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UNCOMMON PRESENTATION- MESENTERIC CYST LYMPHANGIOMA

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ABSTRACT **Background:** Lymphangiomas are rare congenital malformations due to maldevelopment of primitive lymphatic sacs. Cystic lymphangioma present in sigmoid colon mesentery are even rare. **Case Presentation:** We describe 3 months male child presented with huge abdominal distension and multicystic lobulated mass located in the sigmoid colon mesentery measuring more than 12 cm, resected surgically. The pathological and immunohistological findings were suggestive of cystic hemangiomas. **Conclusions:** Sigmoid colon mesentery lymphangiomas are rare presentation presenting in early infancy. It requires high index of suspicion.

KEYWORDS : mesentery, cystic lymphangioma, infancy, sigmoid colon, abdominal mass.

BACKGROUND:

Lymphangioma are congenital malformation characterized by abnormal dilatation of the lymphatic spaces and most they are most commonly found in the neck and axilla (1). These are due to abnormal connection between lymphatic duct and venous system during fetal life. The presentation varies depending on the site. If the site is in gastro-intestinal system, the children have different presentations including abdominal distension, intestinal obstruction or vomiting. In children lymphangiomas are extremely rare vascular lesions and uncommon locations are retroperitoneal and intra-abdominal. Most of them are detected in early infancy and >90% are diagnosed before 2 years of age (2). Surgical excision is treatment of choice. They need to followed up for recurrences. Abdominal cystic lymphangiomas are more common in boys (3). The clinical symptoms depend on the size and location of the cysts.

Case Presentation:

A 3 month old, appropriate for gestational age male infant born at term gestation to a 29 year old primi gravida mother via normal vaginal delivery after an uncomplicated pregnancy. The mother had regular antenatal care. Perinatal laboratory evaluation findings were unremarkable. APGAR scores at delivery were 8 and 9 at 1 and 5 minutes, respectively. After an uncomplicated postnatal course, the infant was discharged from the hospital on day 2 after birth. On 11th day of life mother brought the infant back to hospital because of a noticeable swelling in the left inguinal area. Ultrasonography was confirmed the diagnosis of the left side reducible inguinal hernia for which infant underwent herniotomy. Around 3 month of age baby was readmitted with complaints of abdominal distension and excessive crying. On history evaluation, the infant was having watery loose stools, abdominal distension, abdominal tenderness and straining while passing stools for the past one week.

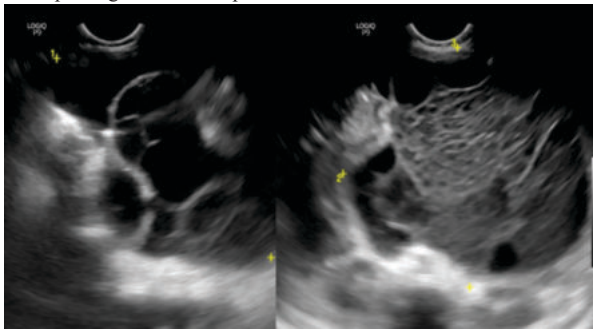

Figure 1: Abdominal USG Showing Internal Septae And Echos.

On admission the baby was irritable and hemodynamically stable with normal vitals. On physical examination, we found abdominal distension and palpable mass in lower abdominal quadrant below umbilicus with tenderness. The rest of the physical examination findings are unremarkable. Ultrasonography of abdomen shows large loculated collection around 500ml fluid in abdominal cavity showing internal septae and echoes in left hypochondriac region (Fig 1). Initially it raised suspicion of malignancy, as the collection of fluid very high in the abdominal cavity. The diagnostic abdominal paracentesis showed clear, yellow fluid and sent for analysis. Relevant laboratory findings on presentation and during hospitalization are shown in the Table 1.

In view of these reports and findings, the differential diagnosis kept were infective pathology with peritonitis, loculated ascites and malignancy. Paediatric surgeon opinion was taken and advised for surgery. Intraoperative findings showed large localized cyst in mesentery of sigmoid colon measuring about more than 13 cm. (Fig 2,3). Lymphatic cyst along with part of Sigmoid colon is resected and anastomosed. Specimen was sent for histopathological examination which revealed mesenteric lymphangioma and not showed evidence of malignancy. In view of low hemoglobin, blood transfusion was given. Antibiotics were given for total of 7 days. Baby was kept nil per mouth for 4 days and gradually restarted feeds on fifth post operative day and tolerated feeds well. Sutures were removed and baby was discharged. Now baby is active and feeding well and follow up for recurrences. The final diagnosis was mesenteric lymphangioma of sigmoid colon.

