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Case Report

Mesentric lymphatic malformation presented with a small bowel internal hernia: A case report ☆

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ARTICLE INFO

Article history:
Received 28 July 2022
Revised 1 October 2022
Accepted 6 October 2022
Available online 2 November 2022

Keywords:
Cystic peritoneal lesions
Lymphangioma
Lymphatic malformation
Internal hernia
Abdominal distention

ABSTRACT

We report a case of a 2 years and 8 months boy presented with a small bowel obstruction occurring in a setting of mesenteric lymphatic malformation (LM). The case did not showed typical features of LM and thought to be a simple ascites. Nevertheless, the mass effect of the lesion was the salient finding and the case presented with a small bowel internal hernia which is a rare presentation. This emergent case was managed surgically.

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Introduction

Mesenteric lymphatic malformation (LM) is known as mesenteric lymphangioma or mesenteric cyst is a benign developmental anomalies in which focal lymphatic channels fail to establish connections with the central lymphatic system. LM occur in many anatomic locations, 95% in the neck and axillary regions and 5% in the abdomen. The rare abdominal LM location include the retroperitoneum, mesentery, solid organs (liver, spleen, and pancreas) and gastrointestinal tract [1].

Case report

A 32-month-old boy, normally developed, had frequent visits to the emergency room with constipation and non-bilious

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2022.10.021

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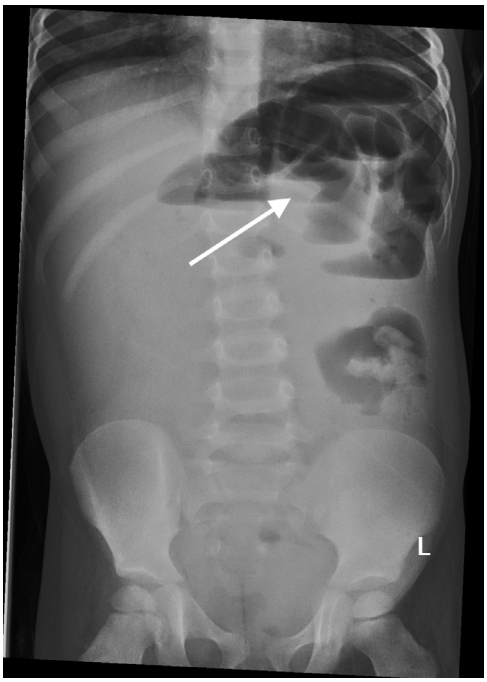


Fig. 1 – Abdominal X-ray. Displaced bowel loop into the left upper quadrant (white arrow), no abnormal calcification, free air, or pneumatosis.

vomiting that improved with fleet enema and prescribed laxative. Abdominal radiographs were showed gaseous and non-specific bowel loop distributions. Abdominal ultrasound demonstrated bilateral hydronephrosis and mild ascites that thought to be insignificant at one emergency room visit. After three months, the patient presented with worsening constipation, development of abdominal distention, pain, tenderness all over the abdomen, and non-bilious vomiting. Laboratory workup revealed normal blood chemistry and other laboratory result. However, abdominal radiograph demonstrated abdominal mass-effect displacing the bowel loops into left upper quadrant, distended loops with multiple air-fluid level Fig. 1. Corresponding ultrasound presented dilated bowel and ascites with floating debris, there were stable bilateral hydronephrosis Fig. 2. Computed tomography (CT) scan indicated features of mechanical proximal small bowel obstruction with bowel loops localized in the expected region of the lesser sac, and large ascites/cystic lesion. The impression was strangulated lesser sac, internal hernia with large ascites and no bowel ischemia Fig. 3.

Consequently, the patient was booked for laparotomy, possible bowel resection and anastomosis, and possible stoma creation.

During the surgery, there were 2 huge cysts, arising from the mesentery of the small bowel 15 cm from the duodenojejunal junction. These cysts were huge in size. There was no

