

Duplication Cyst Presenting as Intussusception: Report of Two Cases

SANJEEV B. JOSHI, ROHITH MUDDASETTY, HARISH B.N. PATEL

ABSTRACT

Duplication cysts are one of the rare congenital anomalies affecting the gastrointestinal tract. Small intestine is the most common site followed by colonic and gastric duplications. The commonest presentation is intestinal obstruction followed by bleeding and rarely volvulus. These

cysts can be detected on an antenatal scan. Postnatally these cysts can be diagnosed on Ultrasonography or Computed Tomography of the abdomen. We report two cases of cystic duplications, presented with acute intestinal abdomen due to intussusception, in the Department of Surgery.

Keywords: Gastrointestinal tract, Marsupialization, Segmental Resection

CASE REPORT

Case 1

An infant aged one month presented with history of bilious vomiting and abdominal distension for a period 2 days. On examination, child was dehydrated and sick with a distended abdomen, with step ladder pattern of peristalsis evident over the abdomen. Plain radiograph of abdomen showed multiple air fluid levels involving the Jejunal loops. Ultrasonography of abdomen revealed a jejunojejunal intussusception with a cyst. At laparotomy, jejunojejunal intussusception [Table/Fig-1] was reduced manually revealing the duplication cyst on the mesenteric side of the jejunum [Table/Fig-2]. A segmental resection of the jejunum including the duplication cyst was done and gut continuity was restored by a jejunojejunal anastomosis [Table/Fig-3,4]. Post operative stay in hospital was uneventful. Post-operatively, regular follow-ups were done for 6 months. Child respond well, he was healthy and gained appropriate weight.

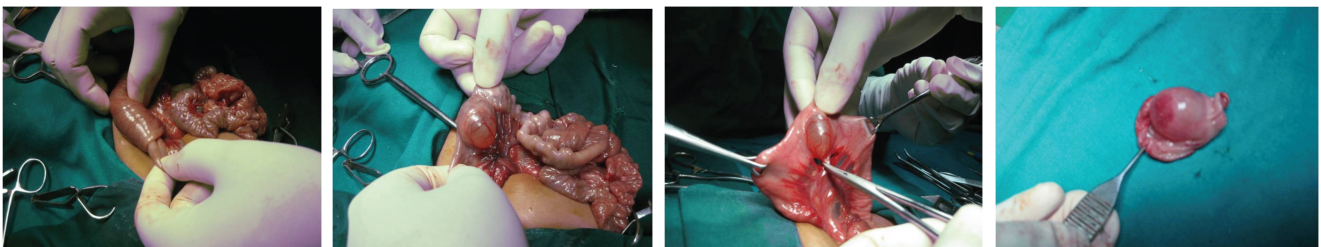
Case 2

A 12-year-old boy, presented with history of intermittent

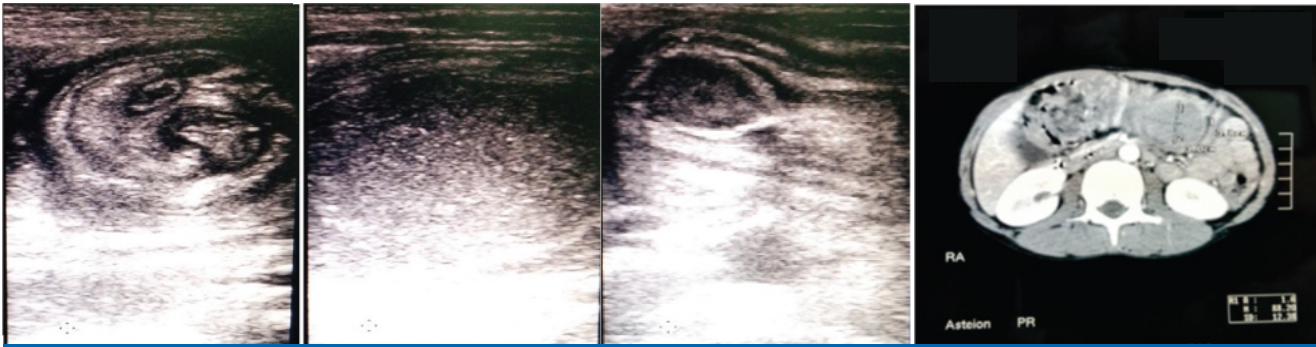
colicky abdomen pain. On examination, a firm mass was felt in the right iliac fossa extending to right lumbar region. Bowel sounds were increased. There was ballooning of the rectum suggestive of intestinal obstruction.

