



Cecocolic Intussusception in an Infant with Cecal Duplication Cyst: Case Report with Review of Literature

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Abstract

Background: Intussusception is a quite common gastrointestinal emergency in children. In nearly 2% to 12% of patients, a lead point can be Meckel diverticulum, mesenteric lymph nodes, intestinal tumors (NHL), polyps, ganglioneuroma, hamartomas or mesenteric or duplication cysts. The latter are rare congenital malformations of the gastrointestinal tract. These cysts are commonly found in the ileum. Colonic duplications occur in 3% to 20% of cases, whereas cecal duplications are extremely rare with a reported incidence of 0.4% of all alimentary tract duplications.

Methods: We report the case of a 2-month-old female who was referred to our hospital with abdominal pain and rectal bleeding. Diagnostic imaging included Ultrasonography, plain abdominal radiography and contrast enema. The indication to surgery was a suspected intussusception whose lead point was a duplication of the cecum. The correct diagnosis was established only with surgical assessment. Enucleation of the cyst was not possible due to its anatomical localization and a common muscular wall with cecum. Hence a complete mucosectomy of the cyst was performed.

Results: The post-operative period was uneventful and the infant was discharged on the 4th p.o day. Follow up after 6 month was regular.

Conclusion: Cecal duplication is a really rare congenital condition. About 80% of these cases are detected in the first 2 years of life as a result of an acute intestinal obstruction. The correct diagnosis is generally subsequent surgical exploration. Mucosectomy is a surgical choice which allows preserving the ileo-cecal valve without resecting the cecum.

Keywords: Duplication cyst; Cecum; Intussusception; Mucosectomy

Introduction

Intestinal duplications are rare congenital anomalies and most of them are detected in the first 2 years of life or antenatally. The timing and clinical presentations depend on their location, size and the presence of gastric mucosa. The symptoms are unspecific and vary from minor digestive problems to intestinal obstruction, bleeding and perforation. Prenatal diagnosis may prevent the development of acute clinical conditions and allows early treatment. Surgical management depends on the location of the duplications in relation to adjacent bowel.

Patients and Methods

A 2-month-old female was referred to our hospital with abdominal pain and rectal bleeding. Once arrived to the pediatric emergency department, the infant began vomiting bile-stained fluid. Clinical findings were normal; abdominal tenderness and rebound were observed in the right lower quadrant. Biochemical analysis revealed the following: haemoglobin level 12.7 g/dl; haematocrit 36.9%; white blood cell count 18.590/L; PCR<1.

Ultrasonography showed a “target sign” on the right side of the abdomen associated with an anechogenic pseudocystic image. Subsequently, with the aim to solve the leading point of the intussusception, a contrast enema was done without success. During the colon series the passage of contrast could be observed from the left to the right colon without passing through the small bowel. Thereafter the operation was performed. The patient underwent a laparoscopic exploration that identified the intussusception area. The umbilical incision was enlarged allowing to perform an abdominal exploration and to manipulate the long intussusception until it was completely resolved. The leading point resulted to be a duplication of cecum located between the ileo-cecal valve and cecum. With the aim of avoiding a resection of colon with the ileo-cecal valve, the duplication was incised, its fluid was aspirated and a complete mucosectomy was done respecting the integrity of the

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Received Date: 27 Jun 2018

Accepted Date: 11 Dec 2018

Published Date: 14 Dec 2018

Citation:

De Biagi L, Ratta A, Federici S. Cecocolic Intussusception in an Infant with Cecal Duplication Cyst: Case Report with Review of Literature. *World J Surg Surgical Res.* 2018; 1: 1081.

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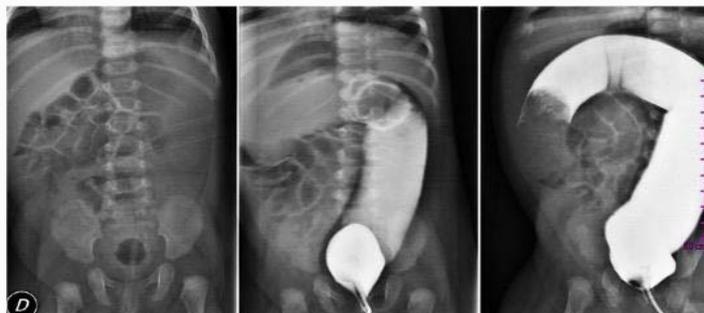


Figure 1: Barium enema series show the intestinal intussusception.



