Caecal Duplication Cyst Presenting with Intussusception: A Case Report

Maria Munawar, Laraib Amjad and Muhammad Bilal Mirza

Department of Paediatric Surgery, University of Child Health Sciences, The Children's Hospital, Lahore, Pakistan

ABSTRACT

Caecal duplication is a rare entity and usually presents with intestinal obstruction. Even more unusual is its presentation as a lead point for intussusception. A nine-month boy presented with signs of intestinal obstruction. On examination, a mucosal mass was found in the rectum. At surgery, a caecocolic intussusception was found. After reduction, a caecal mass was appreciated as a lead point. Limited haemicolectomy was performed, and ileostomy was formed. Histopathology confirmed it as a caecal duplication cyst.

Caecal duplication cyst as a lead point for caecocolic intussusception is a rarity. It should be considered in the differentials of caecal masses in children.

Key Words: *Caecum, Duplication cyst, Intussusception, Rectum, Prolapse*.

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INTRODUCTION

Enteric duplication cysts are rare congenital anomalies that can occur anywhere from the mouth to the anus and are more common in males under the age of two years, with an incidence of 1 in 4,500 live births.[1] The most prevalent location for these cysts is the small bowel, particularly the ileum, with rarer occurrences in the oesophagus or colon.[2] A duplication cyst of the caecum is an even rarer entity, with only 43 cases reported in the literature so far.[3]

Intussusception is a common surgical emergency in infancy and typically occurs without a lead point. One of the significant complications of enteric duplication cysts is intussusception, where the cyst acts as a lead point.[4] Patients often present with symptoms such as abdominal pain, a palpable abdominal mass, vomiting, and, in rare cases, prolapsed intussusception.[4]

We report a rare case of a nine-month boy with a caecal duplication cyst causing intussusception, which, upon examination, was found to have prolapsed into the anus.

CASE REPORT

A nine-month term male baby presented to a paediatrician at a private clinic with complaints of loose motion, generalised abdominal pain, and vomiting for two days, as well as bleeding per rectum for one day.

Correspondence to: Dr. Maria Munawar, Department of Paediatric Surgery, University of Child Health Sciences, The Children's Hospital, Lahore, Pakistan
E-mail: munawarmaria11@gmail.com

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Treatment for dysentery was initiated. The following day, the baby developed bilious vomiting. While changing the diaper, the mother noticed an open anus with a pinkish mass visible within the rectum, although it was not protruding from the anal verge. The patient was then referred to a paediatric surgeon.

On examination, the baby's pulse rate, blood pressure, and respiratory rate were unremarkable. The abdomen was soft and distended with palpable bowel loops, and a sausage-shaped mass was palpable in the left haemi-abdomen. A digital rectal examination revealed the pinkish mass as a prolapsed gut into the rectum, with the finger able to pass between the mass and the rectal wall. The anus remained open due to the mass, which was easily visible within the lumen of the anus and rectum but did not protrude from the anal verge. Ultrasonography of the abdomen showed a doughnut sign. Laboratory investigations, including complete blood count (CBC) and serum electrolytes, were within normal limits.

The patient was optimised for surgery and underwent an exploratory laparotomy. Intraoperatively, a caecocolic intussusception was found, prolapsing into the rectum (Figure 1). The intussusception was reduced, but during reduction, a full-thickness tear occurred in the sigmoid colon which was repaired, and a serosal tear occurred in the transverse colon. A firm mass was palpable in the wall of the caecum after reduction (Figure 2), which was resected, and a limited right haemicolectomy was performed. An ileostomy and mucous fistula were created in the same incision. Biopsy samples were taken from the caecum, ascending colon, and lymph nodes. The biopsy report suggested a caecal duplication cyst. The stoma started functioning on the second postoperative day, and oral intake was resumed. The patient was discharged on the third postoperative day and had an uneventful recovery. The stoma was reversed after two months, and the baby is doing well on follow-up.

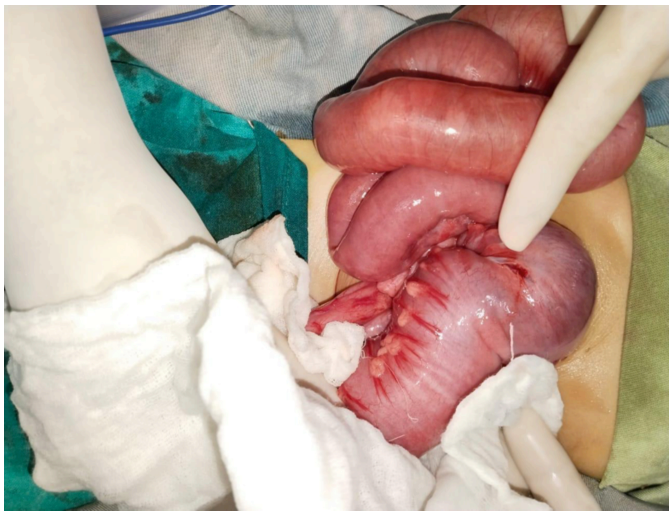

Figure 1: Intraoperative photograph showing a caeco-colic intussusception.

