Pediatric Surgery in Tropics
2025 (Apr-Jun); Volume 2, Issue 2: 109-113
DOI: 10.70947/pst.2025.18

Clinical Audit

Intestinal Duplications: Our experience and Review of Literature

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Keywords
Abdominal pain
Bowel surgery
Gastrointestinal bleeding
Intestinal Duplication
Intestinal obstruction
Mesenteric cysts

Abbreviations
CIDC - Completely isolated duplication cyst
ED - Enteric duplication
GIDC - Gastrointestinal duplication cyst
USG - Ultrasonography

Abstract

Introduction: *Gastrointestinal duplication cysts (GIDC) are rare and they occur anywhere from the oro-pharynx to the anal canal. They can be cystic or tubular and they may or may not have communication with the lumen of the adjacent bowel. Accordingly, they have varying clinical presentation. In this article we share our Indian experience with GIDC.*

Methods: *This study is a retrospective analysis of 6 cases of GIDC treated over a 3-year period (2013-16). Case records were reviewed for details of clinical presentation, nature of treatment and outcome.*

Results: *All the patients were below 6 years of age with presentations ranging from intestinal obstruction to gastrointestinal hemorrhage and pain. The male-female ratio was 5:1. One of them had a congenital isolated duplication cyst, which is very rare. The duplicated bowel was cecum in 2, jejunum in 2 and colon in 1. Pre-operative diagnosis was made in all except one patient who presented with features of intestinal obstruction. Ultrasonography and computed tomography scan were the main imaging modalities of diagnosis. They were treated by segmental resection in 3 children and cyst enucleation in 1 case. Histopathology confirmed the diagnosis. Four patients did well in the follow-up, 1 died and 1 was referred elsewhere.*

Conclusions: *GIDC commonly presented with sub-acute intestinal obstruction. With modern imaging studies, preoperative diagnosis is possible in a majority of cases. Final outcome is consistently good.*

INTRODUCTION

Gastrointestinal duplication cysts (GIDC) are rare and can involve any part of the alimentary tract, from the mouth to the anus.[1,2] They may be cystic or tubular in nature and are primarily present on the mesenteric side of the gut.[2] They are commonly seen within the first 2 years of life.[1-3] Their incidence is 1:4,500 births.[4] About 30% of all the GIDC were diagnosed in adulthood, with the majority being discovered incidentally in antenatal scans.[5,6] About 75% of GIDC are located in the abdomen, and the most common of which is ileal duplication (53%) and the least common is eso-

phageal duplications (2%). Cystic duplication of the cecum is particularly uncommon, with only 19 cases reported in the English literature.[4] GIDC may manifest as solid or cystic tumors, intestinal obstruction, perforation, gastrointestinal bleeding or intussusception. Therefore, a high index of suspicion is essential to diagnose this condition. In this report we describe our clinical experience with 6 cases of GIDC.

MATERIALS AND METHODS

This is a retrospective analysis of 6 cases of GIDC that were treated by the authors between December 2013 and December 2016. Data of all patients with respect to clinical features, radiological findings and operative findings were analyzed.

RESULTS

Details of clinical presentation, operative findings and treatment are summarized in table 1. Age at the time of presentation varied from 1 month to 6 years. Out of these, 5 were male and 1 was female. Preoperative diagnosis of enteric duplication (ED) cyst was made in five cases by ultrasonography (USG) and computed tomography (CT) scan. One case presented with gross abdominal distension, and an X-ray of the abdomen was suggestive of intestinal obstruction. USG could not detect the cecal duplication cyst due to an excessive amount of luminal gas. In 2 cases, non-communicating cystic duplication of the cecum was occluding the lumen. (Fig. 1) One case had a completely isolated, non-communicating duplication of the jejunum. (Fig. 2) This was the only child who presented with pain without any obstruction. A 6-year-old malnourished child with duplication cyst of the transverse colon presented with sub-acute intestinal obstruction. The child was very sick on admission and died of encephalitis before being considered for a surgical operation. One girl presented with intermittent gastro-intestinal bleeding and her hemoglobin was 5 g/dl. She was found to have a 100 cm long communicating tubular duplication of the terminal ileum. (Fig. 3) No action was taken at laparotomy, as resection of a long segment of ileum was ought to cause short-bowel syndrome and the expertise for doing Bianchi procedure was not available in our hospital. Hence, after correcting anemia, she was referred to an advanced center for further management. Three patients were managed by resection of the duplicated segment and enucleation of the cyst was possible in one. Postoperative recovery in all the 4 was uneventful.