Figure 2: Intraoperative photographs show the opening of the cyst and its mucosectomy.

common wall with cecum. The post-operative period was uneventful and feeding was started on the second post-operative day. Ectopic gastric mucosa was observed at the histological exam.

On long term follow up (12 months) stools were regular.

Discussion

Duplications of the gastrointestinal tract are uncommon congenital anomalies. Ladd applied the term to congenital lesions having three characteristics: (1) the presence of a well-developed coat of smooth muscle, (2) an epithelial lining representing some type of intestinal mucosa and (3) intimate anatomic association with some portion of the gastrointestinal tract. Cysts are defined as spherical or tubular structures (the latter being less commonly found in the small intestine than the spherical type) [1-3].

There are 3 categories for classification: (1) localized duplications (2) duplications associated with spinal cord defects and vertebral malformations and (3) duplications of the colon.

Multiple duplication cysts are found in 10% to 20% of cases [1].

Children with enteric duplications seen to have a high incidence of other associated anomalies. Urinary tract anomalies have been reported with midgut and hindgut malformations; and in one series, all patients with multiple duplications also had a skeletal or urinary tract anomaly.

The etiology of intestinal duplications remains unclear. The persistence of embryonic diverticulae during the development of the gastrointestinal tract, intrauterine vascular accidents, and recanalization and fusion of embryonic longitudinal folds are some of the etiological hypothesis that has been proposed [1,4,5].

Duplications of the alimentary tract may present at any age, but 80% present in first 2 years of life; the ileum is the most common

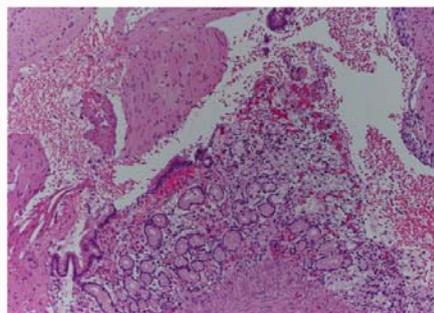


Figure 3: Histological exam shows ectopic gastric tissue.

site of involvement and only 13% of all alimentary duplications are colonic [6]. Most cases develop within the first 3 months of life and a palpable mass is the most frequent mode of presentation, seen in 50% of these infants. Duplication cysts of the cecum are very rare. In an 18-year review, Grosfeld et al. [6] could find only one case of cecal duplication in a neonate; barium study revealed a palpable abdominal mass with a filling defect. Oudshoorn and Heij [7] have re-viewed 362 cases of duplication cysts reported in the literature and found only 16 cases of cecal duplications.

Symptoms of intestinal duplication cysts vary according to the size, morphology and location of the cysts. Common symptoms include acute intestinal obstruction, vomiting, recurrent abdominal pain, recurrent gastrointestinal bleeding, constipation or an incidental detection. 30% of the patients have ectopic gastrointestinal mucosa that may present as gastrointestinal bleeding or even perforation [1-3]. Ultrasonography and barium studies are helpful imaging modalities for the diagnosis of duplication cyst [8,9]. Ultrasound shows an echogenic inner mucosal layer with a hyperechogenic outer muscle layer. Some enteric duplication cysts contain gastric mucosa

and may be demonstrated with a 99T cm-pertechnetate scintigraphy [10,11]. Although rare, duplication cysts are an important differential diagnosis to consider in children, especially in neonates who present with a palpable abdominal mass such as mesenteric cyst, ovarian cyst, pancreatic cyst or choledochal cyst.

Various surgical procedures have been proposed in the treatment of cecal duplication cysts [12]. Complete excision should be performed when possible. Sometimes complete excision is not achievable and end-to-end anastomosis after segmental resection of bowel with duplication cysts is the preferred choice of treatment [1,5] even if removal of the ileocecal valve can lead to a reduced intestinal transit and to an impairment of absorption clinically evident with diarrhea, electrolyte imbalance and finally malnutrition [13,14].

In the case reported a mucosectomy was preferred in order to avoid the resection of the ileo-cecal valve and this surgical choice has demonstrated to be safe and easy to perform.

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