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Ileal Duplication Cyst: A Rare Cause of Intestinal Obstruction in a Neonate

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Abstract: ***Duplications of the alimentary tract are uncommon congenital anomalies that usually present duringinfancy and early childhood. The case of a neonate presenting with small bowel obstruction secondary to a duplication cyst is presented.***

Keywords: duplication cyst, intestinal obstruction, neonate, congenital anomaly

1. Introduction

Alimentary tract duplications are relatively rare congenital anomalies that may be found anywhere along the gastrointestinal tract from the mouth to the anus.[1] They have a reported incidence of 1 in 4500 in an autopsy series [2] ; there seems to be a slight preponderance in males [3] . In 1733 Calder first reported intestinal duplication [4]. Ladd introduced the term "duplication of the intestinal tract" to encompass a group of congenital anomalies that have three characteristics [5] First, they have a well-developed coat of smooth muscle; secondly, their epithelial lining represents some part of the alimentary tract and thirdly, they are attached to some part of the alimentary tract. Duplications are either cystic or tubular in shape [6]. Though they are more often observed in the small intestine, Duplication cysts can also more rarely be rectal, duodenal, gastric, and thoracoabdominal [7].They become symptomatic depending on their localization. They generally evidence signs of compression. Complications include bleeding into the cyst, obstruction, perforation, fistulization, and, rarely, malignancy [8]. A case of a neonate presenting with small bowel obstruction is presented and the management of duplication cysts is discussed.

2. Case Report

A three-day-old new-born male was referred from the paediatrics department with a history of abdominal distension and bilious vomiting on the second day following birth. On examination the abdomen was distended, rectum was empty and the neonate was sluggishly active. Routine haematological tests revealed a total leukocyte count of 17700 /mm3. X-ray abdomen (Figure 1) showed gasfilled small bowel and stomach, following which an abdominal ultrasonography was done which showed a 3.7x 2.5 cm cystic lesion with a few septae in the right hypochondrium. A decision to perform an explorative laparotomy was taken, intraoperatively a 5 x3.5x 4 cm cyst with atretic bowel was seen in the ileum (Figure 2), 15cm proximal to the ileo-caecal junction. The cyst contained straw coloured mucoid fluid 10 ml. The cyst was completely obstructing the lumen of the ileum. The segment of the ileum, mesentery along with the cyst was resected and an ileo-ileal end- to- end anastomosis was done in two layers. The post-operative course was uneventful.

The pathologic analysis of the specimen reported a bowel duplication cyst, with two separate tubular structures with lumen (Figure 3).

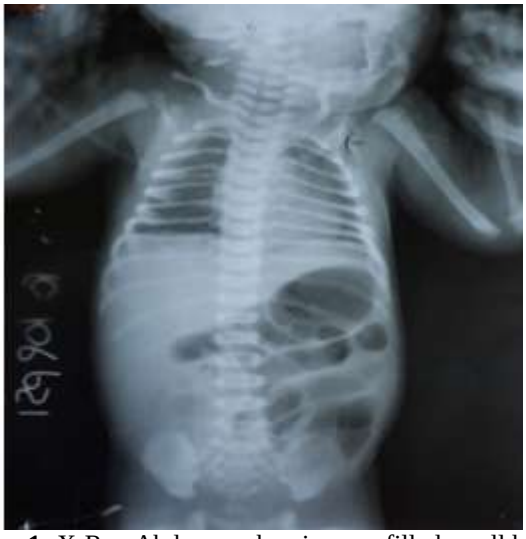

Figure 1: X-Ray Abdomen showing gas filled small bowel and stomach

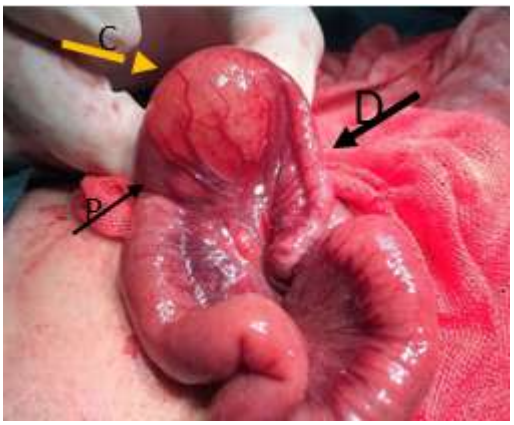

Figure 2: Intraoperative photograph showing the Duplication cyst (D) with dilated proximal (P) And collapsed distal bowel segments