Ultrasonography of abdomen showed ileocecal intussusception with a cystic lesion [Table/Fig-5]. Keeping the possibility of duplication cyst acting as the lead point for ileocecal intussusception, a contrast enhanced computed tomography of abdomen and pelvis was done which confirmed ileocecal intussusception and duplication cyst [Table/Fig-6].

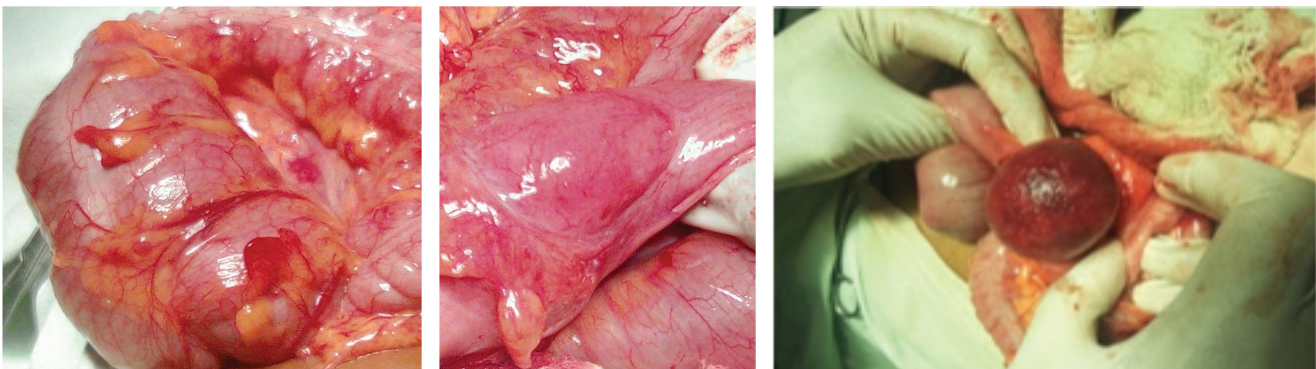
Patient underwent laparotomy through a right transverse supraumbilical incision. During surgery, ileocecal intussusception was manually reduced [Table/Fig-7]. The duplication cyst was on either side of ileocaecal valve, distal part of which was projecting into cecal lumen, with proximal extension onto terminal ileum [Table/Fig-8]. A decision was made to preserve the ileocaecal junction, along with the ileocaecal valve. Appendectomy was done and incision was extended from base of appendix along the anterior



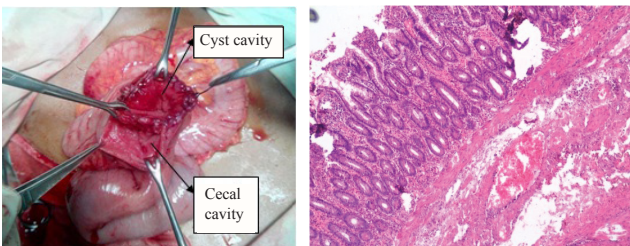
[Table/Fig-1]: Picture showing Jejunojejunal intussusceptions. **[Table/Fig-2]:** Picture showing a cystic type duplication cyst. **[Table/Fig-3]:** Segmental Resection. **[Table/Fig-4]:** Duplication cyst with adjacent bowel after resection.



[Table/Fig-5]: USG pictures of the intussusception with a cyst. **[Table/Fig-6]:** CECT Abdomen images showing Intussusception and Duplication cyst.



[Table/Fig-7]: Intra operative picture of intussusceptions. **[Table/Fig-8]:** Picture of duplication cyst with proximal extension onto terminal ileum. **[Table/Fig-9]:** Cyst within ceecal cavity.



[Table/Fig-10]: Marsupialization of the cyst.

[Table/Fig-11]: Histopathology showing ileal mucosa.

tenia. The cyst was visualized, needle aspiration showed thick chylous fluid [Table/Fig-9]. The cyst was opened and around 200 ml of thick fluid was drained. Cyst was marsupialized into the caecum and the mucosal lining of the cyst was approximated with the caecal mucosa thus leaving a wide opened mouth of the cyst into the caecum [Table/Fig-10]. Cecum was closed primarily. Post operative stay in hospital was uneventful. The histopathological evaluation of the cyst wall showed ileal mucosa [Table/Fig-11]. There was no evidence of any ectopic tissue. In the next follow-up after 1 year, child was asymptomatic, ultrasonography of abdomen was also normal.

