

## INTESTINAL DUPLICATION CYST PRESENTING AS VOLVULUS: A Rare Case Report

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**Background:** Intestinal duplication cyst is a rare congenital anomaly occurring anywhere along the alimentary tract, from mouth to anus. They can present with numerous complications like perforation, bleeding or intestinal obstruction. We report a rare case of intestinal duplication cyst of ileum with acute intestinal obstruction due to volvulus with review of literature.

**Keywords:** Intestinal duplication cyst, Intestinal Obstruction, Volvulus

### INTRODUCTION

The first case of intestinal duplication cyst (IDC) was reported by Calder in 1733.<sup>1</sup> Clinical presentation is either in early childhood or as an incidental finding at a later age. The symptoms may range from vague abdominal pain to acute abdomen. IDC may be complicated by perforation, bleeding, fistulisation or intestinal obstruction.<sup>2</sup> Rarely, malignancy has been reported, especially in adults. In this study, we present an interesting case of IDC presenting with an unusual complication.

### CASE REPORT

A six year old boy presented with complaints of acute abdominal pain, vomiting and absolute constipation of 2 days duration. Examination revealed a pulse rate of 112/min, BP of 80/50 mm Hg, and temperature of 38.5°C. He was dehydrated. His abdomen was distended with guarding and rigidity. Plain radiograph of abdomen showed multiple air fluid levels, suggestive of acute intestinal obstruction. Laboratory data showed polymorph nuclear leucocytosis with a white cell count of 20,000/mm<sup>3</sup>. His haemoglobin level was 13.6 gm%. He was resuscitated and taken up for exploratory laparotomy. Intra-operatively, a volvulus of 20 cm long ileal loop with dilated proximal bowel loops was found (Figure 1). Surprisingly, after untwisting the bowel loop, we found a 15x10 cm long tubular structure, attached parallel to the ileal segment situated 2 feet proximal to the ileo – caecal junction (Figure1).

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It was diagnosed as an ileal duplication cyst as it had a common wall with the ileum and the blood supply was shared with the duplicated parallel segment. There were two communications of the cyst to the ileum (one proximal and the other distal) and the cyst is filled with stool which has resulted in development of volvulus. The ileal segment along with the duplication cyst was resected and end to end anastomosis was done (Figure 2). Macroscopic examination of the resected segment revealed a solitary 0.5x0.5 cm communication between normal and duplicated bowel loops on cut section (Figure 3).



Figure 1  
Per-Operative picture showing ileal duplication cyst

Both macro and microscopic examination revealed hypertrophic gastric mucosa in the duplication cyst and a common wall with normal ileal loop. Postoperative period was uneventful without any morbidity at 6 months follow up.



Figure 2  
Resected specimen of IDC



Figure 3  
Resected ileal loop with duplication showing a communication at centre (tip of haemostat) and gastric rugosity like mucosa in duplication cyst

## DISCUSSION

Intestinal duplication cyst (IDC) of the alimentary tract is a rare congenital malformation, which can occur anywhere from mouth to anus. The incidence is 1 in 4,500 births. Small bowel is commonly involved and ileum is the commonest part of the small gut to be affected by this disorder. The first case of intestinal duplication cyst was reported by Calder in 1733.<sup>1</sup> Though, various nomenclatures have been proposed, Ladd unified the different terminology to coin the term "Duplication of the alimentary tract".<sup>3</sup> They can be cystic or tubular and are usually lined by alimentary tract mucosa. The criteria for diagnosis of duplication cysts include (a) intimate attachment to the GI tract, (b) a layer of smooth muscle in the wall, and (c) an epithelial lining resembling a part of the gastro intestinal tract.<sup>4</sup> They either have common blood supply to both native bowel and IDC or an independent blood supply. The communication between native bowel and IDC is usually proximal or distal to the

adjacent bowel or rarely non-communicating type. Occasionally, the cysts are lined by heterotopic gastric issue. The etiopathogenesis of duplication cysts is still enigmatic. Various theories such as; (a) Split notochord theory, (b) Second theory suggests the failure of normal regression of embryonic diverticula, (c) Theory of median septum formation and (d) The association of complex tubular duplication of colon and rectum with urogenital anomalies due to partial or abortive twinning have been proposed.<sup>5-7</sup>

Sonography is an important tool in diagnosing IDC, which is seen as an echogenic inner margin representing mucosa and outer hypo echoic rim representing smooth muscle. This was termed as "Muscular rim sign".<sup>8</sup> Computerised tomography and magnetic resonance imaging have fared poorly, though they help in diagnosing associated anomalies. Many differential diagnoses such as mesenteric cyst, omental cyst, ovarian cyst and pancreatic pseudo cyst can be confused with IDC. In case of Small-intestine duplication cysts, the clinical presentation depends on the type, size, location, and mucosal lining of the cyst.<sup>9</sup> For example, a small duplication cyst can act as anchor point for intussusception or can result in volvulus, whereas a long tubular duplication cyst with proximal communication, which drains poorly, can cause intestinal obstruction due to retention of intestinal contents. Surgery is recommended for complicated IDC, but there is no consensus on management of asymptomatic cases due to rarity of reported cases.<sup>10</sup> The indications for surgical resection are possibility of recurrent volvulus or blind loop syndrome and a complicated IDC. As, the normal bowel and IDC have a common muscle layer, isolated resection of IDC is impractical. The optimal procedure is resection of adjacent intestine along with IDC after ligation of mesenteric vascular supply. In the present case, IDC appeared to cause acute intestinal obstruction due to volvulus. The cause of the volvulus could be due to large quantity of intestinal contents in the duplication cyst forming its epicentre. On extensive review of literature, our case was found to be the only second case of its kind with volvulus reported.<sup>11</sup> Only few cases of IDC with intestinal obstruction are reported in the literature. Another interesting finding in this case was the site of communication between normal ileum and IDC, which was through a 0.5x0.5 cm communication in the centre of the IDC. This could lead to blind loop syndrome, because of non-emptying nature of IDC through this tiny communication.

## CONCLUSIONS

IDC should be kept in the differential diagnosis of acute volvulus, especially in children and also for other intra peritoneal cists. Resection is

the treatment of choice in intestinal duplication cyst as found in the literature.<sup>12</sup> Incidentally found duplication cysts should be surgically managed to avoid future complications like bleeding and obstruction etc. Resection of IDC with parallel native bowel appears to be the optimal surgical management of choice, as was done in our case.

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#### Conflicts of interests - Nil



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