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Case report

Chylolymphatic mesenteric cyst with midgut volvulus in an adolescent: a peculiar presentation [☆]

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ARTICLE INFO

Article history:

Received 25 October 2021

Accepted 30 October 2021

Keywords:

Mesenteric cyst

Chylolymphatic cyst

Midgut volvulus

Whirlpool sign

Intestinal obstruction

ABSTRACT

Mesenteric cysts are rare intra-abdominal masses in the pediatric population, with the chylolymphatic variant comprising only 7.3% of all abdominal cysts. These cysts can have a varied clinical presentation, ranging from asymptomatic cystic masses to intestinal obstruction. We report a 16-year-old female, who presented with acute abdominal pain and vomiting, and was diagnosed with an intra-abdominal cystic mass with midgut volvulus accompanied by the classical “whirlpool sign” on imaging.

She underwent laparoscopic abdominal exploration, which revealed a large chylolymphatic mesenteric cyst associated with feeder lymph vessels. This cyst had resulted in volvulus of the small bowel. The bowel was detorsed and found to be viable, and the cyst was removed en bloc after suction evacuation with ligation of the lymphatic feeder vessels.

Midgut volvulus in the pediatric population is usually secondary to malrotation, but in this case the patient's small intestine was not found to be malrotated, and hence we present this case and urge physicians to consider a diagnosis of midgut volvulus even in the absence of malrotation in a child with a cystic abdominal mass presenting as intestinal obstruction.

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Case report

We present a previously healthy 16-year-old female who presented to our emergency department with severe back and abdominal pain as well as vomiting. She complained of intermittent back pain for the preceding one week which worsened on the day of presentation, accompanied by multiple episodes of vomiting with epigastric pain. On examination she

was afebrile at 36.6C, with a normal blood pressure and a normal heart rate. On palpation, the abdomen was soft with mild diffuse tenderness but no signs of peritoneal irritation. Costovertebral angle tenderness was absent bilaterally, but the child complained of pain on the left side of the back on palpation. She was given a fluid bolus as well as IV ketorolac for pain. CBC was unremarkable including a normal hemoglobin at 12.4 and a normal white blood cell count at 8000 cells/mL.

[☆] Competing Interests: The authors have declared that no competing interests exist.

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<https://doi.org/10.1016/j.radcr.2021.10.066>

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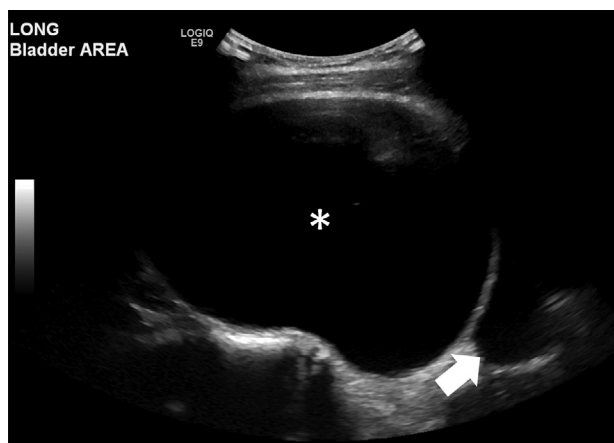


Fig. 1 – 16-year-old female with a pelvic chylolymphatic mesenteric cyst. Grayscale, longitudinal sonographic image demonstrates a very large cystic mass (asterisk) superior to the bladder (arrow).

Urine dipstick was also normal, with no nitrites, leucocytes, or occult blood.

An ultrasound of the kidneys was obtained to evaluate for renal obstruction and hydronephrosis. It revealed a 13.5 cm cystic lesion in the pelvis (Fig. 1) and right sided hydronephrosis, likely secondary to ureteral compression from the cyst. Computed tomography (CT) with intravenous contrast redemonstrated the pelvic mass, most consistent with either a mesenteric cyst or duplication cyst (Fig. 2). The CT also noted swirling of the mesenteric vessels consistent with volvulus (Fig. 2).

She was taken to the operating room for laparoscopic abdominal exploration and a large jejunal mesenteric cyst identified (Fig. 3A), along with intestinal volvulus of at least 360 degrees with no evidence of malrotation. On decompressing

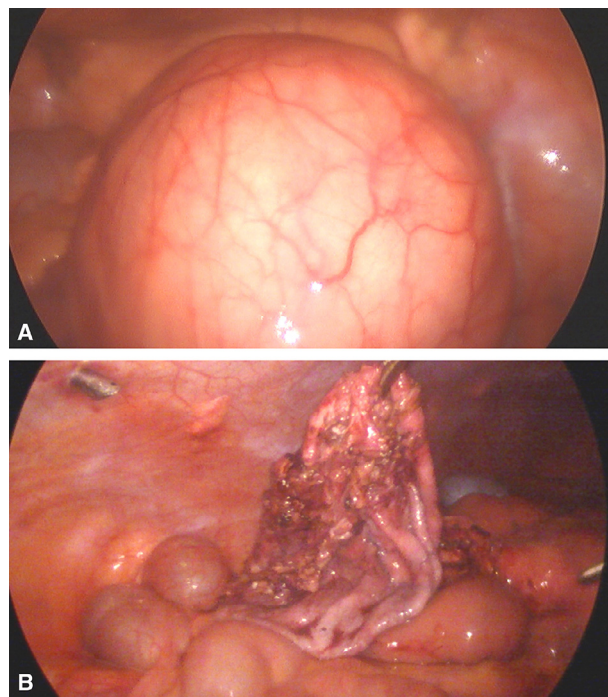


Fig. 3 – 16-year-old female with pelvic chylolymphatic mesenteric cyst. (A) Intraoperative findings confirmed a large cyst in the mesentery with volvulus. (B) Dissection of the cyst freed from the mesentery with noted feeding structures.

