Pediatric Chylolymphatic Mesenteric Cyst: A Rare Entity

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Abstract

Mesenteric cysts are uncommon cystic lesions with variable clinical presentations. These are infrequently encountered lesions reported both in children and in adults. Chylolymphatic cysts are rare variants of mesenteric lesions and constitute 7.3%–9.5% of all abdominal cysts. Very few cases of pediatric chylolymphatic cysts have been reported in the literature. Herein, we report a case of chylolymphatic cyst in a 1½-month-old child, which is a rare entity.

Keywords: *Chylolymphatic, mesenteric cyst, pediatric*

Introduction

Mesenteric cysts are rare entities of diverse etiology with variable presentations and have surgical indication in pediatric patients.[1] A chylolymphatic cyst is a rare variant of a mesenteric cyst, and it is frequently observed in association with small bowel.[2] The mean age of children affected is 4.9 years.[1] These cysts present within the mesentery and are considered to arise from lymphatics lacking an efferent communication with the lymphatic system.[2] Although mesenteric cysts have been reported in the literature quite frequently, in the pediatric age group, chylolymphatic cysts are rarely reported with little information available regarding their presentation and complications.[1] We report a case of chylolymphatic cyst in a neonate who presented with abdominal lump and intestinal obstruction.

Case Report

A 1 and a half month old child presented with complaints of excessive crying, abdominal lump, and signs and symptoms of subacute intestinal obstruction. On ultrasonography, a loculated cystic mass was identified in the periumbilical region of approximate size 10.2 cm × 9 cm. Further, exploratory laparotomy was performed which revealed a cystic mass of approximate size 12 cm × 12 cm adjacent to ileal segment. The mass was excised along with the involved intestinal segment, and a resection anastomosis was performed. The specimen was sent for histopathological examination. Gross examination revealed a cystic mass measuring 12 cm × 11 cm × 8 cm along with a segment of intestine measuring 7 cm in length [Figure 1]. External surface was mildly congested, and cut surface showed multiloculated cyst filled with milky fluid. The thickness of cyst wall varied from 0.1 to 0.2 mm. The mucosa of the attached intestinal segment was thinned out with focal areas of congestion. Histopathological examination from the cyst wall revealed fibrous tissue along with dilated lymphatic channels lined with flattened cells [Figure 2a]. Further, on immunohistochemistry, these cells were positive for lymphatic marker D2-40 [Figure 2b], thus confirming the diagnosis of chylolymphatic cyst.

Discussion

Mesenteric cysts, first described in the 16th century, are one of the rarest abdominal tumors, and their incidence varies from 1 per 100,000 to 250,000 admissions.[3] Mesenteric cysts are most frequent in the second decade of life but are also reported to occur in the first decade as well.[2] Among these uncommon cystic lesions of the mesentery, chylolymphatic cysts are extremely rare and very few cases of pediatric chylolymphatic cysts have been reported in the literature. These cysts seem to arise in the sequestered lymphatic channels or ectopic lymphatic tissue in the small bowel mesentery and enlarge by accumulating both lymph and chyle.[3] Beahrs *et al*. classified mesenteric

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Figure 1: Photograph showing a cystic mass filled with milky fluid along with an attached intestinal segment. External surface is mildly congested

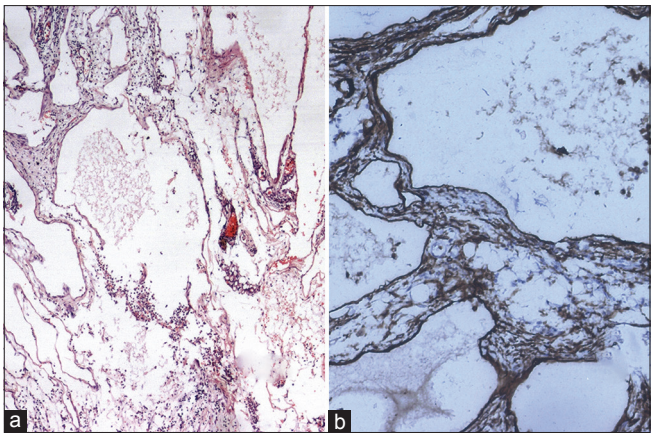

Figure 2: Photomicrograph showing (a) dilated lymphatic channels lined with flattened cells and surrounded by fibrous tissue (H and E, ×400) and (b) positivity of D2-40 in flattened cells lining the lymphatic channels (immunohistochemistry, ×400)

cysts into four groups based on etiology: embryonic or developmental; traumatic or acquired; neoplastic; and infective or degenerative.[4] Based on the contents of the cyst, the mesenteric cyst can be divided into serous, chylous, hemorrhagic, and chylolymphatic cyst.[1]

Chylolymphatic cyst is a rare variant of mesenteric cysts, comprising 7.3%–9.5% of all abdominal cysts. The cysts may be asymptomatic and may cause abdominal distension or an abdominal lump or may present with complications, such as intestinal obstruction, hemorrhage, infection, rupture of the cyst, and volvulus or obstruction of the urinary or biliary tract.[1]

Radiological investigations form an integral part of the management of these lesions.[1] Ultrasonography should be performed before surgical intervention so as to exclude other causes of an abdominal lump.[2] A plain abdominal radiograph may show a gasless, homogenous mass displacing the bowel loops around it. In patients presenting with complaints of intestinal obstruction, multiple air-fluid levels will be seen on an erect abdominal radiograph. On ultrasonography, a "fluid–fluid level" may be seen due to formation of an upper fluid level by lighter chyle over a lower fluid level of heavier lymph. Computed tomography may contribute some additional information in the form of the presence of fluid levels of differing echodensities, with an upper fatty echodensity of chyle on top of the water echodensity of lymph in a well-defined cystic lesion. However, contrast-enhanced film better shows the relationship of the bowel and other vital structures to the lesion.[1]

Surgical intervention forms the main treatment plan for chylolymphatic cysts. The different surgical approaches that can be used are marsupialization, sclerotherapy, drainage, enucleation, percutaneous aspiration, and excision of the cyst with or without resection of the involved gut. Due to high recurrence rates of chylolymphatic cyst in patients treated with marsupialization and drainage, complete excision of the cyst should be attempted whenever possible.[1] In adults, preferred mode of treatment is enucleation, in which cyst is shelled out from between the two leaves of the mesentery; however, in pediatric patients, bowel resection is frequently required along with removal of the cyst.[5]

The differential diagnosis of chylolymphatic cysts includes cystic lymphangioma, retroperitoneal cystic teratoma, renal cysts, splenic cysts, periappendiceal abscess, caseating tuberculous lymph nodes, hydatid cysts, and other cystic lesions of ovarian or enteric origin. Even lymphoma and duplication cysts may also give similar appearances.[1,2] Excision biopsy followed by histopathological examination establishes the diagnosis and helps in differentiating chylolymphatic cyst from all other cystic lesions. Cystic lymphangioma has a striking resemblance to chylolymphatic mesenteric cysts both grossly and microscopically. Some authors consider chylolymphatic mesenteric cysts to be a type of cystic lymphangioma, but the medical literature also shows some authors describing chylolymphatic cysts as a variant of mesenteric cysts.[1]

Conclusion

Chylolymphatic mesenteric cyst, although a very rare entity, should be kept in mind as one of the differential diagnoses of cystic masses of the abdomen. Surgical removal of the cyst is usually curative. Chylolymphatic cysts have well-established histopathological features that help in differentiating it from other cystic lesions of the abdomen, including cystic lymphangioma.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The

patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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