Journal of Medical and Health Studies
ISSN: 2710-1452
DOI: 10.32996/jmhs
Journal Homepage: www.al-kindipublisher.com/index.php/jmhs

JMHS
AL-KINDI CENTER FOR RESEARCH AND DEVELOPMENT

| RESEARCH ARTICLE

Cystic Lymphangioma of the Mesentery Complicated by Intestinal Obstruction in Children: A Case Report

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| ABSTRACT
Cystic lymphangioma is a benign tumor of malformative origin of the lymphatic vessels and lymph node tissue. This case is about a 6-year-old child who presented with a clinical symptomatology of obstruction. Surgical treatment included detorsion and resection of the mass and resection of the small bowel anastomosis. Ultrasound is a useful examination for both diagnosis and follow-up.

| KEYWORDS
Cystic lymphangioma; obstruction, tumor.

| ARTICLE INFORMATION
ACCEPTED: 20 June 2025 **PUBLISHED:** 09 July 2025 **DOI:** 10.32996/jmhs.2025.6.3.12

1. Introduction

Cystic lymphangioma is a benign tumor of malformative origin of the lymphatic vessels and lymph node tissue (1). It is a rare pediatric condition; adult forms are exceptional. Mesenteric localization is rare (2), often with a silent evolution so that its discovery occurs after a complication, such as post-traumatic rupture in the peritoneal cavity, is classic (3) or in a clinical symptomatology of intestinal obstruction. These malformations are present at birth and in early childhood. Their antenatal diagnosis is possible. They are, most often, isolated but they can be part of genetic anomalies: NOONAN syndrome, or chromosomal anomalies such as TURNER syndrome; KLINEFELTER; trisomy 21 and trisomy 18 [4,5].

2. Clinical case:

This case is about a 6-year-old child with no significant pathological history who presented to the emergency department with a clinical symptomatology of obstruction (cessation of solids and gas and bilious vomiting). Clinical examination revealed abdominal bloating and diffuse abdominal tenderness on palpation. CT scan showed a septate cystic mass and small bowel obstruction(Fig1). Surgical treatment included detorsion and resection of the entire mass (Fig2) and resection of the small bowel anastomosis(Fig3).

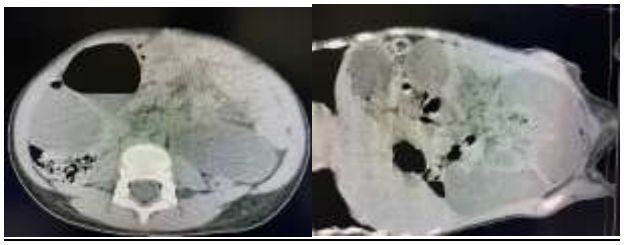

Fig1: CT images showing the cystic lymphangioma causing the intestinal obstruction

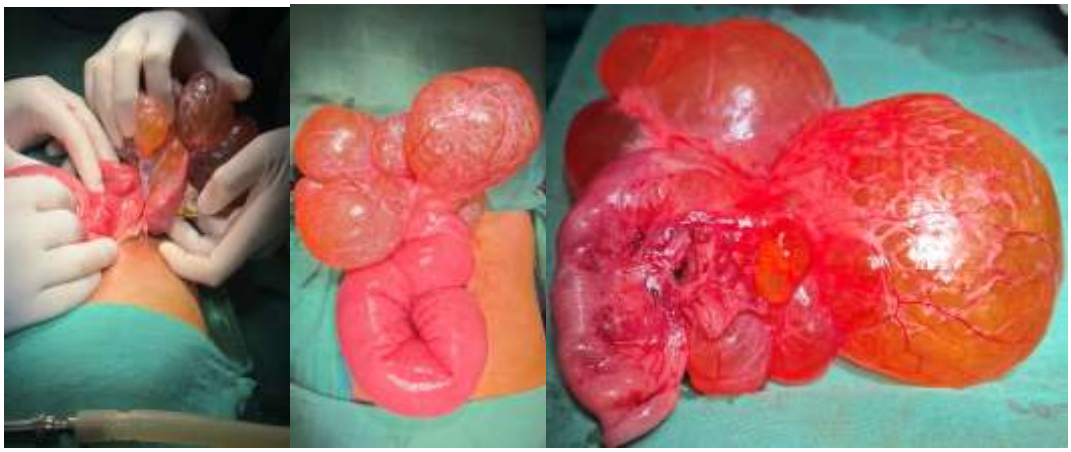

Fig2: Images of cystic lymphadenitis during surgery

Fig3: after cyst removal and intestinal anastomosis suture

3. Discussion :

Cystic lymphangioma is a rare benign tumor, most commonly seen in children. Its most common location is the cervical or axillary region (95% of cases), and less commonly the mediastinum or abdomen (5-10% of cases).

