the lymphatic system. Mesenteric lymphangiomas in the small bowel are extremely uncommon.

CASE SUMMARY

We present a 21-year-old female patient who complained of abdominal pain. The diagnosis of ovarian torsion was suspected after abdominopelvic unenhanced computed tomography and ultrasound revealed a large cyst in contact with the bladder, ovary, and uterus. The patient underwent emergency laparotomy performed by gynecologists, but it was discovered that the cystic tumor originated from the jejunum. Gastrointestinal surgeons were then called in to perform a cystectomy. Pathological examination confirmed the diagnosis of cystic lymphangioma of the mesentery. The patient had an uneventful postoperative recovery.

CONCLUSION

Mesenteric lymphangiomas can cause abdominal pain, and imaging techniques can help determine their characteristics, location, and size. Complete surgical excision and pathological examination are considered the standard treatment and diagnostic method.

Key Words: Rupture; Jejunum; Mesenteric cystic lymphangioma; Ovarian torsion; Surgical excision; Pathological examination; Case report

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Core Tip: Cystic lymphangioma is a benign malformation tumor of the lymphatic system. Mesenteric lymphangiomas in the small bowel are rare.

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INTRODUCTION

Lymphangiomas are benign tumors which may evolve from congenital malformations of lymphatic vessels[1]. Lymphangiomas are rare cystic tumors which are usually located in the axilla and neck of children[2]. Mesenteric lymphangiomas of the small bowel in adults are extremely rare and are fewer than 1% of all lymphangiomas[3]. In this report, we present a case wherein the misdiagnosis of ovarian torsion occurred due to the rupture of a sizable jejunal mesenteric cystic lymphangioma.

CASE PRESENTATION

Chief complaints

A 21-year-old woman suffered abdominal pain for one day.

History of present illness

The patient suffered abdominal pain for one day.

History of past illness

She had no history of kidney disease, coronary heart disease, hypertension or diabetes.

Personal and family history

No relevant personal or family history.

Physical examination

Tenderness was found around the umbilicus.

Laboratory examinations

Most laboratory test results, including serum amylase, glutamic-pyruvic transaminase, and creatinine levels, were within normal limits. However, serum C-reactive protein level was significantly elevated (90 mg/L) (Table 1).

Imaging examinations

Based on ultrasound (US) (Figure 1A) and abdominopelvic unenhanced computed tomography (CT) results (Figure 1B), the diagnosis of ovarian torsion was suspected. The imaging studies revealed a large cyst (14 cm × 12 cm) in contact with the bladder, ovary, and uterus.

FINAL DIAGNOSIS

The final diagnosis was cystic lymphangioma of the mesentery.

TREATMENT

Emergency laparotomy was performed by gynecologists, but during the procedure, it was discovered that the cystic tumor originated from the jejunal mesentery (Figure 2A). A small rupture in the cyst led to the leakage of chylous fluid. Gastrointestinal surgeons were then called in to perform a cystectomy (Figure 2B-D). Pathological examination confirmed the diagnosis of cystic lymphangioma of the mesentery (Figure 3). The patient's postoperative recovery was uneventful (Figure 4).

OUTCOME AND FOLLOW-UP

The patient's postoperative recovery was uneventful.

Table 1 Laboratory results before the patient underwent surgery

	Laboratory result	Normal
WBC ($\times 10^9/L$)	7.9	3.4-9.5
EO ($\times 10^9/L$)	0.05	0.4-8.0
HB (g/L)	129	115-150
Platelet ($\times 10^9/L$)	351	125-350
PT (s)	11.6	9.7-13.5
Dimer (mg/L)	0.45	0.00-0.55
ALT (U/L)	13	9-50
TB ($\mu\text{mol/L}$)	14	3-20
Cr ($\mu\text{mol/L}$)	43	40-80
CA199 (kU/L)	16.8	0-35
CEA ($\mu\text{g/L}$)	0.3	0-5
CRP (mg/L)	90	0-10
Amylase (U/L)	91	35-135
Lipase (U/L)	84	0-190
Glucose (mmol/L)	5.6	3.0-6.1
HbsAg	Negative	Negative
HIV Ab	Positive	Negative
Sp Ab	Negative	Negative
ANA	Negative	Negative
HC Ab	Negative	Negative

