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Short Communication

A Huge Cystic Mesenteric Lymphangioma of Small Intestine Involving Ileum: An Unusual Cause of Abdominal Pain

Verma Prashant[1], *Kumar Madhu*[1*] *and Rawat JD*[2]

[1]Department of Pathology, King George's Medical University, India

[2]Department of Pediatric Surgery, King George's Medical University, India

Abstract

Background: Lymphangioma of the small-bowel mesentery are very rare, and lymphangiomas in the jejunum or ileum are extremely rare, accounting for less than 1% of all lymphangiomas.

Methodology: We report a case of huge cystic mesenteric lymphangioma of small intestine involving ileum in a 10 years old child presenting with on &off severe pain in the abdomen. Radiological diagnosis was suggestive of enteric duplication cyst. Patient underwent for explorative laparotomy, and the mass was excised completely along with the part of small intestine.

Results: The final pathological diagnosis was cystic mesenteric lymphangioma of small intestine with multiple focal areas of involvement of mucosa, submucosa, and muscularis propria of ileum, on the basis of histopathology & immunohistochemistry.

Conclusion: Cystic lymphangiomas of small intestine are rare, should be considered as a possible cause of abdominal pain.

Keywords: Cystic lymphangioma; Mesenteric; Small intestine

Introduction

Lymphangioma is a benign vascular tumor characterized by proliferation of thin-walled lymphatic spaces. Lymphangioma of the small-bowel mesentery is rare, with an incidence of 1:250,000, representing less than 1% of all lymphangiomas. Mesenteric cystic lymphangioma is a rare benign intraabdominal anomaly with uncertain aetiology, predominantly occurring in children [1]. The intra-abdominal location is extremely rare [2,3]. We report a case of huge cystic mesenteric lymphangioma of small intestine involving the ileum in a 10 years old child presenting with on &off severe pain in the abdomen.

Materials and Methods

A 10 years old male child presented with on &off pain in the periumblical area since last one year. Severity of pain was increased for 10 days. Abdominal swelling, vomiting and altered bowel habits are not evident. No history of previous medical & surgical illness. His haemogram, kidney functional test, and serum electrolytes were within normal limit. All serum viral markers were negative. Ultrasonography of abdomen showed a well-defined complex cystic lesion with vascularity & internal septate, close proximity to small bowel loop, posteriorly abutting the anterior wall of urinary bladder. Radiological diagnosis was enteric duplication cyst. CT Scan of abdomen showed a cystically enlarged lesion which was closely abutting the small bowel loop, measuring 7 cm × 5 cm in size in the pelvis region. Findings are suggestive of mesenteric cyst. Patient underwent for explorative laparotomy and the mass was excised completely along with the part of small intestine.

Results

We received a part of ileum measuring 3.5 cm × 2.0 cm in size, attached with globular cyst measuring 7.0 cm × 6.0 cm in size (Figure 1). Both luminal ends of ileum were patent and 1.5 cm in dimension. On cutting ileal mucosa was unremarkable. Outer surface of the cyst was smooth & on cutting serous fluid came out. Multiloculated cyst with internal septae and cyst wall thickness was 0.2 cm in size. Representative sections taken for histopathology. Sections taken from ileal wall with cyst wall displaying unencapsulated cystic neoplasm and showed lymphatic channels of variable sized involving the mucosa, submucosa, and muscularis propria. These dilated thin-walled lymphatic vessels are lined by endothelial cells and supported by loose connective tissue stroma and filled with proteinaceous lymphatic fluid (Figure 2). There was no evidence of granuloma or malignancy. Provisional diagnosis was cystic lymphangima and enteric duplication cyst. On Immunohistochemical examination, CK was negative, CD31, CD34 and D2-40 were positive in lymphatic vessels (Figure 3). The final pathological diagnosis was Cystic mesenteric lymphangioma of small intestine with multiple focal areas of involvement of mucosa, submucosa, and muscularis propria of ileum, on the basis of histopathology & immunohistochemistry.

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***Corresponding author:** Madhu Kumar, Department of Pathology, King George's Medical University, Chowk, Shahmina Road, Lucknow 22603, Uttar Pradesh, India, Tel: 9005201980; E-mail: madhukumar@kgmcindia.edu

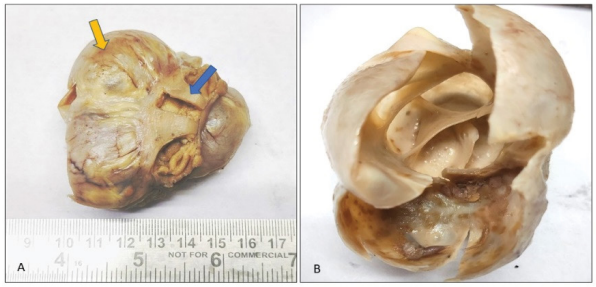

Figure 1: A-Part of ileum [Blue arrow] attached with globular cyst [Yellow arrow]. B- On cutting multiloculated cyst with internal septae [Gross Specimen].