Depending on its growth rate (7), cystic lymphangioma can appear at any age, but is observed in 60% of cases before the age of 5 (6, 7, 8).

The non-specific and polymorphic clinical presentation of cystic lymphangioma is linked to the tumor volume, the location and the types of complications it causes (mechanical/infectious/hemorrhagic) [9,10]. Cystic lymphangioma can thus be revealed by palpation of an abdominal mass, abdominal pain (38% of cases), fever, hematemesis, volvulus, etc. (7,8). Finally, there is a particularly rare clinical form, in the form of cystic dissemination mimicking peritoneal carcinomatosis, called peritoneal cystic lymphangiomatosis (7).

From an etiopathogenic perspective, two theories have been suggested regarding the origin of the condition: the first suggests an acquired origin resulting from obstruction of the lymphatic vessels following inflammation, trauma, or degeneration; the second theory suggests a congenital defect in the lymphatic-venous connections leading to the isolation of a lymphatic bud that would evolve into a cyst, gradually filling with lymph (11-13).This second theory is currently the most widely accepted because it explains the insidious progression of cystic lymphangioma and the risk of recurrence if resection is incomplete. The progression is toward a gradual increase in the size of the lymphangioma, punctuated in many cases by inflammatory flare-ups. In other cases, rearrangement of the lymphangioma itself may occur, due to hemorrhage or superinfection. Finally, there are cases of spontaneous regression (13).

Macroscopically, the lesion appears whitish or translucent, arranged in a grape cluster with cysts of variable number and size, independent or communicating with each other. Histologically, the walls are reduced to an endothelial surface lined externally by connective tissue (14, 15). When the lesion is symptomatic, the clinical picture is polymorphic: abdominal swelling, abdominal pain, subocclusive syndrome or even intestinal obstruction or volvulus, more rarely peritonitis or traumatic rupture of the cyst

into the large cavity (3, 16). For our patient, the diagnosis of mesenteric cystic lymphangioma was late and was only made at the stage of complication, which is interstinal obstruction.

Ultrasound is a useful examination for both diagnosis and follow-up. It reveals a mass filled with cystic cavities with hypoechoic fluid content, of variable sizes, and with well-defined thin walls. These ultrasound features remain nonspecific.

The unprepared abdomen may be normal, or show a water-toned opacity repressing gastrointestinal lucencies. CT shows a homogeneous, hypodense tumor with thin walls, not enhanced by contrast (7).

MRI is used only as a second-line examination; it allows for a more precise study of the anatomical relationships of the lesion with neighboring structures. Cystic lymphangioma has a fluid signal: hyposignal on T1 and hypersignal on T2.

The treatment of choice is surgical and consists of complete excision of the lesion, thus avoiding recurrence. This must be complete to minimize recurrence, by laparotomy or laparoscopy, and is the most organ-conserving due to the benign nature of lymphangioma (7). During surgery, attention must be paid to lymphostasis to limit postoperative complications such as lymphocele or the development of chylous ascites (7). Nevertheless, there is a recurrence rate of 40% after incomplete resection and 17% after macroscopically complete excision (6, 7). A therapeutic alternative is aspiration of the cyst contents with or without injection of a sclerosing agent (bleomycin, Tissucol, OK-432 (picibanil), ethibloc (zein)). This treatment is useful in cases of oligo-macrocystic lesions and is easily accessible. However, the recurrence rate is high and therefore, it is preferentially used for symptomatic purposes for unresectable lesions without extensive intestinal sacrifice (6).

Diagnostic certainty is provided by the anatomopathological analysis of the tumor. Histologically, three criteria are necessary for diagnosis: 1) it is a cystic formation; 2) the septa are made up of a connective stroma with lymphoid tissue and smooth muscle; 3) the cyst is lined with a lymphatic-type endothelial lining (positivity of factor D2-40) demonstrating the vascular origin of the tumor [7,11, 17, 18]. The immunohistochemical study is positive for CD 31 factor and actin [19].

4. Conclusion :

Mesenteric cystic lymphangioma in children is a rare, progressive benign tumor requiring total and complete surgical excision to prevent complications and recurrence. The diagnosis may be suggested by a presentation of acute intestinal obstruction in a child under 5 years of age. Radiology can guide the diagnosis, and confirmation requires a pathological examination.

Funding: This research received no external funding.

Conflicts of Interest: The authors declare no conflict of interest.

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