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Case Report

Ileo-cecal Sphincteric Duplication-Total Cyst Excision with Ileo-cecal Valve Preservation

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Abstract

We report a case of prenatally diagnosed and postnatally confirmed ileal duplication cyst situated close to ileo-cecal valve. The size of the cyst was smaller than 5 cm diameter and an elective admission with laparoscopic excision was planned at 3 months of age to avoid any anesthetic or hospitalization related complications. However, the infant presented in emergency with massive lower gastrointestinal bleeding requiring transfusion. At exploration, a terminal ileal duplication cyst in the ileocecal sphincteric area was found which was excised without resection of bowel or the ileocecal valve. Histopathological examination confirmed ectopic gastric mucosa with ulceration in the transitional area. The case is very interesting and the debate about the conservation or the resection of the ileocecal valve in ileocecal cyst duplication has been presented with review of relevant literature.

Keywords: Infant; Ileal; Duplication; Cyst; Ectopic gastric mucosa; Gastrointestinal bleeding; Ileo-cecal valve; Preservation

Introduction

Ileocecal valve appears to be a key vital structure in children in general and neonates and infants in particular. It plays a fundamental role in the growth and development of the body, regulates intestinal transit and acts as barrier to delay passage of intraluminal small bowel material increasing their absorption [1]. It is a strong anti-reflux mechanism to prevent reflux from the cecum into the terminal ileum acting as an important barrier water shade and keeping small bowel contents free from gross contamination and bacterial overgrowth [2]. We wish to report a unique case of an infant with ileo-sphinteric duplication cyst very close to the ileocecal valve in whom we could excise the whole cyst with preservation of ileocecal valve and avoided resection and anastomosis.

Case Presentation

A term baby girl weighing 3540 gm was born by vaginal delivery. Prenatal scans detected an abdominal cyst which was confirmed on postnatal scan measuring 24.2 X 25.6 mm and having double lines suggestive of a cystic duplication cyst (Figure 1). She was sent home at birth and an elective laparoscopic excision was planned at 3 months of age.

At 8 weeks of age she presented with significant bright fresh painless intermittent lower gastro-intestinal bleeding. On arrival she was pale; abdomen was soft with a palpable mobile mass in right lower quadrant. Her haemoglobin was 80 G/L.

After initial resuscitation and stabilisation, she underwent exploration through periumbilical incision which showed terminal ileal sphincteric duplication cyst with some blood in the lumen of bowel. The ileo-cecal area and cecum was very mobile. Cyst was aspirated. Ileo-cecal area with collapsed cyst was exteriorised. It was attached to ileal sphincter 4 cm away from the ileo-cecal valve via a small stalk on the mesenteric side. Both leaves of the mesentery were opened by displacing the vessels and the cyst wall was mobilised. Cyst was excised totally with closure of the mesenteric defect.

Post-operative period was uneventful and was discharged home on 3rd post-operative day. Histology confirmed ileal duplication cyst with ectopic gastric mucosa and ulceration of adjacent mucosa. At one year follow up, patient was thriving well and asymptomatic and was discharged.

Discussion

Congenital gastrointestinal duplications are rare, with an incidence of 1 in 4,500 neonatal autopsies or 1 in 18,000 live births. Although the small bowel duplications are the most common of all one third are situated in the jejuno-ileum (30%) and one third are situated close to ileocecal valve (30%) with the half of all lesions may contain ectopic gastric mucosa [3,4]. Over 90% are cystic and the remaining one may be tubular.

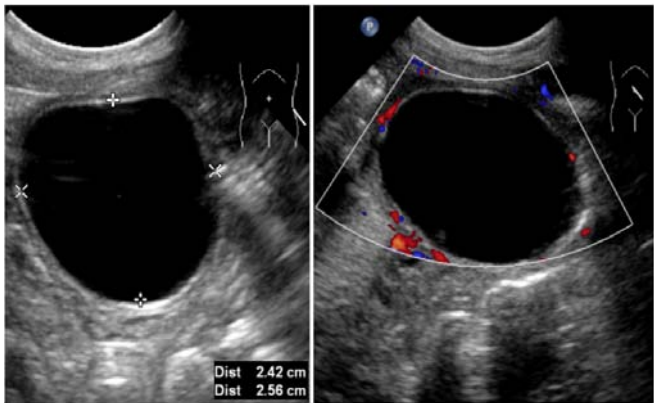

Figure 1: Postnatal ultrasound scan and colour Doppler- Note double lining of enhancing intestinal mucosa and simple cyst less than 5 cm in diameter.

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A duplication cyst diagnosed by prenatal and postnatal scans may create problems even when they are smaller than 5 cm in diameter, without having ectopic gastric mucosa and even before birth [5-7]. An anatomically intact and functionally preserved ileocecal valve is essential for better growth and nutrition in the neonates and infants and this fact has been taken into consideration at large neonatal and paediatric surgical centers with special interest in bowel preservation and lengthening rather than going on the transplant option.

Terminal ileum and ileocecal region has high incidence of congenital and acquired pathology in neonates and infants in the form of ileal atresia, congenital internal hernia, midgut malrotation, necrotizing enterocolitis, intussusception , etc. and a plea has been made recently for preservation of the ileocecal valve to retain its important functions should be considered in cases of benign lesions.

A simple abdominal cyst less than 5 cm diameter diagnosed prenatally and confirmed by postnatal scans as duplication cyst should be taken seriously. It can become symptomatic and present with life threatening emergency even before the planned excision and could be fatal.

Traditionally segmental bowel resection and anastomosis is performed which would sacrifice ileo-cecal sphincter and valve. Total excision of the cyst with preservation of ileo-cecal sphincter and valve is crucial in infants and it is feasible, safe and effective through small peri-umbilical incision.

Resection of duplication alone without the resection of the ileum or the ileocecal valve is the ideal treatment, and whenever possible laparoscopic resection is advised in uncomplicated cases. Recently a large series defending the conservation of the ileocecal valve has been published and traditional resection of the ileum with the ileocecal valve has been questioned [8].

Although considered to be congenital and in this era of prenatal diagnosis, most are diagnosed on antenatal scans but few may be missed and present later in life with catastrophic complications. Apparently benign with excellent prognosis when treated early and appropriately, such lesions may lead to lethal complications and few cases of sudden deaths have been reported even in the twenty first century [9-12].

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