ALT: Alanine aminotransferase; ANA: Anti-nuclear antibodies; CEA: Carcinoembryonic antigen; CA199: Carbohydrate antigen 199; Cr: Creatinine; CRP: C-reactive protein; EO: Eosinophil; HB: Hemoglobin; HbsAg: Hepatitis B surface antigen; HC Ab: Hepatitis C antibody; HIV Ab: Human immunodeficiency virus antibody; PT: Prothrombin time; Sp Ab: Syphilis antibody; TB: Total bilirubin; WBC: White blood cell.

DISCUSSION

Mesenteric cysts have a reported incidence ranging from one per 100000 to one per 250000 of in-patients[4]. Due to the lack of lymphatics, cystic lymphangiomas cannot affect the central nervous system[5]. The incidence of mesenteric lymphangiomas is approximately 1/20000 in children and 1/100000 in adults[6]. The cause of mesenteric lymphangiomas may be attributed to congenital abnormalities of the lymphatic system which induces sequestration of lymphatic tissues during development of the embryo[7]. However, other causes such as abdominal trauma, inflammation, lymphatic obstruction, and radiation therapy should be considered[8]. Mesenteric lymphangiomas are normally asymptomatic until they become large. The main symptoms include an abdominal mass and abdominal pain[9]. Histopathology of the surgical specimen may identify a unilocular or multilocular cyst containing serous or viscous fluid with cholesterol crystals, chylomicrons, and triglycerides encircled by a single layer of flattened immunoreactive mesothelial cells with cytokeratins and a fibrous wall with lymphocytes[10,11].

In this case report, the patient was asymptomatic prior to rupture of the cyst but experienced abdominal pain localized around the umbilicus. It is worth noting that abdominal pain can be associated with various other medical conditions, making it difficult to establish the accurate diagnosis based on symptoms. Moreover, no definitive blood tests are available to confirm the diagnosis. In this particular case, both unenhanced CT and US imaging revealed a large cyst in proximity to the bladder, ovary, and uterus. Unfortunately, the radiologist, sonographer, and gynecologist misdiagnosed the patient's condition as ovarian torsion. Consequently, it became evident that complete surgical excision of the cyst, followed by pathological examination, was necessary to achieve a definitive diagnosis and provide appropriate treatment.

Surgical excision can prevent potential malignant transformation, in addition to complications and recurrences[12]. Vigilance is imperative during the follow-up period to promptly detect any recurrence of mesenteric cystic lymphangioma.

