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Newborn intestinal obstruction due to mesenteric lymphangioma: A diagnostic challenge

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ABSTRACT

Lymphatic malformations are uncommon benign cystic lesions that typically present in the neck and axilla during infancy. They are more rarely observed in the abdomen and extremities. This study presents the case of a newborn infant who was admitted to the hospital with vomiting. While the cause of the vomiting was investigated, a complete intestinal obstruction developed, and an emergency surgical intervention was performed. During a laparotomy, a cystic mass originating from the mesentery was discovered. A lymphangioma mass resulting in intestinal obstruction during the third week of life was not considered in the differential diagnosis.

1. Introduction

Lymphangiomas are tumors of the lymphatic system and are characterized by anastomosing lymphatic channels and cystic spaces varying in size; the diameter of such a mass is usually smaller than 0.5 cm, but they can reach much larger sizes [1]. These tumors occur due to an abnormal connection between lymphatic ducts and the venous system during the fetal period [2] and can be observed in any part of the body. Tumors are usually located in the neck, with a rate of 75%, and in the axilla, with a rate of 20%. Lymphangiomas occur less often in the thoracic and intra-abdominal visceral organs, with a rate of 4–5%. They affect the mesentery, omentum and retroperitoneal space at a rate of less than 1% [3]. The clinical symptoms depend on the size and location of cysts. This report presents the case of a male newborn with mesenteric lymphangioma and an unachievable preoperative diagnosis. Cases of mesenteric lymphangioma and newborn bowel obstruction in the literature were also reviewed.

2. Case report

A 20-day-old male infant was brought to the hospital after one week of vomiting, being unwilling to feed and passing less stool. The full-term infant was born vaginally to a 30-year-old mother with a gravidity of three and a parity of three. His birth weight was 2500 g. No specific features were present in the family history, and prenatal follow-up included ultrasonography (US). The infant did not present any complaints during the first two post-natal weeks; his feeding and defecation were normal, and the symptoms occurred and progressed in week three. On physical examination, mild dehydration, respiratory distress and abdominal distension were observed. No palpable abdominal mass was found. Mid-penile hypospadias and chordee were noted. Blood biochemistry and other laboratory results were normal. No metabolic alkalosis was observed. An erect plain abdominal radiograph demonstrated only a wide-based stomach with an air-fluid level and no gas or calcification in the remaining abdomen; the intestines were not visible on the erect radiograph (Fig. 1). US examinations were repeated twice under emergency and elective conditions; the resulting differential diagnoses included malrotation, annular pancreas or midgut volvulus. During that period, non-bilious vomiting gradually became bilious, and pyloric stenosis, an initial diagnosis, was eliminated. A contrast upper gastrointestinal (UGI) series was performed to evaluate the possibility of incomplete duodenal obstructions, such as type 1 atresia, intraluminal duodenal web (wind sock), superior mesenteric artery syndrome, intestinal malrotation-Ladd's bands, midgut volvulus, and duodenal stenosis. The contrast UGI results showed an enlarged stomach with contrast material retention, nondilated loops of the small bowel and no evidence of a double bubble (Fig. 2).

On day 5 after admission, a complete intestinal obstruction developed with a gradual deterioration in general condition. The amount of bilious gastric aspirate and abdominal distension

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Abbreviations

US	ultrasonography
UGI	upper gastrointestinal
CT	computed tomography

increased, stool discharge ceased, and an empty rectum was detected on rectal examination. Emergent surgical exploration using a right transverse incision above the level of the umbilicus was performed. Intraabdominal serous fluid was noted, and a sample was collected. A lymphangioma approximately 8 cm in diameter was observed; it had developed in the mesentery of the 7 cm proximal jejunum segment of the small intestine and completely occluded the intestinal lumen. The lymphangioma had thin walls, contained chylous fluid, and exhibited hemorrhagic areas in various sections; in addition, dilated lymph channels were visible (Figs. 3–5). No other pathology was observed during the abdominal exploration. The multiseptated cyst was excised along with the small intestine segment that it surrounded. Intestinal continuity was maintained with end-to-end anastomosis. Although no malrotation was observed, an elective appendectomy was performed after a risk and benefit assessment (Fig. 6). An intraabdominal drain was not inserted. The nasogastric catheter remained for 48 h. Oral feeding began on the third day. The patient was discharged on day 7. Growth culture and fluid cytology results were negative for the fluid from the abdominal cavity. A gross examination of the specimen showed enlarged lymph vessels, which contained lymph fluid. The diagnosis of lymphangioma was confirmed by a histopathological analysis. The cystic mass, which was completely located within the mesenteric boundaries, had a type 2 Losanoff pathological classification. The patient remained asymptomatic during a follow-up period of three years.

