Asian Journal of Healthy and Science
p-ISSN: 2980-4302
e-ISSN: 2980-4310
Vol. 4 No. 5, May 2025

COLONIC DUPLICATION CYST: A RARE CASE REPORT

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Abstract

Intestinal duplication cyst (IDC) is a rare congenital anomaly that can occur anywhere along the alimentary tract. The percentages were reported in the literature regarding different site as following commonly distal ileum 30% followed by esophagus, colon 5%–15%, duodenum 10%, jejunum 8%, stomach 8%, and rectum 4%–5% of all duplication anomalies. Clinical manifestations of intestinal duplications cyst are variable and determined by the type, site, and size of the duplication. Complications, such as perforation, intussusception, bowel obstruction, and volvulus can occur. Case Presentation 1-month-old male patient presented with an intra-abdominal mass since birth. Clinical examination and radiology findings suggest an intestinal duplication cyst. Laparotomy was performed and a non-communicating ascending colon duplication cyst was found. The management was achieved by resection with primary anastomosis. The non-communication cyst was located at ascending colon. Conclusion IDC can present with different clinical symptoms and it is quite difficult to diagnose in neonate patients. Early recognition and treatment can prevent further complication. In this study, the patient was done laparotomy and resection with primary anastomosis.

Keywords: Keyword: Intestinal duplication cyst, abdominal mass, ascending colon duplication, tubular duplication

INTRODUCTION

Intestinal duplication cyst (IDC) is a rare congenital anomaly that can occur anywhere along the alimentary tract. These cysts are epithelium-lined structures attached to the gastrointestinal tract, and they share the same blood supply.[1] Ectopic gastric mucosae are found in 20–30 % of these cysts. This condition had incidence of two or three cases per year in pediatric referral centers with estimated incidence thought to be 1 in 4500 births which represent its rarity, with slight male predominance. The most common site of presentation is the ileum.[2] The pathogenesis is still unclear; however, the split notochord theory has been postulated most often.[3] Clinical manifestations of intestinal duplications cyst are variable and

determined by the type, site, and size of the duplication. Abdominal symptoms may include the presence of pain, distension, palpable mass, vomiting, and bleeding. Complications, such as perforation, intussusception, bowel obstruction, and volvulus can occur.[4]

CASE REPORT

A 1-month-old male patient that was referred to our hospital for evaluation of an intra-abdominal mass. The baby was first order, born at term gestation, was appropriate for gestational age with uneventful antenatal and perinatal history and was doing well postnatally.

Upon questioning, the mother denied drug or alcohol consumption and there was no other relevant information concerning genetic malformations from both sides of the family.

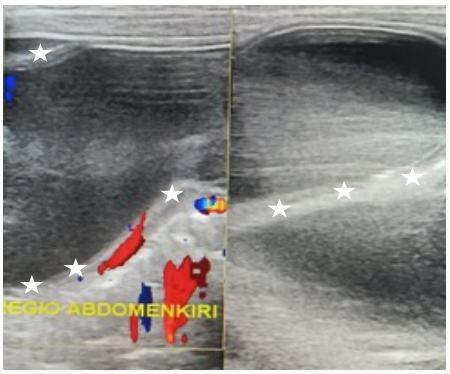

Fig. 1. Longitudinal US view of the epigastrium: thickened wall (*stars*) cystic lesion.

The parents mentioned that since birth, the infant presented abdominal distension and abdominal pain mainly after nursing manifested by constant crying and cramping. Approximately 1 weeks prior to admission he started to present occasional vomit and intolerance to oral feeding.

On physical examination the child was conscious, oriented with normocephalic, symmetrical eyes with isochoric pupils, with no signs of cardiopulmonary pathology, abdomen distended, painful to palpation, soft and depressible with increased peristalsis and no signs of peritoneal irritation or visceromegaly, normal extremities. Vital signs at hospital on admission: Temp: 36.5 °C, Respiratory rate: 40 bpm, Pulse: 136 bpm.

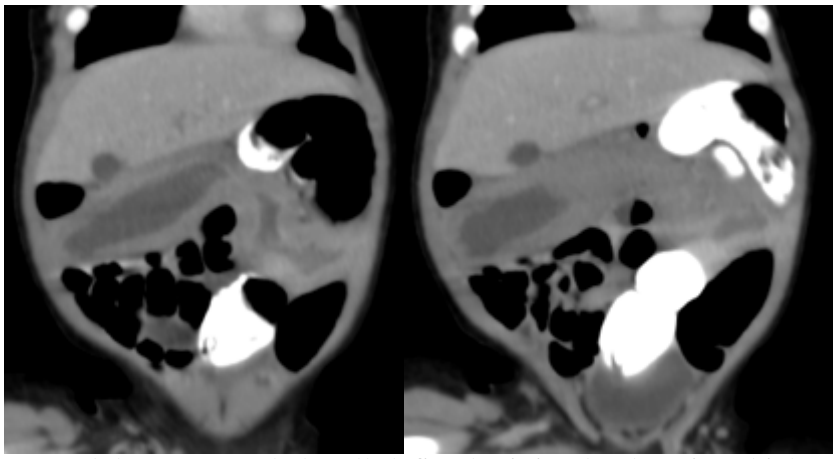

Fig 2&3. CT abdomen revealed well-defined minimal enhancing thick-walled cystic lesion.