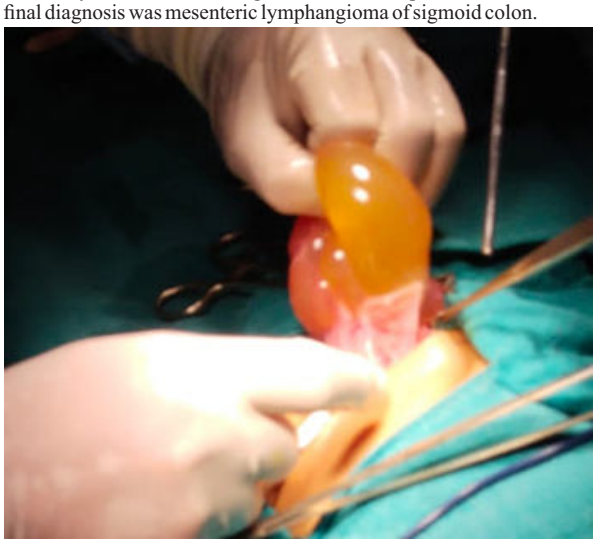

Figure 2: Large Mesenteric Cyst Attached To Sigmoid Colon.

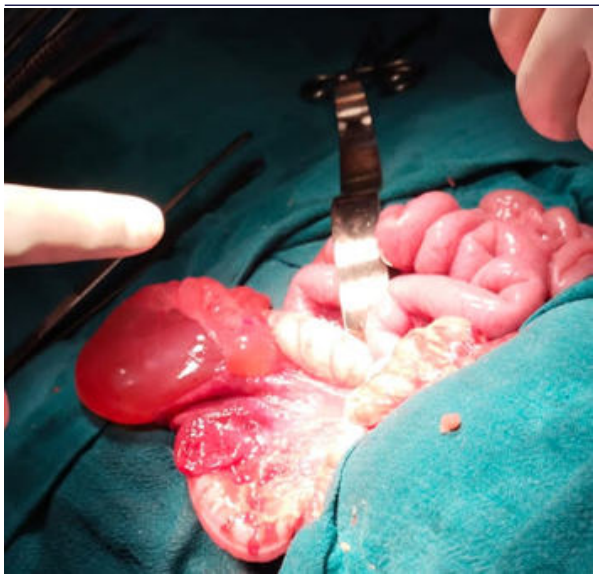

Figure 3: Large Mesenteric Cyst Attached To Sigmoid Colon Around 13 cm.

DISCUSSION:

In this case, baby presented with mixed symptoms of intestinal obstruction and infective pathology. In second week of life, baby had inguinal hernia but we couldn't find any correlation between them. They can have varied presentation starting from life threating complications to remain asymptomatic throughout the life. Kuroiwa et al. (4) reported that 67% of mesenteric cysts occurred in the small intestine and 33% in the colon. Of the 33% of cases of colonic mesenteric cysts, almost all cases were detected at the transverse or sigmoid colon. Neukirch□s classification (5) has been used to classify mesenteric cysts. However, the cause of this disease is controversial and there are two possible hypotheses: one theory identifies cysts as acquired, stating that fluid collection within a cyst is caused by the obstruction of lymph vessels due to trauma, inflammation, or pregnancy; the other theory postulates that cysts are congenital and that the fluid collection results from the proliferation of aberrant lymph tissue during embryonic development. A previous Western report has suggested that cysts are caused by sequestration or budding of lymphatic vessels, obstruction of lymphatic vessels, or ectopic lymphatic tissue.

Mesenteric cysts mostly remain asymptomatic and only come into attention when they cause abdominal fullness / distension or vague discomfort or features of intestinal obstruction. Abdominal ultrasonography usually provides working diagnosis in these patients and other investigations may be avoided though a more detailed picture can be provided by CT scan abdomen which may help in planning surgery. The recommended treatment for the intra-abdominal cystic lymphangioma is a complete surgical resection. The intervention should be performed as soon as possible because of the risk of infection, torsion, hemorrhage, or obstruction. The correct technique depends on the size and shape of the tumor, which can be resected with or without the adjacent intestinal loop or other adjacent involved organ. Prognosis is excellent when the complete resection is feasible. However, relapses may occur if vesicles or part of the tumor remain unresectable.

CONCLUSION:

Mesenteric cysts arising from the sigmoid colon are very rare, and benign in most cases; however, they should be considered as a possibility in cases of cysts occurring in the abdominal cavity. Patients who are symptomatic generally present with pain, abdominal distension, a palpable mass, and vomiting. Cysts are misdiagnosed or neglected as evening colic as some other more common abdominal conditions. Ultrasonography and CT are the most useful imaging modalities for diagnosing mesenteric cysts. Complete surgical excision is the treatment of choice. Following surgery, prognosis is excellent and recurrence is low.

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