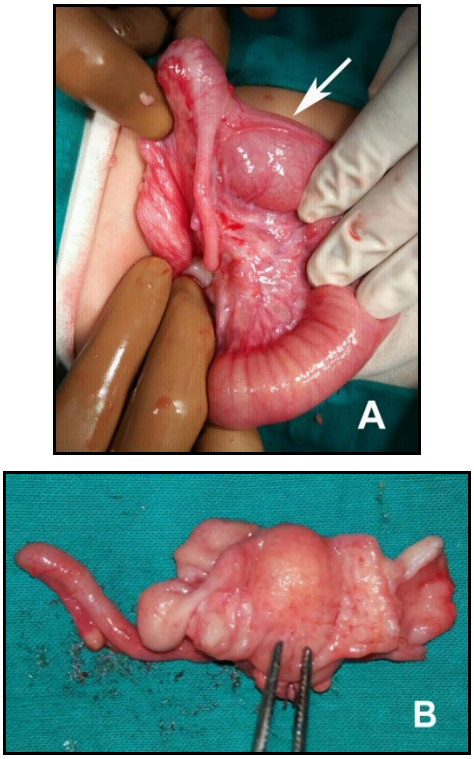
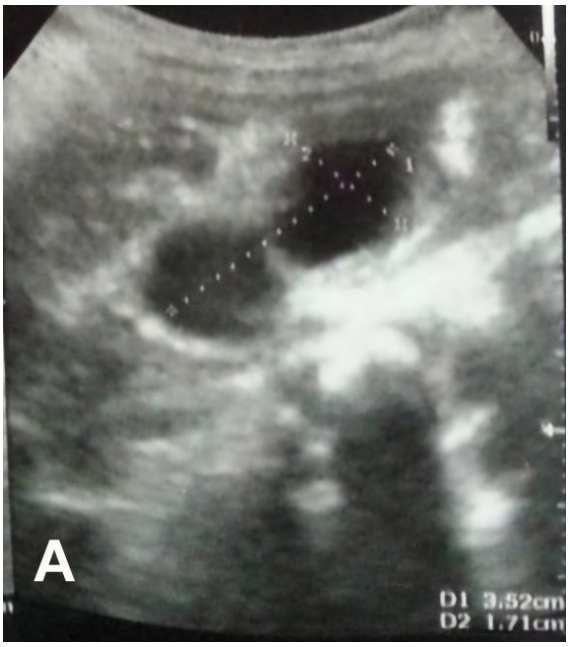
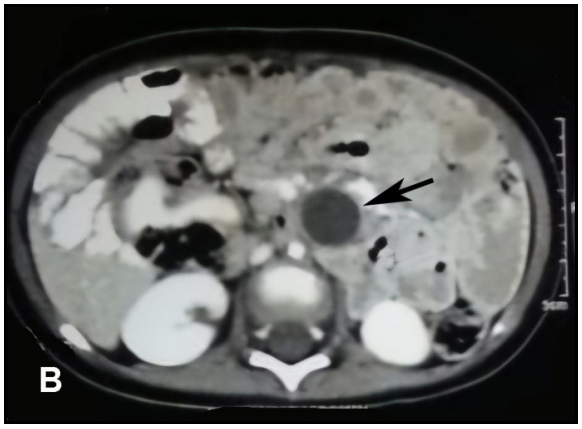


Fig 1. *Cecal duplication cyst (A) External view of the non-communicating cyst (arrow), (B) Internal view showing the luminal encroachment of the cyst*