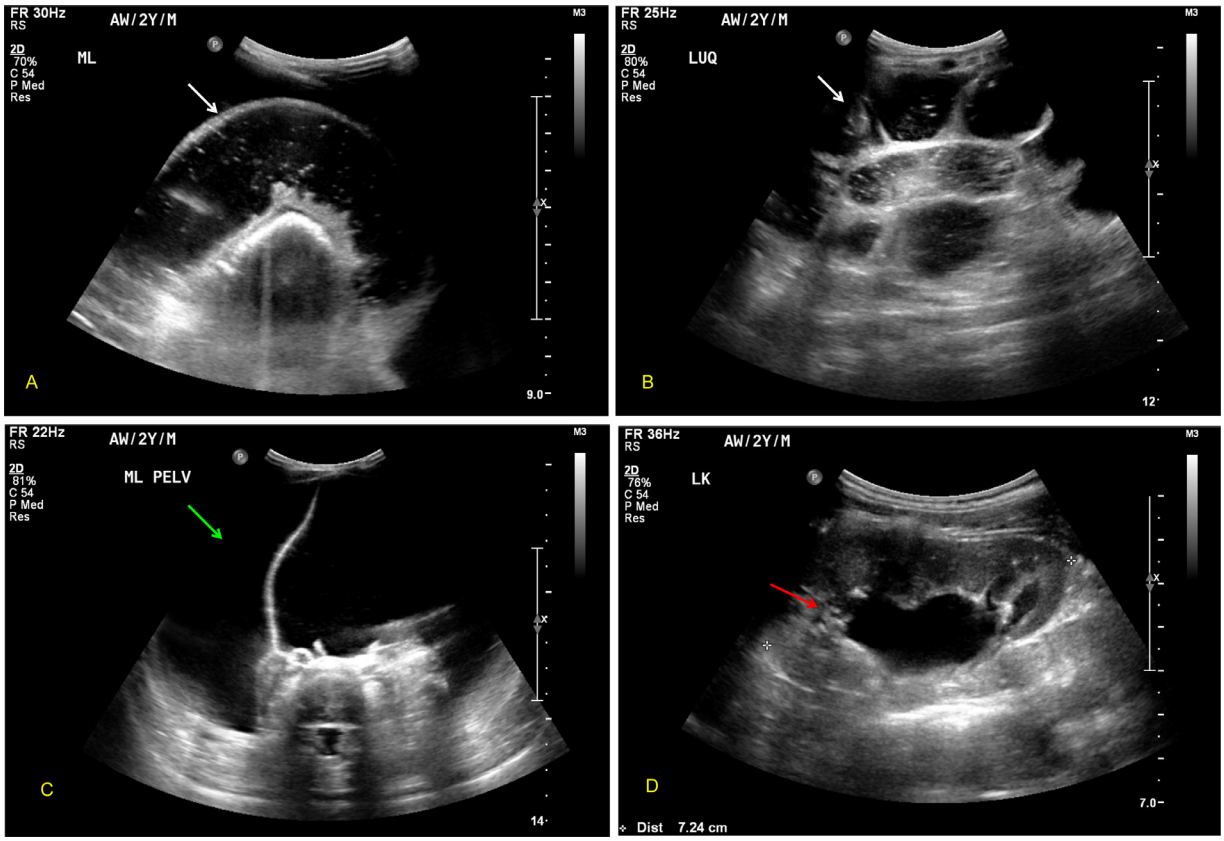


Fig. 2 – Abdominal ultrasound. (A, B) Dilated owl segments at midline and LUQ (white arrows). (C) Large cystic lesion occupying the pelvis with separation and floating debris (green arrow). (D) Left kidney with dilated renal pelvis (red arrow); hydronephrosis were noted bilaterally.

