Case Report

Chylolymphatic Mesentric Cyst: A Rare Case Report, Mimicking as Dermoid Cyst on Imaging

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Abstract

Introduction – A chylolymphatic cyst is a rare variant of a mesenteric cyst. These cysts present within the mesentry, lined with thin endothelium or mesothelium and filled with chylous & lymphatic fluid. Although often asymptomoatic, patient may present as vague abdominal pain, with or without palpable mass. Ultrasonography and CT may suggest the diagnosis but histopathological examination is required for confirmation after surgical excision. A chylolymphatic cyst should be considered in one of the differentials of the cystic lesion of abdomen with fat-fluid level, same as mesenteric cystic teratoma. **Aim**- We reported this case, as chylolymphatic mesenteric cysts are extremely rare & very little information is available regarding their presentation, differentials & complications. **Case Report**- We reported a case of 22 years old woman, admitted with occasional abdominal pain for 4- 5 years, aggravated since 6 months. She was evaluated with an ultrasound and subsequently CT scan of the abdomen was done.USG revealed a hypoechoic lesion measuring 10x10 cm in left upper abdomen, separated from the tail of pancreas. CT was planned, showed a well defined rounded hypodense cystic mass with fluid attenuation & fat-fluid level. On contrast examination, it showed only wall enhancement. As taking into consideration of characteristic fat- fluid level, presumptive diagnosis of mesenteric cystic teratoma was given. The patient got operated. On histopathological examination, it showed milky white fluid, cyst wall lined with endothelium with lymphoid follicles & foam cells. All these findings confirmed the diagnosis of chylolymphatic cyst. **Discussion** – Mesentric cyst represent a rare pathologic entity. Their existence was first reported in 1507 by Bennevieni. It may occur anywhere in the mesentry of GIT , but most commonly in the mesentry of the small intestine. Among these uncommon cystic lesions of the mesentry, chylolymphatic cyst is extremely rare sub classification of mesenteric cyst, specially of lymphatic origin. It constitute 7.3% to 9.5% of all abdominal cysts. They were first described by Rokitansky in 1842. These cysts may be asympomatic and may cause vague abdominal pain, lump or abdominal distention or It may present with complications. Chylolymphatic cysts, as name suggests, contain both chyle & lymph. Abdominal USG is initial investigation of choice. CT/MRI may additionally demonstrate the fluid attenuation of the lesion. Some authors have described the characteristic appearance of fluid level of differing echodensities, i.e. an upper fatty echodensity of chyle, on top of the water echodensity of lymph in a well defined cystic lesion. Although imaging findings may suggest the diagnosis but It will be confirmed only on histopathological examination after surgical exploration. **Conclusion**- Although very rare, chylolymphatic cyst should be kept in mind as one of the differential diagnosis of cystic masses of the a bdomen with fat-fluid level. The definitive diagnosis of chylolymphatic cyst is only confirmed after histopathology. Complete surgical excision of the cyst ensures excellent prognosis.

Keywords- chylolymphatic cyst, mesenteric cyst, fat-fluid level

Introduction

A chylolymphatic cyst is a rare variant of a mesenteric cyst[1,2]. These cysts present within the mesentry, lined with a thin endothelium or mesothelium and filled with chylous & lymphatic fluid[3]. Although mesenteric cysts in general have been reported in the literature fairly frequently, chylolymphatic cysts are extremely rare in modern medical literature[2].

Therefore very little information is available regarding their presentation, differentials and complications.

Chylous mesenteric cysts have an estimated incidence of 7.3% of all abdominal cysts[4]. It was first described by Rokitansky in 1842[5]. Patient generally present with abdominal distension associated with vague abdominal pain with or without a palpable mass [6, 7]. Although often asymptomatic 10% of patients with such cysts present as acute abdomen [8] chylous cyst should be differentiated from other fluid filled abdominal cystic masses such as mesenteric, omental and dermoid cysts[9,10,11].

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Ultrasonography and computed tomography suggest the diagnosis but histopathological examination is required for confirmation . Complete excision of the cyst ensures excellent prognosis and is curative.

Case report

A 22 year old woman was admitted with occasional abdominal pain for 4-5 years, aggravated since 6 months. The pain was moderate intensity. She had no other complaints and his physical examination was unremarkable. His biochemistry parameters including serum amylase and lipase were within normal range. She was evaluated with an ultrasound and subsequently a CT scan of the abdomen done.

