## **Case Report**

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# **Ileal duplication cyst: a rare cause of intestinal obstruction in infants**

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#### **ABSTRACT**

Ileal duplication cyst (IDC) is a rare congenital anomaly where there is an abnormal portion of intestine attached to or intrinsic with the normal bowel. A 6-month-old male child presented with obstipation and bilious vomiting at emergency. X-ray abdomen showed multiple air fluid level suggestive of intestinal obstruction. Laparotomy was performed. Diagnosis of IDC was made and resection with primary anastomosis was done. Post-operative period was uneventful, and patient did well during 3 month follow-up checkup. IDC is a rare cause of intestinal obstruction which can present with different clinical symptoms posing a diagnostic dilemma. Diagnostic laparotomy is a suitable approach for both diagnosis and treatment to avoid delays in treatment where imaging method is unavailable for exact diagnosis.

Keywords: Exploratory laparotomy, Ileal duplication cyst, Intestinal obstruction, Resection anastomosis

#### INTRODUCTION

Ileal 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 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.<sup>2</sup> This condition was first reported in 1733 by Calder followed by Fitz in 188 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.<sup>3</sup> 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.<sup>4</sup> This anomaly most commonly affects the small bowel with around 44%. It is either asymptomatic or presents with vague symptoms mimicking other more common pathologies and is encountered unexpectedly intra-operatively. A case report of a 6 month old male infant with ileal duplication cyst who presented with acute intestinal obstruction is reported.

#### CASE REPORT

A 6 month old male child was brought to Pediatric Surgery Dept, Nil Ratan Sircar Medical college & Hospital at midnight with history of abdominal distension and obstipation for 5 days, bilious vomiting for 3 days. On examination the child was dehydrated, febrile with

tachycardia and hypotension. Abdominal examination showed grossly distended abdomen with visible intestinal peristalsis and diffuse tenderness. Bowel sounds were present. Per-rectal examination showed an empty rectum with anterior wall tenderness. Blood investigation showed elevated white cell count (25,000/ mm³) with predominance of polymorphs (75%). Plain X- ray abdomen erect showed multiple air fluid level with absent colonic gas. Emergency laparotomy via right transverse supra-umbilical incision was performed after adequate resuscitation. A multi-loculated cystic mass brownish in colour was attached to the mesenteric side of the ileum (Figure 1) which was communicating with the lumen of the ileum through a narrow pedicle.



Figure 1: Intraoperative picture of Ileal duplication cyst attached to the mesenteric side of the ileum.



Figure 2: Resection of ileal duplication cyst with primary anastomosis.

It was located 30 cm proximal to the ileocecal junction. The lumen of the cyst contained feculent smelling turbid brown fluid. Resection of the segment was done with primary anastomosis (Figure 2). A peritoneal lavage was done and abdomen closed with a peritoneal drain.

Postoperative period was uneventful, feeding resumed 4th day post operatively and patient was discharged on 5th postoperative day. Histopathologic examination reported a duplication cyst sharing a common wall structure with the ileum and advanced narrowing of the ileal lumen. There was no malignancy or dysplasia. At three months of follow up the patient was normal.

#### **DISCUSSION**

Duplication cyst is an uncommon malformation of the gastrointestinal tract which is a diagnostic and therapeutic challenge. 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.5 Approximately two-thirds of all intestinal duplications are discovered within the first 2 years of life with one-third identified in the newborn period.<sup>6</sup> 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, intussception), 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 intussception, volvulus and intestinal obstruction. 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.<sup>7</sup> 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.8 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.9 The treatment of choice for enteric duplication cysts is complete surgical excision with anastomosis 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. 10

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