Cecal Duplication Cysts Mimicking Intussusception in Female: A Case Report

Khalid A
Department of Pathology, Sahiwal Medical College Sahiwal, University of Health Sciences Lahore, Pakistan

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1. Abstract
Enteric duplication cysts are a rare congenital anomaly, occurring anywhere throughout the length of the intestine. More than 80% cases occur in the first two years of life. However, asymptomatic individuals may also present in adult life. Cecal duplication cysts occur only in 0.4% of all the gastrointestinal tract duplication cysts. The wall of cyst contains an inner mucosal and an outer muscular layer. Most common clinical presentations include palpable abdominal mass, pain mimicking appendicitis, bleeding per-rectum, abdominal distension, and intussusception with acute or chronic intestinal obstruction. The delay in the diagnosis can lead to high morbidity. We present a case of cecal duplication cyst in a female presenting with pain and vomiting since one day.

2. Introduction
Duplication cysts in the intestine are a rare congenital anomaly, most commonly occurring in the ileum, 13% in the colon and rarely from the cecum. Out of all of them, two third cases occur in the first two years [1]. However, some individuals have no symptoms and usually present in adult life. Until now, more than two dozen cases have been reported in the literature, mostly occurring in the first two years of life. Their incidence is one in 4500. Patients can variably present depending upon the size, location, underlying structures and the presence of ectopic gastric tissue. The wall of cyst contains an inner mucosal, echogenic layer (on US) and an outer hypoechoic (On ultrasonography) muscular layer. Most common clinical presentations include palpable abdominal mass, pain mimicking appendicitis, bleeding per-rectum, abdominal distension, and intussusception with acute or chronic intestinal obstruction. The delay in the diagnosis can lead to high morbidity [2]. The reported cases are around 50 to the best of our knowledge [3]. We present a case of cecal duplication cyst in female presented to our teaching hospital emergency department (tertiary care center) and it is in accordance with the SCARE guidelines. Surgical management is the best option with excellent prognosis.

3. Case Report
A 42 years old female patient, Rasheeda, presented to us in emergency on 15th June, 2022, with presenting compliant of generalized abdominal pain and vomiting from 1 day.

Pain was sudden in onset, colicky, progressive in nature, non-radiating, non-shifting, lasted for one day, aggravated on eating and lying flat with no specific relieving factor, not associated with constipation or abdominal distension but with sudden onset and multiple episodes of vomiting, yellowish in color, containing food particles, equals 4-5 cups in each episode and non-projectile. There was no significant medical history of any inherited or congenital condition. However, she had her cesarean section 9 years back. The family history was positive for diabetes mellitus in her mother and elder sister. The gynecological history depicts a regular menstrual cycle 7/28 with age of menarche at 15-year.

On clinical examination, pulse 119beats/min, B.P. 110/70 Oral Temperature 100F, SpO2 98%, Respiratory rate 32breaths/min. On abdominal examination, abdomen was distended, umbilicus inverted and central. There was a visible cesarean scar mark in lower abdomen. There was no visible pulsation or prominent vein. Fullness seen in right hypochondrium. On palpation, abdomen was tense as a whole; a vague tender mass was palpable in the left lumbar region, firm in consistency with ill-defined borders and not moving with respiration. No other viscera was palpable. Per-vaginal and per-rectal examination were unremarkable. Percussion note was resonant all over the abdomen except left lumbar region where it was dull. On auscultation, bowel sounds were audible. Remaining physical examination was unremarkable.
Lab reports show complete blood parameters with decreased Hb, leukocytosis, lymphocytopenia and raised platelets. Serum electrolytes show hypokalemia with normal Na, Cl, and Mg levels. Hep B and Hep C serology was negative. Detailed LFT’s were done and they were within normal range. Renal Function tests were also within normal limits. Serum HCG and remaining lab parameters were unremarkable.