DISCUSSION

Duplication cysts are hollow, epithelial lined structures present in the mesenteric border of the intestine. Ileum is commonest site constituting 44% of cases followed by colonic duplications (15%), gastric duplications (7%) [1,2]. Two thirds of the cases present in first two years of life. Two types are described, tubular and cystic. Cystic duplications are common (90%). The commonest presentation is intestinal obstruction [3]. Sometimes ectopic mucosa can be present in which case they present with pain, ulceration, bleeding or perforation.

Gastrointestinal tract duplication cysts are found 1 in 4500 neonatal autopsies [4] and 1 in 18000 live births. Duplications are hollow structures that involve the mesenteric side of the associated GI tract. They tend to share a common muscular wall and blood supply with its mature bowel. Small bowel duplications constitute 45% of all alimentary tract duplications followed by colonic and gastric duplication. The cysts usually contain chyle or mucus.

Several theories have been proposed regarding the development of these duplication cysts. Persistence of the endomesenchymal tract between amnion and yolk sac, partial twinning of the foregut or hindgut, persistent

embryological diverticula, aberrant luminal recanalization is some of them [5].

Two varieties are described, cystic duplications and tubular duplications. The cystic duplications commonly present with obstruction by exerting external pressure on the lumen, by acting as a lead point for intussusception or occasionally by causing a volvulus. Tubular duplications have the same features as the cystic variety but obstruction occurs due to the external pressure. Ectopic mucosa can be present in these duplication cysts. Duplications containing gastric mucosa are at risk of peptic ulceration, perforation and hemorrhage. Ectopic pancreatic tissue has been reported in duplications of the stomach, ileum and colon [6,7].

Antenatal diagnosis can be made by prenatal scans which show dilated bowel loops. Postnatally diagnosis is usually made by ultrasonography or barium contrast studies of abdomen. However, CT and MRI are helpful.

Treatment of choice is segmental resection wherever possible. Other procedures include mucosal stripping and drainage procedure (Bishop Koop) [8] especially for long tubular duplications.

CONCLUSION

Duplication cyst is a rare anomaly which usually present in early period of life. The commonest presentation is acute intestinal obstruction. The treatment of choice is segmental

resection of the involved bowel including the cyst. In cases where segmental resection is not feasible drainage procedures or mucosal resections can be done.

CONSENT

Both cases have been presented here after getting informed written consent from the parents.

REFERENCES

- [1] Corroppolo M, Zampieri N, Erculiani E, Cecchetto M, Camoglio FS. Intussusception due to a cecal duplication cyst: a rare cause of acute abdomen. *Pediatr Med Chir.* 2007;29(5):273-74.
- [2] Gross RE. Duplications of the alimentary tract. *The Surgery of Infancy and Childhood.* 1953:221-45.
- [3] Holcomb GW 3rd, Gheissari A, O'Neill JA Jr, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. *Ann Surg.* 1989;209(2): 167-74. [Medline].
- [4] Jancelewicz T, Simko J, Lee H. Obstructing ileal duplication cyst infected with Salmonella in a 2-year-old boy: a case report and review of the literature. *J Pediatr Surg.* 2007;42(5):E19-21.
- [5] Fitz RH. Persistent omphalomesenteric remains: their importance in the causation of intestinal duplication, cyst formation and obstruction. *Am J Med Sci.* 1884;88:30-57.
- [6] Ladd WE, Gross RE. Surgical treatment of duplications of the alimentary tract. *Surg Gynec Obstet.* 1940; 70:295-307.
- [7] Smith JR. Accessory enteric formations: classification and nomenclature. *Arch Dis Childh.* 1960; 35:87-89.
- [8] O'Neil J, Rowe M. Duplications of the gastrointestinal tract. In: *Essentials of Pediatric Surgery.* St. Louis, Mo: Mosby Yearbook; 1995:520-25.

AUTHOR(S):

1. Dr. Sanjeev B. Joshi
2. Dr. Rohith Muddasetty
3. Dr. Harish B.N. Patel

PARTICULARS OF CONTRIBUTORS:

1. Associate Professor, Department of Paediatric Surgery, Vijayanagar Institute of Medical Sciences, Karnataka, India.
2. Post Graduate, Department of General Surgery, Vijayanagar Institute of Medical Sciences, Karnataka, India.
3. Post Graduate, Department of General Surgery,

Vijayanagar Institute of Medical Sciences, Karnataka, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Rohith Muddasetty,
Room Number 4, New PG Boys Hostel, VIMS Campus,
Bellary-583104, Karnataka, India.
E-mail: rohith.m.setty@gmail.com

FINANCIAL OR OTHER COMPETING INTERESTS:

None.

Date of Publishing: Jul 01, 2016