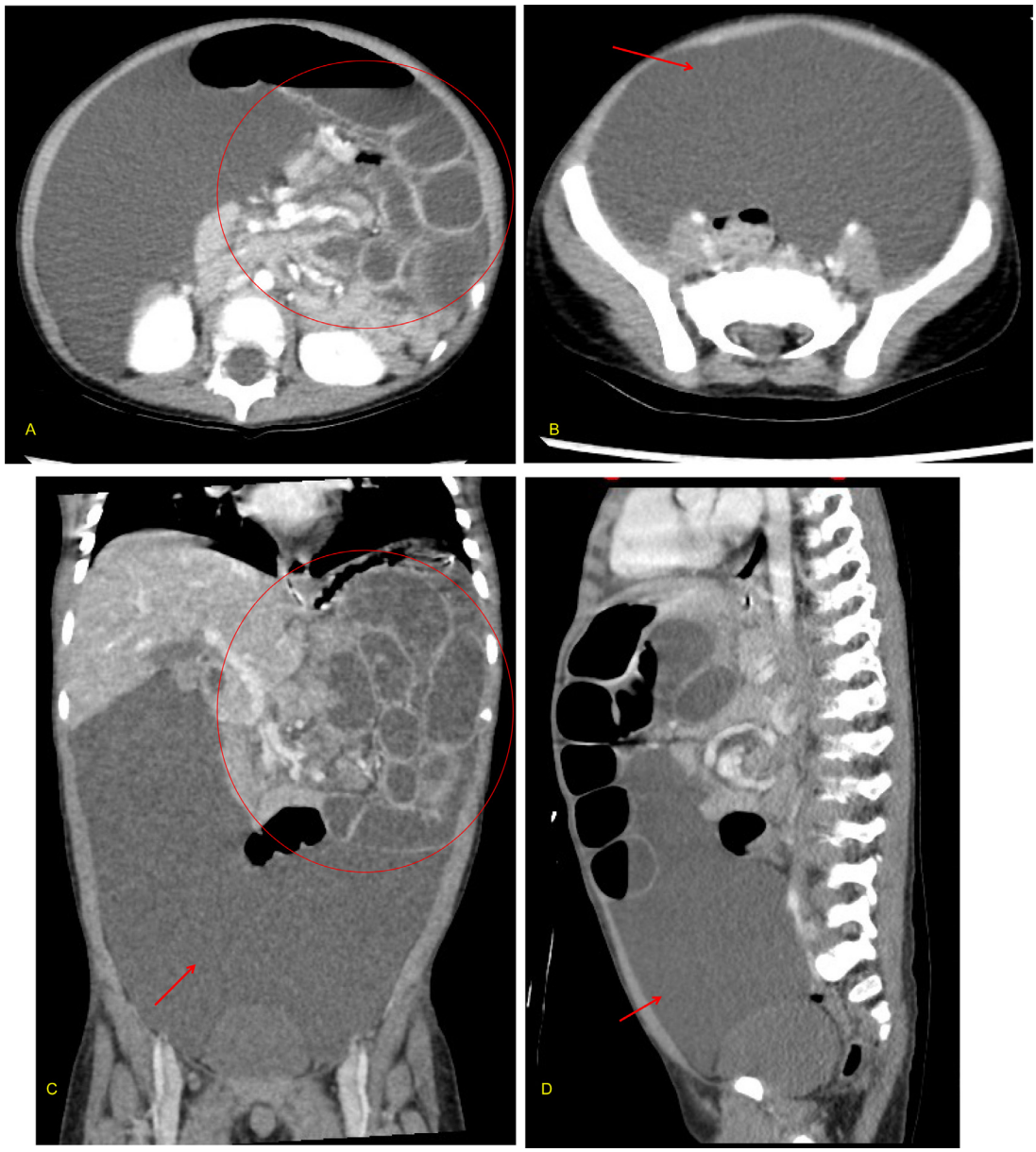


Fig. 3 – CECT. Bowel loop are localized into the LUQ, with patent vessels (red circles). The cystic lesion occupying the whole abdomen with homogenous contents (red arrows). There were no sold component, abnormal abdominal calcification, pneumatosis, or free air.

other obvious pathology. The decision was made to resect the lesion with part of the small bowel because of the arising of the cysts from the mesentery and resection of the cyst alone might jeopardize the bowel.

The lesion consists of 2 intact separated cysts attached to the mesentery of a segment of small intestine (Fig. 4A). The first cyst measures 15 × 13 × 2.5 cm and the second one measures 15 × 10 × 2.5 cm. Segments of small bowel measures 10 × 1 × 1.5. The outer surface of both cysts is smooth and glistening. Opening of both cysts reveled green to brown serous fluid. No solid components identified. The inner lining of both cysts is unilocular and smooth. Smaller cysts are noted within the mesentery fat containing white milky material. The mucosa of the small bowel is unremarkable.

Clinically the patient is doing well, following up regularly for hydronephroses, and instructed for dietary modification to avoid constipation.

Discussion

Of all lymphangiomas, 65% occur at birth, and 90% occur during the first 2 years of life. The clinical presentation depending on the size and location of the mesenteric lymphangioma. It can be asymptomatic for a lifetime [2–4], or produce serious complications such as hemorrhage, intestinal ischemia, obstruction, and volvulus [5]. In our study, bowel obstruction was the presenting symptom.