Ultrasound shows mild free fluid with internal debrinous echoic area noted in lower abdomen and pelvis along with fatty hepatomegaly. Contrast enhanced CT scan of the abdomen and pelvis shows twisted appearance of the gut and mesentery in right sub-hepatic region. However, small gut volvulus could not be ruled out. Well defined non-enhancing cystic area also reported in the left lumber region likely representing a mesenteric cyst.

Patient was prepared for surgery and after taking proper consent and under aseptic measures, abdomen was opened with a midline incision and peritoneum reached. A cystic swelling of 10cm×10cm was found to be originating from cecum (as shown in above figure) with twisted and gangrenous gut along with 500ml purulent fluid in peritoneal cavity. Right hemicolectomy was done. Ileo-colic anastomosis was made and abdomen was washed with 4L of Normal Saline. Abdomen closed in reverse order after placing drain in the pelvis.

Antibiotics, fluids and analgesics were given in post-operative period. Nasogastric tube removed after 1 day and patient discharged in satisfactory condition at 5th post-operative day.

4. Discussion

Congenital cysts include chylous, lymphatic, dermoid, enterogenous and urogenital. More than 80% cases occur in the first two years of life. They can anywhere in the alimentary tract on the mesenteric side. Colonic duplication cysts are rare (13%) and cecal duplication cysts are even rarer [6]. Coider reported the first case in 1733 and named as Duplication cyst by Fitz and used by William E Lad in 1930’s. It was classified by Gross in 1950’s.

Based on their location, terminal ileum and ileocecal junction were the most common (53%) with colonic duplication second to it (13%). However, cecal duplication cysts remains the least common with incidence of 0.4% only. Females are more common than males [7]. USG remains the ideal investigation with >95% specificity and PPV 85-100%. Contrast enhanced CT scan is not required in all cases. While, Technetium pertechnetate scan helps in detecting the gastric mucosa present in ectopic gastric mucosa in duplication cysts [8]. Literature reports around 50 cases and they were reviewed along with our findings. Morphologically, cysts can be of tubular and spherical variety. Their exact cause is not known. Possible causes are defective recanalization, fusion of embryonal longitudinal folds, persistant diverticulae of embryonic life and uterine vascular anomalies [7]. In previous studies, four (10%) patients had associated anomalies. Two with colonic duplication, one with duplication of duodenum and the remaining with thoracic meningocele and vestibular fistula. However, no anomaly was found in our patient. Literature reports show that 67% cysts were present in the first year of life. While, only 5% cases were diagnosed antenatally. Similarly, females were more commonly affected (67%) than males. Symptoms depends upon their location, size and morphology [9]. Our review shows vomiting as the

Figure 1: shows duplication cysts of cecum (blue arrows).

Figure 2: shows cecum (white arrows) and duplication cysts (blue arrows).
most common clinical presentation (57%). Palpable mass in 45% patients, 41% with abdominal distension, 34% with abdominal pain, and hematochezia in 18% of patients. Only 14% had compliant of constipation [6, 8, 10]. Additionally, they can also present with intestinal obstruction as cyst can lead to volvulus or intussusception as seen in 10 patients. However, malignancy can occur occasionally, as adenocarcinoma was reported in two cases and a case with carcinoid as well [11, 12]. In our case, histopathology report is unremarkable. 11% cases had a communication between the gut and the cyst, and 2% cases had an ectopic gastric mucosa. Ultrasound is the best investigation with 95% specificity and PPV of more than 90%. But the findings are operator dependent. Our literature review shows only ten cases with cecal duplications and radiological intussusception was depicted. Out of these four had hydrostatic reduction with incomplete reduction and laparotomy for the rest. Other diseases mimicking the enteric cysts include intussusception, appendicitis, pancreatitis, mesenteric and ovarian cyst. The treatment of choice is resection along with anastomosis. The overall prognosis is good. However, two deaths secondary to sepsis occurred.

5. Conclusion
Dduplication cysts of the cecum are rare with limited reported literature. It is more common in females. Most common presentation include palpable abdominal mass, pain mimicking appendicitis, bleeding per-rectum, abdominal distension, and intussusception with acute or chronic intestinal obstruction. The delay in the diagnosis can lead to high morbidity.

References