USG revealed a hypoechoic lesion measuring 10x10 cm in left upper abdomen, separated from the tail of pancreas, CT scan showed a well defined round hypodense cystic mass with fluid attenuation value & fat fluid level, show only wall enhancement on post contrast study in small bowel mesentry. As taking into consideration of characteristic fat fluid level, presumptive diagnosis of Dermoid cyst was given.

The patient was planned for surgery, open lapratomy was considered because in laparoscopy enucleation of the cyst could not be performed safely due to adhesion of the cyst wall to surrounding mesenteric tissue. On surgical exploration a 10x10 cm cyst was seen arising from the mesentry of proximal jejunum. The cyst was successfully

enucleated from the mesentry. The cyst contained milky white fluid consistent with a chylolympatic cyst. On histopathology, it revealed a cyst wall lined with endothelium having lymphoid aggregates and foam cells.

Her postoperative course was uneventful and feeding was started on 2nd post up day. She was discharged on the 5th post op day. A 6 month follow up with abdominal ultrasound and CT imaging showed no signs of recurrence.

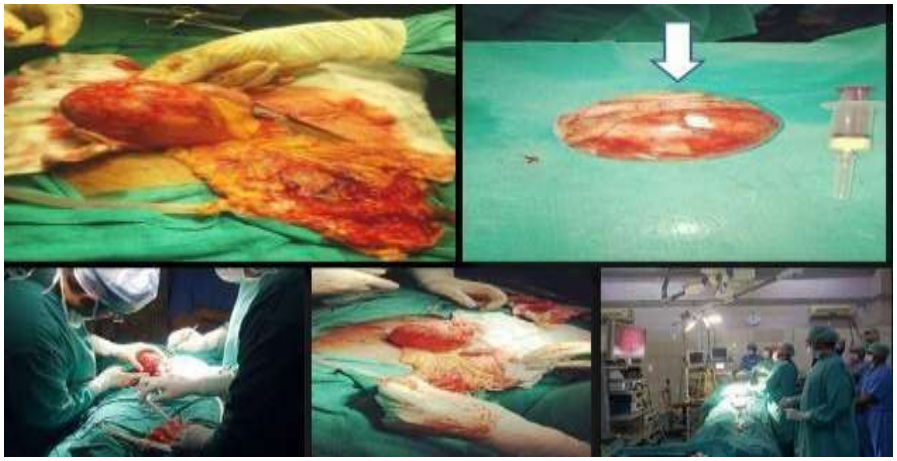

Fig 1: Photographs of the mass after ex situ incision and drainage of the milky white fluid.

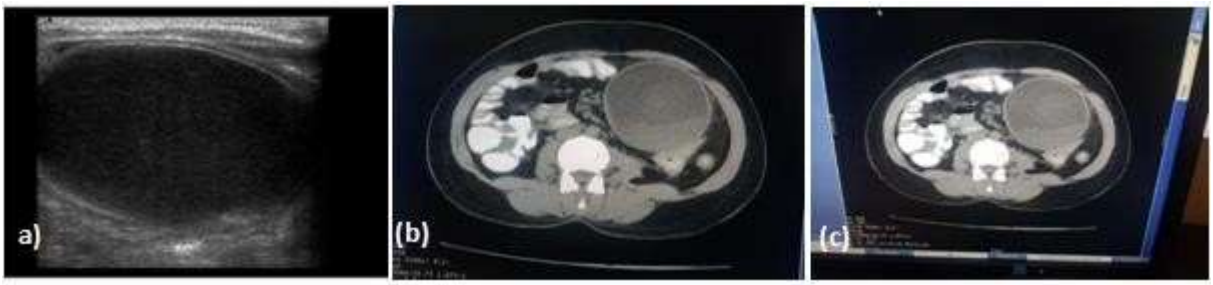

Fig 2: (a) USG shows a well defined hypoechoic cystic lesion in left upper abdomen. CT Scan of the abdomen shows (b) a large cystic lesion with fat fluid level in the small bowel mesentry adjacent to a loop of small bowel, (c) minimal wall enhancement on contrast.