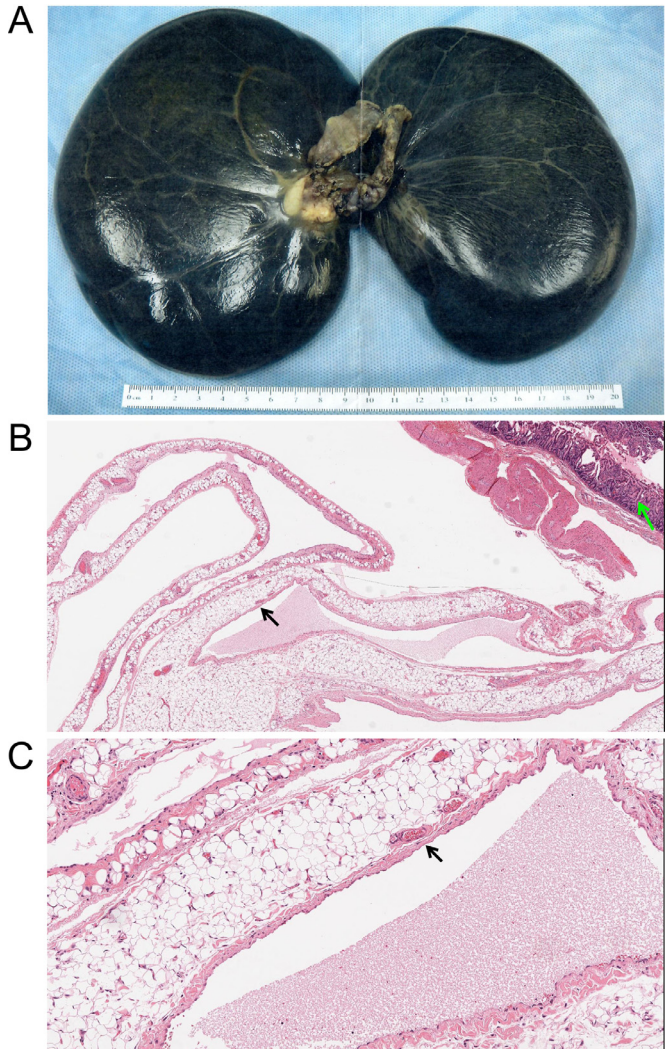


Fig. 4 – (A) Gross specimen. (B) Image from hematoxylin and eosin (H&E) stained slide shows small intestine (green arrow) with mesenteric cystic lymphangioma. (C) There are variable sized lymphatic cysts lined by a single layer of endothelial cells (black arrows).

Accurate anatomic localization is important in preoperative planning because lymphangiomas have an insinuating nature which makes complete surgical excision difficult sometimes. It might be challenging on imaging to differentiation lymphangiomas from other fluid-containing masses and ascites in the abdomen [6]. However, it is possible to differentiated from ascites with these criteria: the presence of septa, compression on adjacent intestinal loops, and lack of fluid in the dependent recesses of the peritoneum such as the paracolic gutters and subhepatic spaces and between the leaves of the small-bowel mesentery [1,7].

Our case was different in several ways. There was only a mass effect on adjacent bowel and appears on imaging as internal hernia. Although the location of the lesion near the duodenojejunal junction, the usual area of twisting in case of a volvulus [8], our patient did developed an internal hernia instead.

Ultrasound is an excellent initial imaging modality when a mesenteric LM is suspected, to confirm the cystic nature of the lesion. When the macrocysts are tubular, mesenteric LMs can mimic dilated fluid filled bowel loops, the presence of a gut signature and peristalsis being differentiating features [9]. Lymphangiomas are usually anechoic or may contain internal echoes or sedimentation, with fluid–fluid levels caused by debris [10].

On CT lymphangiomas are cystic masses, with attenuation values of water if the content is serous, and fat values if the content is chylous [6]. Hemorrhagic content with high attenuation on CT and calcifications are uncommon features [11]. After administration of intravenous contrast lymphangiomas may show enhancement of the cyst wall and septa. On magnetic resonance imaging, lymphangiomas have low-signal intensity on T1-weighted images and high signal intensity on T2-weighted images, similar to signal intensity of fluid. In the presence of chyle, signal drop may be seen in the opposed-phase chemical shift MR images [12]. If hemorrhage or infection occurs within the lesion, the appearance on CT and magnetic resonance imaging can be altered with the lesion appearing more solid [11].

Conclusion

A mesenteric lymphangioma should be considered when there is an abdominal developing cystic mass, causing mass effect on adjacent bowel. In our case, the lesion presented without typical features of septation, fatty attenuation or calcification neither on ultrasound nor CT examinations. However, the clue for radiological diagnosis of mesenteric lymphangioma was the mass effect, and the emergent issue in our case was the internal hernia.

Patient consent

Written informed consent was obtained from the parents.

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