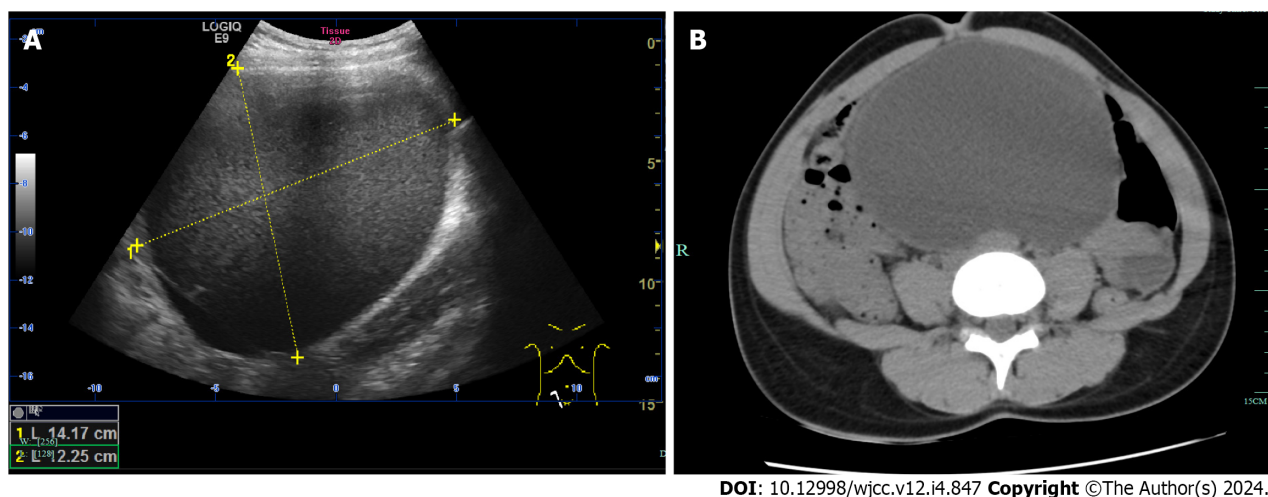


Figure 1 Imaging examinations before laparotomy. A: Ultrasound image of the patient; B: Abdominopelvic unenhanced computed tomography scan of the patient.

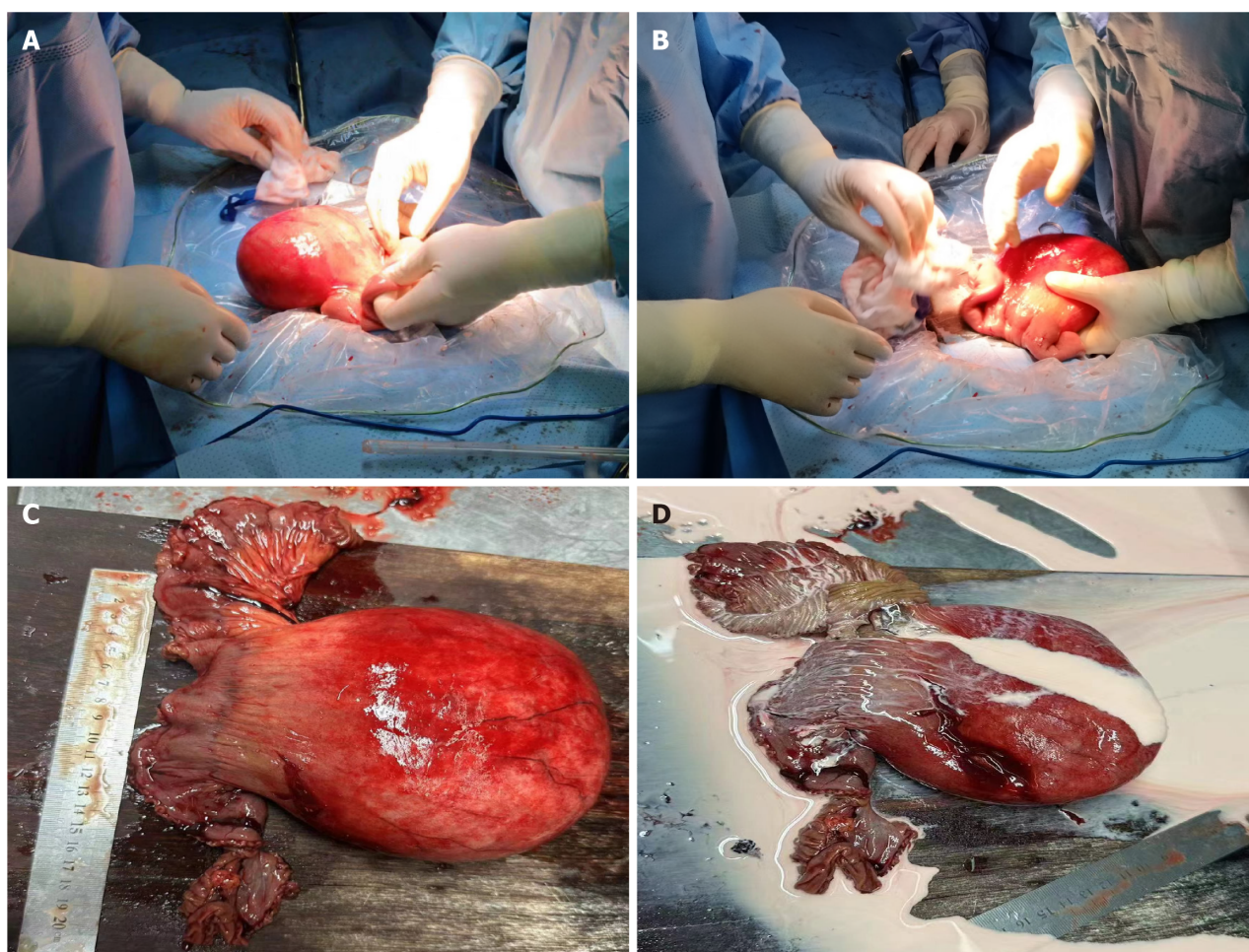
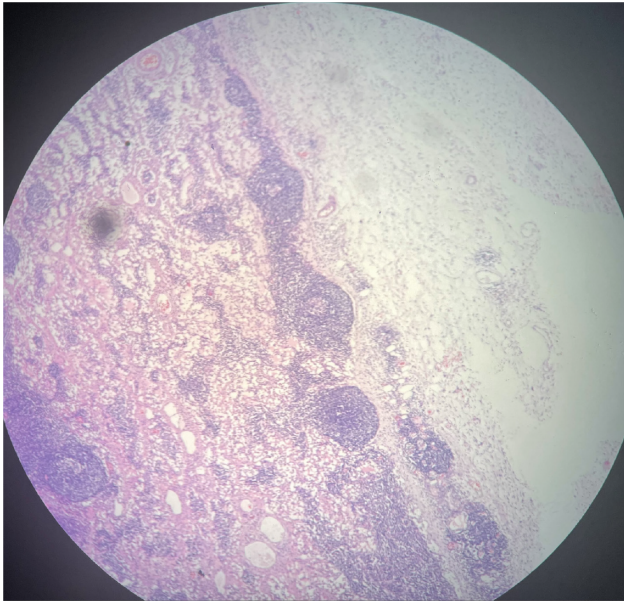