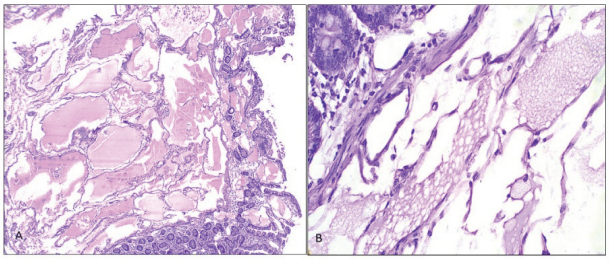

Figure 2: A-Section from ileal wall with cyst wall showed lymphatic channels of variable sized involving the mucosa& submucosa [H&E Stain, X10], B-Lymphatic vessels lined by endothelial cells [H&E Stain, X40].

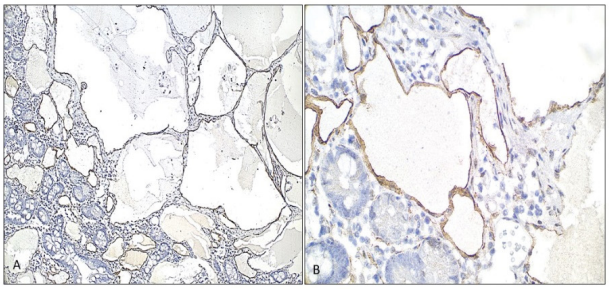

Figure 3: A-Immunohistochemical Stain CD34 positive in lymphatic vessels [IHC Stain, X10], B- Immunohistochemical Stain D2-40 were positive in lymphatic vessels [IHC Stain, X40].

Discussion

Lymphangioma are classified into three histologic types: capillary (simple), cavernous, and cystic [4]. The capillary (simple) type usually originates in the skin and consists of uniform small thin-walled lymphatic spaces. The cavernous type is composed of various sizes of dilated lymphatic spaces associated with lymphoid stroma and shows a connection with the adjacent normal lymphatic spaces. The cystic type consists of dilated lymphatic spaces of various sizes associated with collagen and smooth muscle bundles in the stroma but lacks connection to the adjacent normal lymphatic spaces. Mostly lymphangiomas (95%) occur in the neck and axillary regions; the remaining 5% is located in the mesentery, retroperitoneum, abdominal viscera, lung, and mediastinum [5,6]. Among these, lymphangioma of the small-bowel mesentery are very rare, and lymphangiomas in the jejunum or ileum are extremely rare, accounting for less than 1% of all lymphangiomas [7]. In the present case report cystic lymphangioma arises from the ileum involving the mucularis propria, submucosa and mucosa. The majorities of abdominal lymphangiomas are commonly of cystic type and occur in the mesentery, followed by the omentum, mesocolon, and retroperitoneum [8]. Mesenteric cystic lymphangioma may remain asymptomatic or present with complications depending on the size and location of the lesion. In this case report patient present with on & off lower abdominal pain with increased severity for 10 days. The most common presentation is freely mobile, non-tender abdominal mass with feature of partial small intestinal obstruction [9]. They are usually asymptomatic but can cause acute abdominal symptoms due to complications such as volvulus, bleeding, or lymphangioma rupture that require emergent surgery. Lymphangioma appears to result from congenital malformation of lymphatic vessels rather than a true lymphatic tumor [10]. The former causes sequestration of lymphatic vessels during the embryonic period [4]. However, some data suggest that inflammation, abdominal trauma, abdominal surgery, radiation, or lymphatic obstruction may play a role in the genesis of a tumor. In the present case report study had a history of on& off abdominal pain without any identifiable cause. This supports the theory of lymphangioma resulting from congenital malformation of lymphatic vessels rather than a tumor. Abdominal ultrasound examination is the diagnostic procedure of choice [7]. Laparoscopy is now the gold standard for the treatment of cystic intra-abdominal masses [7]. At follow-up of patient, he was free from disease and symptoms, currently in good health.

Conclusion

Cystic lymphangiomas are benign neoplasm, if occur in small intestine may cause acute abdominal symptoms that require urgent surgery. Therefore, it has to be kept in the differential diagnosis of the acute abdominal pain.

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