An abdominal USG revealed well-defined cystic lesion of size 70×15×10 mm in epigastrium region displacing bowel loops peripherally with thick wall with a double line appearance and fine internal echoes. CT abdomen revealed well-defined minimal enhancing thick-walled cystic lesion of size 80×30×10 mm in left hypochondriac region with displacement of small bowel loops peripherally, no significant communication seen to adjacent bowel loops and no evidence of traversing vessels seen.

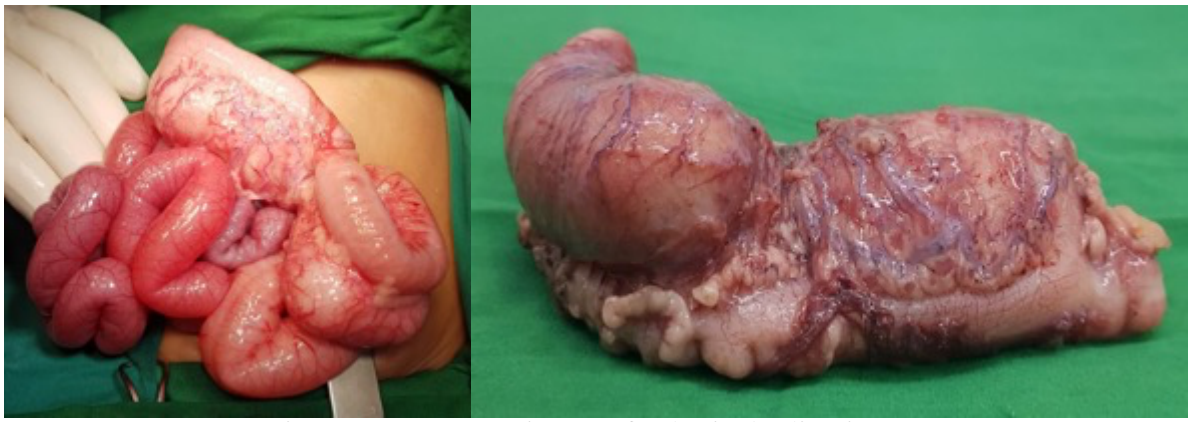

Fig 4&5. Gross speciment of colonic duplication cyst.

The patient was taken for surgery. On exploratory laparotomy, a non-communicating ascending colon duplication cyst of size 130x15x10 mm was found. Resection of the involved segment followed by ileo-colon end to end anastomosis was performed. The cyst was unilocular and containing clear fluid and mucus.

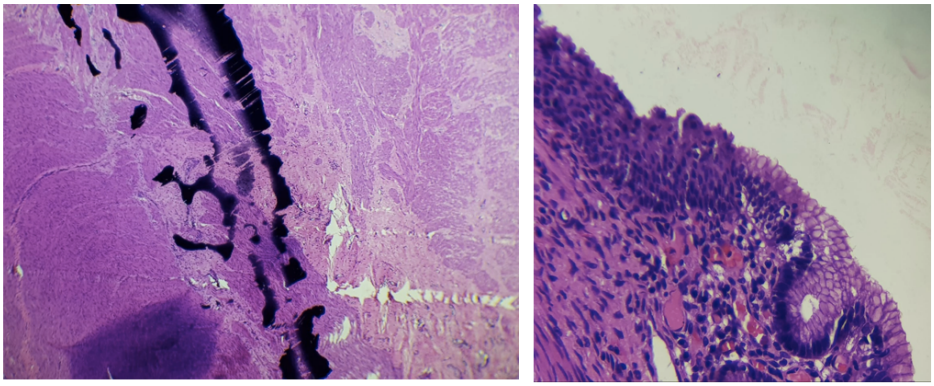

Fig 6&7. Pathologic examination of the patient

Histopathological examination revealed the resected cyst and contiguous portion of colon shared a common muscular wall, although each had its own mucosal lining, confirming it to be a duplication cyst. The patient's post- operative course was uneventful and was discharged on 6th post-operative day. He is doing well on follow-ups after three months.