Figure 2 The patient underwent emergency laparotomy and cystectomy. A: A huge cystic tumor had grown from the jejunum; B: Gastrointestinal surgeons performed a cystectomy; C: The jejunal mesenteric cyst was removed; D: Chylous fluid flowed from the jejunal mesenteric cyst.

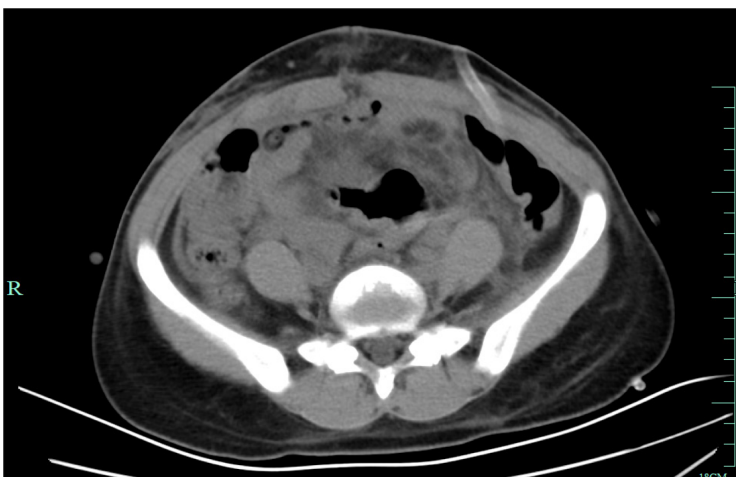
CONCLUSION

Mesenteric lymphangiomas can cause abdominal pain, and imaging techniques can help determine their characteristics, location, and size. Complete surgical excision and pathological examination are considered the standard treatment and diagnostic method.



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Figure 3 Pathological examination confirmed the diagnosis of cystic lymphangioma of the mesentery.



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Figure 4 Following the cystectomy, CT showed that the patient's postoperative recovery was uneventful.

FOOTNOTES

Author contributions: Xu J and Lv TF conceptualized and designed the case presentation; Xu J performed the data collection, analyzed and interpreted the data, drafted and critically revised the manuscript; All authors read and approved the final manuscript.