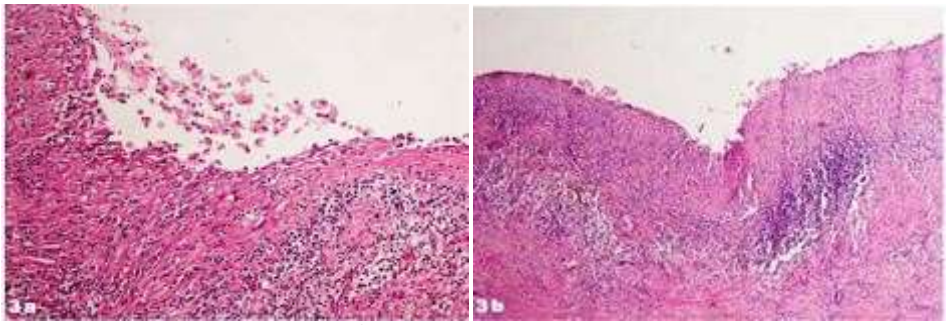

Fig 3: (a):Photomicrograph showing cyst wall with foamy macrophages in the lumen (H& E ,X100), (b): cyst wall with lymphoid aggregates(H&E,X40)

Discussion

Mesenteric cyst represents a rare pathologic entity. Their existence was first reported in 1507 by Bennevieni, an Italian anatomist, when he performed autopsy of an 8yr old boy and the first effective surgical excision was performed in 1880 by Tillaux [12, 13]. These are rare intrabdominal tumour with prevalence about 1:1,00,000 in adults and 1:20,00,000 lakh in paediatric hospitals admission[14]. Mesenteric cysts may occur anywhere in the mesentry of GIT from the duodenum to the rectum but most commonly localized in the mesentry of the small intestine followed by the mesentry of the large intestine and reproperitoneum [14-18]. In a review of 162 cases of mesenteric cysts by Kurtz et al ; they found that 60% were located in the mesentry of small bowel , 24% located in the mesentry of large bowel, and 14.5% were retroperitoneal [14]. Mesenteric cysts may vary in the size from 4 cm to 30 cm [16,19] .

Mesenstric & omental cysts are unique for their diverse clinical presentation, etiology, radiological features, and pathological characteristics – initial classification proposed by Beahers et al 195033 (TABLE:1), recognised 4 categories of mesenteric and omental cysts . It has now been categorized into 6 types by the updated classification by Marc De Perrot 200020 (TABLE:2).

Table 1: Mesentric cyst classification by beaher's et al (1950)

1. Developmental
2. Traumatic
3. Infective

4. Neoplastic

Table 2: New updated classification by Marc De Perrot (2000)

1. Cysts of lymphatic origin – Simple lymphatic cysts, Lymphangioma.
2. Cysts of mesothelial origin – Simple mesothelial cyst, Benign cystic mesothelioma , Malignant cystic mesothelioma.
3. Cysts of enteric origin – enteric duplication cyst, Enteric cyst.
4. Mucinous cystic neoplasms – mucinous cystadenoma, Borderline malignant mucinous cystic neoplasm, Mucinous cystadenocarcinoma.
5. Cysts of urogenital origin.
6. Miscellaneous neoplasms – Mature cystic teratoma (dermoid cysts), Neuroendocrine carcinoma, Cystic spindle cell tumor.
7. Nonpancreatic pseudocysts – Cysts of traumatic origin, Cysts of infectious origin.
8. Nonneoplastic cysts – Hydatid cyst, Mycotic cyst, Parasitic cyst, Tuberculous cyst, Cystic degeneration of lymph nodes and other tissues.

Among these uncommon cystic lesions of the mesentry, chylolymphatic cysts is a extremely rare subclassification of mesenteric cyst, specially of lymphatic origin & usually represents as benign lesions [21,33]. It constitute 7.3% to 9.5% of all abdominal cysts. They were 1st described by Rokitansly in 1842[22]. They are most often unilocular & solitary [23] & frequently observed in association with the small bowel [24]. The pathogenesis remains unclear and there are several theories that have been proposed for the formation of these cysts, one of them suggested that they represent benign proliferation of ectopic lymphatics which lack communication with the main lymphatic system . Another theory proposed that embroyonic lymphatic channel gradually become enlarged, due to the failure of joining the venous system. It may also result from trauma to lymphatic channels. Finally it has been proposed that contusion of the mesentry, results in accumulation of lymphatic fluid within this space[21].

The chylolymphatic cyst, as indicated by its name, contains both chyle and lymph. The accumulation of chyle & lymph is considered to be the result of an imbalance between the inflow and outflow of fluid [1]. These cysts may be asymptomatic, and may cause abdominal distension, lump or vague abdominal pain. It may present with complications such as intestinal obstruction, hemorrhage, infection, rupture of the cyst, volvulus or obstruction of the urinary or billary tract [14-19,25-27]. There are cases in which chylous cysts mimicked rupture of abdominal aortic ancurysm and pancreatitis [28,29] it has also been estimated that malignant transformation may occur in 3% of such cysts [31,32].