with suction, chyle was noted, as was a thick wall separate from the bowel. Laboratory analysis noted the fluid triglyceride content was 8850 mg/dL. The bowel was placed into normal position and the cyst was carefully dissected free from the mesentery without disturbing the blood supply to the associated small bowel. Two lymphatic structures appeared to be

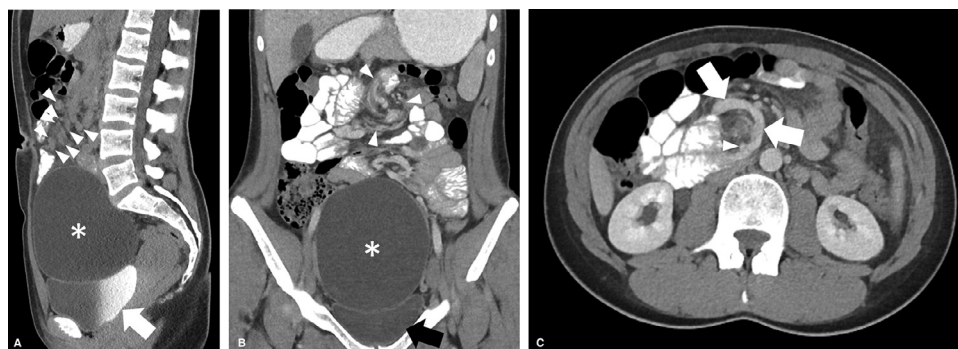


Fig. 2 – 16-year-old female with volvulus secondary to a pelvic chylolymphatic mesenteric cyst. (A) Sagittal CT image in soft tissue windows of the abdomen and pelvis following intravenous contrast material administration demonstrates a large cystic mass (asterisk) superior to the bladder (arrow) which contains layering excreted contrast material. There is swirling of the mesenteric vessels (arrowheads) about the superior mesenteric artery and vein vascular pedal, which has been classically described as a “whirlpool.” (B) Corresponding coronal CT image shows the large cystic mass (asterisk) superior to the bladder (arrow). There is swirling of the mesenteric vessels about the vascular pedicle within the midabdomen (arrowheads). (C) Corresponding axial CT image in the midabdomen demonstrates the classic “whirlpool” sign as the SMV (arrow) wraps around the SMA (arrowhead).

entering the cyst, and these were ligated with 5 mm clip appliers. The cyst (Fig. 3B) was removed via the umbilicus. She was discharged the following day on oral acetaminophen and ibuprofen. On telephonic follow-up with her guardian, she appears to be doing well with school and regular activities and has no further complaints or evidence of recurrence.

Discussion

We report a case of chylolymphatic mesenteric cyst in an adolescent female which presented as midgut volvulus with vomiting and pain abdomen. Mesenteric cysts are a rare diagnosis in adults, and even more so in the pediatric population with a reported prevalence of 1 per 20,000 children, 25% of which are diagnosed by 10 years-of-age [1–6]. They are usually a result of lymphatic malformations, traumatic injuries, or local infections [7]. In 2000, de Perrot et al. proposed a new system of classifying mesenteric cysts based on histopathological features - (i) lymphatic origin (simple cysts and lymphatic malformations); (ii) mesothelial origin (simple, benign and malignant cystic mesothelioma); (iii) enteric origin (enteric cyst and enteric duplication cyst); (iv) urogenital origin; (v) mature cystic teratoma (dermoid cysts); and (vi) pseudocysts (infectious/traumatic cysts) [4,6].

The clinical manifestations of mesenteric cysts can range from asymptomatic intra-abdominal masses discovered incidentally, to acute abdomen secondary to intestinal obstruction (by extrinsic compression or volvulus) or from cyst rupture and hemorrhage. Our patient presented with midgut volvulus, which is most commonly a consequence of embryological intestinal malrotation, resulting from abnormal anatomical fixation of the bowel and mesentery within the abdomen [8–12]. This predisposes the small bowel to undergo volvulus along the axis of the superior mesenteric artery. The normal anatomic relation of the superior mesenteric artery and vein is skewed and presents as the classical “whirlpool sign” on ultrasound and CT (Fig. 2C). In this process, the blood flow via the superior mesenteric artery is occluded, leading to ischemia and even gangrene of the involved bowel segment.

Whirlpool sign was observed in our patient during imaging (Fig. 2C), along with a large cystic intraperitoneal mass (Figs. 1 & 2). This led us to consider the common differential with whirlpool sign that is, volvulus resulting from intestinal malrotation, yet the CT findings noted the ligament of Treitz was in the correct location, and laparoscopic abdominal exploration confirmed this. The thick-walled large cyst in the mesentery of the jejunum appeared to be the fulcrum around which the small bowel volvulized.

The most common differential diagnoses to consider in this case are enteric duplication cysts, mesenteric cysts, and broad ligament/ovarian cysts [12]. Enteric duplication cysts can be difficult to differentiate from mesenteric cysts preoperatively, as both types of cysts can arise anywhere along the gastrointestinal tract from the duodenum to the rectum, and can have identical clinical features of abdominal mass

and intestinal obstruction [12]. As such, a confirmatory diagnosis can only be made after exploration [7]. The high triglycerides confirmed the diagnosis of a lymphatic associated anomaly.

Although mesenteric cysts are rare in the pediatric population and midgut volvulus is a rare occurrence in the absence of intestinal malrotation, these diagnoses should be considered in any pediatric patient presenting with acute abdomen with signs of intestinal obstruction. Complete excision of the cyst along with detorsion of the twisted bowel segment with minimally invasive laparoscopic surgery is the treatment of choice for such patients.

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