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REFERENCES

- 1 **Agha RA**, Borrelli MR, Farwana R, Koshy K, Fowler AJ, Orgill DP; SCARE Group. The SCARE 2018 statement: Updating consensus Surgical CAse REport (SCARE) guidelines. *Int J Surg* 2018; **60**: 132-136 [PMID: [30342279](#) DOI: [10.1016/j.ijssu.2018.10.028](#)]
- 2 **Kitamura H**, Yamamoto D, Kadoya S, Bando H, Okayama Y, Minato H. Jejunal mesenteric lymphangioma treated by pancreaticoduodenectomy: A case report. *Int J Surg Case Rep* 2020; **77**: 165-169 [PMID: [33161289](#) DOI: [10.1016/j.ijscr.2020.10.088](#)]
- 3 **Erguney S**, Teksoz S, Erdamar S, Ainechii S, Olgun DC. Extended pancreaticoduodenectomy for a huge cystic-cavernous lymphangioma: a case report. *JOP* 2012; **13**: 289-291 [PMID: [22572134](#)]
- 4 **Vanek VW**, Phillips AK. Retroperitoneal, mesenteric, and omental cysts. *Arch Surg* 1984; **119**: 838-842 [PMID: [6732494](#) DOI: [10.1001/archsurg.1984.01390190076018](#)]
- 5 **Ouaissi M**, Emungania O, Sebbag H, Lehors-Albous H, Boutboul R. [Cystic lymphangioma of the lesser omentum]. *Presse Med* 2003; **32**: 1165-1166 [PMID: [13677878](#)]
- 6 **Mabrut JY**, Grandjean JP, Henry L, Chappuis JP, Partensky C, Barth X, Tissot E. [IMesenteric and mesocolic cystic lymphangiomas. Diagnostic and therapeutic management]. *Ann Chir* 2002; **127**: 343-349 [PMID: [12094416](#) DOI: [10.1016/s0003-3944\(02\)00770-8](#)]
- 7 **Yasoshima T**, Mukaiya M, Hirata K, Takashima T, Kashiwagi K, Kukita K, Homma H, Niitsu Y. A chylous cyst of the mesentery: report of a case. *Surg Today* 2000; **30**: 185-187 [PMID: [10664346](#) DOI: [10.1007/s005950050040](#)]
- 8 **Suthiwartnarueput W**, Kiatipunsodsai S, Kwankua A, Chaumrattanakul U. Lymphangioma of the small bowel mesentery: a case report and review of the literature. *World J Gastroenterol* 2012; **18**: 6328-6332 [PMID: [23180956](#) DOI: [10.3748/wjg.v18.i43.6328](#)]
- 9 **Watanabe A**, Suzuki H, Kubo N, Kobayashi T, Araki K, Sasaki S, Shimura T, Oyama T, Kuwano H. A case of mesenteric cystic lymphangioma in an adult which caused duodenal stenosis after resection. *Int J Surg Case Rep* 2013; **4**: 212-215 [PMID: [23287062](#) DOI: [10.1016/j.ijscr.2012.10.023](#)]
- 10 **Lee DL**, Madhuvrata P, Reed MW, Balasubramanian SP. Chylous mesenteric cyst: A diagnostic dilemma. *Asian J Surg* 2016; **39**: 182-186 [PMID: [23769746](#) DOI: [10.1016/j.asjsur.2013.04.009](#)]
- 11 **Javed A**, Pal S, Chattopadhyay TK. Chylolymphatic cysts of the mesentery. *Trop Gastroenterol* 2011; **32**: 219-221 [PMID: [22332339](#)]
- 12 **Cudia B**, D'Orazio B, Cali D, Di Vita G, Geraci G. Lymphatic Mesenteric Cyst, a Rare Cause of Surgical Abdominal Pain: Case Report and Review of the Literature. *Cureus* 2020; **12**: e11766 [PMID: [33409014](#) DOI: [10.7759/cureus.11766](#)]



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