The definitive diagnosis of these lesions is difficult prior to surgical exploration as there are no pathognomonic symptoms or characteristic imaging findings[30].

Abdominal radiographs are usually non contributory, however it may reveal dilated bowel loops, with air fluid level in the very rare patients with intestinal obstruction, which may result from compression of the adjacent bowel by the cyst or by mesenteric volvulus[30].

Abdominal ultrasonography is currently the initial imaging procedure of choice. This delineates the nature of the mass, organ or site of the origin, extent and associated mass effects on the kidney or liver, if any. In a chylolymphatic cyst, a 'fluid- fluid level' can be seen on ultrasonography due to formation of an upper fluid level by lighter chyle over the lower fluid level of heavier lymph [34].

A CT/MRI scan may additionally demonstrate the fluid attenuation of the lesion, its relationship with the adjacent viscera and vessels[35]. Some authors have described the characteristic appearance of a chylolymphatic cyst on CT in the form of the presence of fluid levels of differing echodensities, that is to say, an upper fatty echodensity of chyle on top of the water echodensity of lymph in a well defined cystic lesion[34, 36].

As demonstration of the fat fluid level without calcification is also diagnostic of dermoid cyst provided that the element denser than normal fat is gravity dependent[37], cyst was primarily diagnosed as a case of dermoid cyst in our case, however came out as a rare chylolymphatic cyst on histopathological examination after surgical exploration.

Some other pathological lesions like Nonpancreatic pseudocyst also show chylous content & may present as fat-fluid level. Fat fluid level had also been described in case of well differentiated liposarcoma of the retroperitoruem[38].

In case of large chylous cyst, especially in symptomatic cysts, surgical excision is advised to prevent a potential malignant transformation and to avoid development of complication. The different surgical approaches used are marsupialisation, sclerotheraphy, drainage, enucleation, percutaneous aspiration and excision of cyst with or without resection of the involved gut[39-42]. The preferred technique entails open or even laparoscopic enucleation of the mesenteric cyst, that is the atraumatic separation of the cyst from the surrounding leaves of the mesentry[28,29,32,34,35,38]. Whenever enucleation can't be performed safely, due to adhesions of the cyst wall to surrounding mesenteric tissue and or other structures, a resection of adjacent organs may be necessary. It has been reported that bowel resection is necessary for only one out of three of treated adults[43]. Partial excision of cyst, drainage and deroofing have also been described as potential treatment options; however the last 2 options is particularly have been associated with higher likelihood of recurrences and thus are best avoided[34,35,38,43,44]. Endoscopoic removal was also referred in the literature but the method has many limitations[44].

Histopathological examination of surgical specimen may reveal a unilocular or multilocular cyst, containing milky white viscous fluid with chylomicrons. It also shows cholesterol crystals and triglycerides that are diagnostic for chyle[38]. The cysts are usually lined with single layer of endothelium and may contain lymphoid tissue & foam cells[1, 45]. CD[31] is used primarily to demonstrate the presence of endothelial cells and can help to evaluate the degree of tumour angiogenesis.

The differential diagnosis of these cysts include lymphangioma, pseudocyst, haemongioma, endometriosis, loculated ascites (usually tuberculous), peritoneal inclusion cysts, cystic mesenteric panniculitis, sclerosing mesentritis, hydatid cyst, cystic teratoma and urogenital cysts[32]. Cystic lymphangioma has strict resemblance to chylolymphatic cyst both grossly and microscopically, some authors considered chylolymphatic mesenteric cyst to be a type of lymphangioma but some literatures also show authors describing chylolymphatic cyst as a variant of mesenteric cyst[46, 22]. The absence of smooth muscle & lymphatic spaces in the wall of the cyst differentiates chylolymphatic cyst from mesenteric lymphangioma[22].

Conclusion

Although very rare, chylolymphatic cyst should be kept in mind as one of the differential diagnosis of cystic masses of the abdomen. The definitive diagnosis of theses lesions is difficult prior to surgical exploration as there are no pathognomonic symptoms or characteristic imaging findings, histopathological examination is required for confirmation. Complete surgical excision the cyst ensures excellent prognosis and is curative.

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