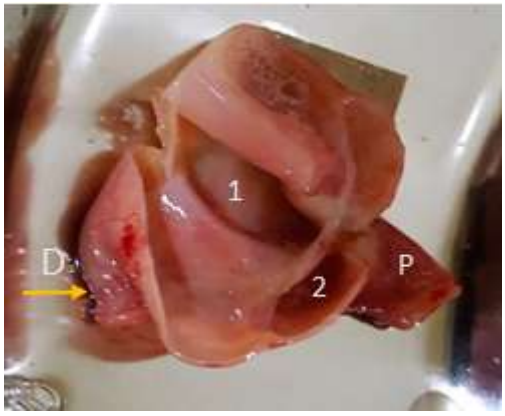

Figure 3: Specimen with two lumina (1, 2), with proximal (P), distal (D) and mucoid fluid.

3. Discussion

Duplication Cysts of the GastrointestinalSystem are the result of one or more congenital anomalies of uncertain etiology. Theories regarding etiology include the persistence of fetal bowel diverticula, vacuolization, caudal duplication, and split notochord, but the theory that has gained the most acceptance has been split notochord syndrome [9]. Duplication Cysts can be distinguished from other cystic lesions as they possess normal GastrointestinalSystem epithelium. Duplication Cysts may be located anywhere in the gastrointestinal tract from the mouth to the anus. They are seen most often in the ileum (30%) and the ileocecal region (30%). Their frequency in other areas of the gastrointestinal tract has been reported as follows: 10% in the duodenum, 8% in the stomach, 8% in the jejunum, 7% in the colon, and 5% in the rectum [10] they are generally located on the mesenteric side and most often appear within the first year of life. The classic scenario seen in children is partial intestinal obstruction and mobile abdominal mass. Depending on the localization of the cyst, rare complications such as obstruction, bleeding into the cyst, volvulus, cyst torsion, cystic rupture, infection of the cyst, urinary or biliary obstruction, or malignancy (3% sarcoma, lymphangiosarcoma) may arise. Among adults they are mostly asymptomatic and may represent an incidental finding during other intra-abdominal procedures. Rarely, adult patients may present with acute abdominal symptoms. [11]. Ultrasonography is among themost widely used imaging methods in the diagnosis of duplication cysts. CT and MRI scansare also helpful. Duplication cysts are defined on ultrasound by the presence of a hypoechoic outer muscular layer with an echogenic internal mucosal layer. Surgical excision is the recommended therapy for Duplication Cysts [12]. Duplication Cysts with different localizations and clinical symptoms may be encountered. Because there is no specific marker orimaging tool for them, we must include them among our differential diagnoses when patients become symptomatic. The diagnosis of Duplication Cysts in the neonatal and infant periods can be quite difficult because patients present with nonspecific symptoms. Due to the difficulty of diagnosis in the period of infancy, fatal cases have been reported in the literature, diagnosed too late after clinical gastrointestinal tract obstruction [10]. Duplication Cysts may be of either a spherical cystic or a tubular type. In either case, the approach to treatment is the same: removal of the Duplication Cysts and anastomosis. The cystic-type Duplication Cysts we found in the ileum fit the classical definition and was exerting pressure on the lumen. We performed a resection and anastomosis as recommended by the literature, and we observed a complete recovery in the patient.

4. Conclusion

In conclusion, Duplication Cysts may be the etiology in neonatal congenital intestinal obstruction, albeit rarely. Delays may occur in diagnosis due to the incomplete obstruction which occurs in most cases. We are of the opinion that Duplication Cysts is an important provisional diagnosis for patients with recurrent complaints of intestinal obstruction.

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