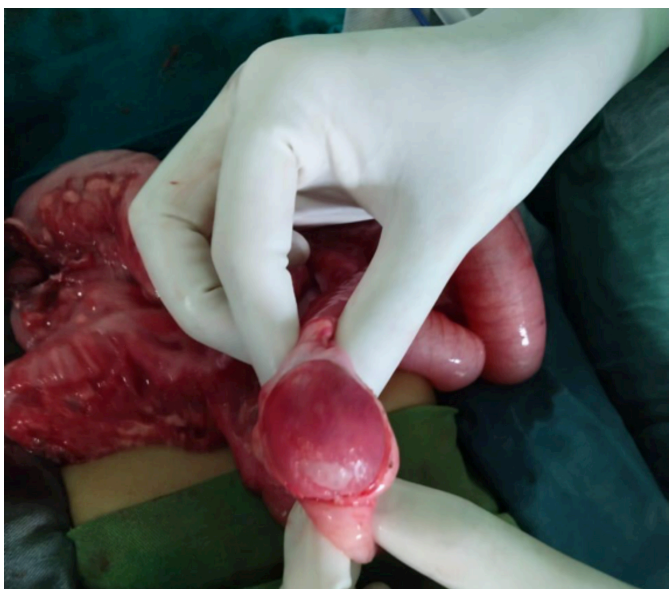

Figure 2: Intraoperative photograph showing a caecal mass within the wall of the caecum, the caecum has been opened to demonstrate the intramural mass.

DISCUSSION

Duplication cysts are among the rarest congenital anomalies of the alimentary tract.[3] They mostly present within the first two years of life, with the majority of cases occurring between 5 and 8 months.[4] The ileum is the most common site of occurrence, while the caecum is an exceedingly rare site for duplication cysts. More than two-thirds of alimentary tract duplications are cystic and non-communicating, as observed in this case.[2]

Over 95% of intussusceptions are idiopathic, with only 5% having a pathological lead point (PLP). The incidence of PLP increases in older children and can include Meckel's diverticulum, polyps, mesenteric lymph nodes, duplication cysts, and gastrointestinal tumours.[5] Our case is rare because despite the patient being a small infant, we found the caecal duplication cyst acting as a PLP for caecocolic intussusception.

Common presentations of duplication cysts include abdominal pain, abdominal distention, vomiting, and per-rectal bleeding. They can occasionally progress to intestinal obstruction or perforation, often due to the twist of the cyst, compression on the adjacent bowel, or development of intussusception.[6] In this case, the patient presented with abdominal pain, distension, and rectal bleeding, with a mucosal mass appearing in the rectum due to prolapse of the intussusception.

The diagnosis of intussusception was made clinically, with the patient presenting with a prolapsed rectal mass and a palpable sausage-shaped mass in the left haemi-abdomen. The presence of currant jelly stool raised suspicion of intussusception. Ultrasonography, the imaging modality of choice for diagnosing duplication cysts, confirmed the diagnosis.[1] However, preoperative diagnosis of the caecal duplication cyst was not possible due to the development of caecocolic intussusception. Even during surgery, the diagnosis was initially suspected to be a caecal mass; however, the final diagnosis was confirmed through histopathological evaluation.

Management of intussusception is a surgical emergency. In early-presenting cases, pneumatic or hydrostatic reduction under fluoroscopy or ultrasound guidance is preferred.[7] In late-presenting cases or when the intussusception has prolapsed, as in this case, open or laparoscopic surgery is more effective. Resection is necessary in cases of gangrenous gut or the presence of a PLP. While resection and intestinal anastomosis are preferred, a temporary stoma may be required under certain conditions. In this case, the presence of a full-thickness tear in the sigmoid colon and a large serosal tear in the transverse colon necessitated a temporary stoma following a limited haemicolectomy for the caecal mass.

In conclusion, caecal duplication cysts are very rare congenital anomalies. Their presentation with caecocolic intussusception prolapsing into the rectum is exceedingly rare. Caecal duplication should be considered in the differential diagnosis of caecal masses, especially those acting as PLPs for intussusception.

PATIENT'S CONSENT:

Informed consent was obtained from the patient's attendants to publish the clinical details and images included in this case report.

COMPETING INTEREST:

The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:

MM: Conception, design of the work, and analysis and interpretation of data.
LA: Revising and refining the content.
MBM: Supervision.
All authors approved the final version of the manuscript to be published.

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