RESULT AND DISCUSSION

Intestinal duplication cyst (IDC) is a rare congenital anomaly where there is an abnormal portion of intestine attached to or intrinsic with the normal bowel and can involve any part of gastrointestinal (GI) tract from the mouth to the anus.1 This condition had incidence of two or three cases per year in pediatric referral centers with estimated incidence thought to be 1 in 4500 births which represent its rarity, with slight male predominance. This anomaly most commonly affects the small bowel with around 44%.2 The percentages were reported in the literature regarding different site as following commonly distal ileum 30% followed by esophagus, colon 5%–15%, duodenum 10%, jejunum 8%, stomach 8%, and rectum 4%–5% of all duplication anomalies.2 This condition may be associated with other abnormalities, such as complete colonic duplication, gastric diverticulum, and neurenteric cysts. The most common cell type lining this condition is gastric mucosa, accounting for 50% of cases followed by intestinal, pancreatic, or respiratory epithelium.[3]

IDC was first reported in 1733 by Calder followed by Fitz in 1884 and finally popularized by Ladd in 1937 as a tubular or spherical shaped anomaly that is attached or adherent to the normal alimentary tract and share the similar phenotypic characteristics with the three following properties: (a) the cyst is surrounded by smooth muscle, (b) the cyst must contain the GI system mucosa from which it takes its own origin, and (c) the cyst must have a wall in common with the anatomic region in which it is found. [3]

There are two types of intestinal duplication cyst (IDC) in general they can be cystic which accounts of 80% of the cases, they are spherical in shape and not communicated with the bowel lumen or tubular which accounts for 20% of the cases and communicated directly with bowel lumen. Li et al. classified small IDC

depending on the blood supply into two types: (a) parallel (Type I) defined as duplication is more toward mesentery, having separate blood supply from native bowel. (b) intramesenteric (Type II) defined as duplication is cantered in mesentery having vessels from both side of mesentery.[5]

Although many of the duplications are diagnosed incidentally, most patients present with a combination of pain and/or obstructive symptoms. Clinically, intestinal duplication cyst (IDC) may present as an asymptomatic, especially in adults or it can present as occlusive symptoms (volvulus, intussusception), the classical presentation seen in children is abdominal pain, abdominal mass, bright red blood per rectum due to ulceration of ectopic gastric mucosa and less commonly it is present with intussusception, volvulus and intestinal obstruction.4 Complication could occur rarely like bleeding into the cyst, volvulus, cyst torsion, cystic rupture, infection of the cyst, urinary or biliary obstruction, or malignancy (3% sarcoma, lymphangiosarcoma) may arise.[6]

The etiology is unknown, but multiple and various theories exist regarding this condition which include the following: (a) abortive twinning theory, (b) split notochord theory, and (c) intrauterine vascular accident theory, but the most accepted explanation is that duplication of the gut occurs due to pinching off of a diverticulum during embryological development.[3]

IDC can be suspected prenatally by sonographic demonstration of an intra-abdominal cystic mass. Sonographic findings suggestive of an intestinal origin of the cyst are peristaltic muscular contractions of the cyst wall, double-layered wall, and close contact with the mesenteric border. However, in adults, multiple diagnostic tools are reported to be useful in the investigation of IDC including abdominal ultrasonography, GI endoscopy, and contrast-enhanced CT scan. Ultrasound sonography is an important tool and the most widely used for the diagnosis intestinal duplication cyst (IDC) will be seen as hypoechoic outer muscular layer with an echogenic internal mucosal layer, this was termed as "Muscular rim sign" in addition to barium studies. CT and magnetic resonance imaging scans are considered less necessary. The literature suggested Technetium-99 m pretechnetate scintigraphy to be the first test to be done in order to diagnose IDC.[1]

The treatment of choice for enteric duplication cysts is complete surgical excision with reanastomosis even if the cysts are found incidentally. The resection of the adjacent normal bowel wall is required due to the potential complications such as malignant changes, ulceration, and hemorrhage due to ectopic gastric mucosa. However, complication as short bowel syndrome can occur with resection of large tubular duplication cyst, so mucosal stripping offers alternative surgical option in these cases, eliminating the possibility of subsequent peptic ulceration or carcinogenesis.[7]

In our study, the patient diagnosis was made after ultrasound and contrast-enhanced CT scan examination. On exploratory laparotomy, a non-communicating ascending colon duplication cyst. Resection of the involved segment followed by ileo-

colon end to end anastomosis was performed as recommended by literature, and the patient recover uneventfully. Histopathological examination revealed the resected cyst and contiguous portion of colon shared a common muscular wall, although each had its own mucosal lining, confirming it to be a duplication cyst.

CONCLUSION

IDC can present with different clinical symptoms, and it is quite difficult to diagnose in neonate patients. Early recognition and treatment can prevent further complication. In our study, the patient was done laparotomy and resection with primary anastomosis.

Conclusions describe the answers to hypotheses and/or research objectives, or scientific findings obtained. The conclusion does not contain a repetition of the results and discussion, but rather a summary of the findings as expected in the objectives or hypotheses. If necessary, at the end of the conclusion can also be written things that will be done related to the next idea of the research.

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