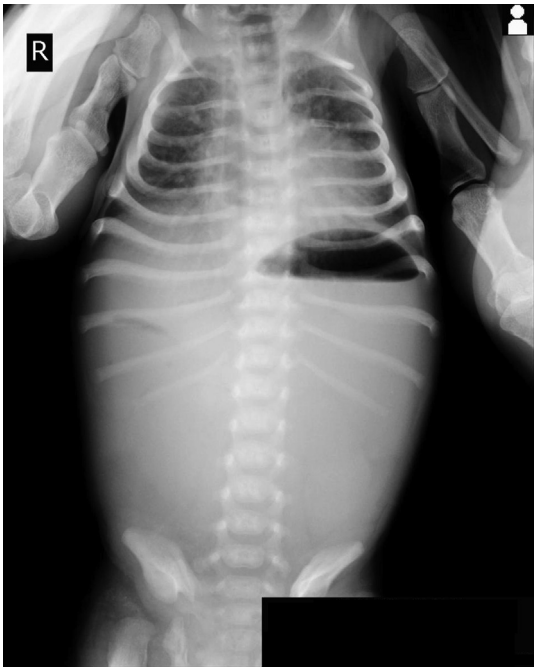

Fig. 1. Erect plain abdominal film showing a gasless abdomen with the exception of the air-fluid level in the stomach.

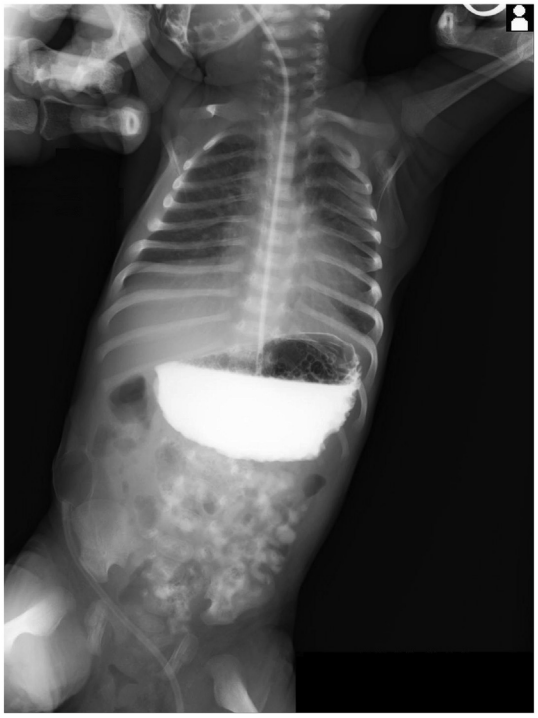

Fig. 2. Contrast UGI results showing a dilated stomach and a normal bowel configuration.

3. Discussion

The incidence of lymphangiomas is not precisely known because some cases are asymptomatic. The patient presented in this study was male, and hypospadias existed as an additional abnormality. The incidence of this condition in males and females varies according to reports in the literature. In this work, an electronic search of the English literature was performed to screen for relevant papers and calculate the male:female ratio; the 8 largest studies considering only mesenteric lymphangiomas included a total of 110 patients, and the male:female patient ratio was calculated to be 1.5/1 (66/44) [2,4–10]. Few associated pathologies were found in the search, including vascular malformation in three studies [4], Russell-Silver syndrome in one study [7], left renal agenesis in one study, pancreatic and renal cysts in one study [10], and intestinal malrotation in four studies [11–13]. No additional abnormalities have been reported in other case reports or series. In contrast to Antoniou et al., Weeda et al. suggested in a report of 2 cases that mesenteric lymphangiomas could be an acquired pathology that occurs as a result of malrotation and volvulus [11–13]. In this case, the cecum was in its normal location, and no malrotation of the intestine was present. Obstruction occurred due to extrinsic pressure instead of volvulus. This study supports the idea that mesenteric lymphangioma is an isolated disease; although it is a congenital disease, it is observed in generally healthy individuals [14], and the clinical symptoms can occur at any time.

Of all lymphangiomas, 65% occur at birth, and 90% occur during the first two years of life [3]. They can remain asymptomatic for a lifetime or can produce life-threatening complications. While small intraabdominal lymphangiomas cause only the accumulation of a small volume of peritoneal fluid, symptoms will occur if the mass increases in size [1]. They are characteristically slow-growing lesions but can also reach alarming sizes due to rapid massive growth