DISCUSSION

GIDC are rare anomalies with a population incidence of 0.02%. A majority of them present during infancy.[7,8] In our study, there were two infants. Lined with intestinal mucosa, these cysts share a common wall and mesenteric blood supply with the adjacent intestine. Usually they do not communicate with the gut lumen. The term 'duplication' was popularized by William Ladd in 1930, and Robert Gross further classified it in 1950s.[9,10]





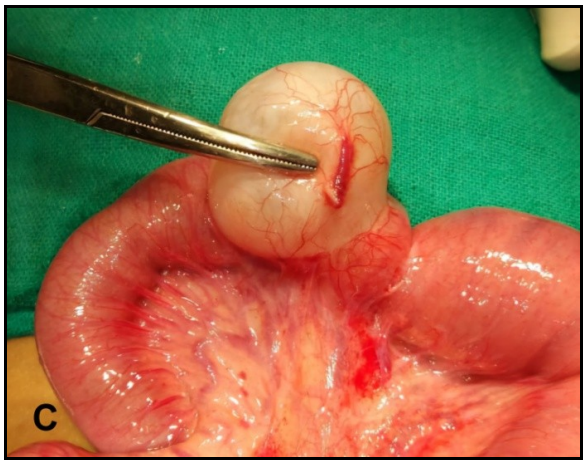


Fig 2. *Completely isolated duplication cyst of the proximal jejunum. (A) USG showing dumb-bell shaped cyst, (B) CT scan showing thick walled cyst (arrow), (C) Intraoperative photograph of the cyst*

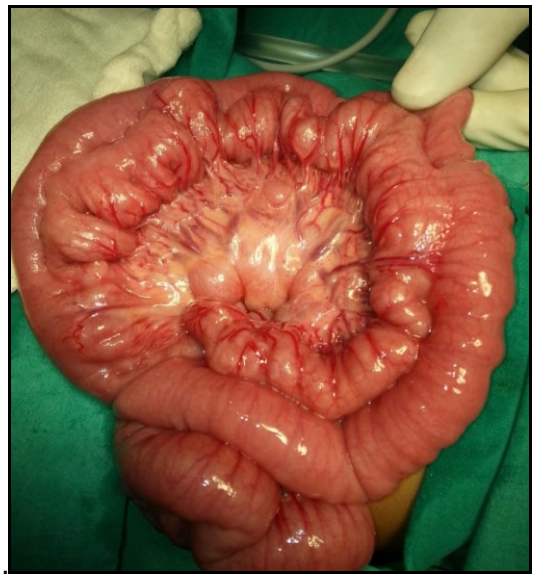


Fig 3. *Long, tubular duplication of the ileum, extending over 100 cm in length and freely communicating with the ileal lumen*

The 3 essential diagnostic criteria of GIDC are:[11]

a) Cyst wall with well-developed smooth muscle layer
b) Mucosal lining consistent with that of adjacent intestine
c) Proximity of the cyst to the alimentary tract

The ileum is the commonest site of GIDC, followed by the colon, jejunum, stomach, and duodenum.[12] However, these cysts are rare in the cecum and esophagus.

The exact etiology of GIDC remains uncertain. One theory suggests that they are the result of defective recanalization of the intestinal lumen after the solid stage of embryological development.[13] The split notochord theory posited, neural tube traction as the cause of ED, as the latter is often associated with vertebral and spinal cord anomalies.[14]

Symptoms vary according to the size, morphology, and location of GIDC. Asymptomatic lesions are diagnosed incidentally. ED may also present with abdominal pain, distension, vomiting, constipation gastrointestinal bleeding, or intestinal obstruction.

Table 1. Clinical details of intestinal duplication in children

Pt. No	Age	Sex	Clinical Features	Location	Pathological nature	Management	Outcome
1	6 mo	M	Epigastric pain	Jejunum	NC	SRPA	Survived
2	6 yr	M	SAIO	Transverse colon	Cystic	Not operated	Died of encephalitis
3	1.5 yr	M	SAIO, Palpable lump	Jejunum	Cystic	Cyst enucleation	Survived
4	2 yr	M	SAIO	Cecum	Cystic, NC	SRPA	Survived
5	4 yr	F	Massive GI bleed	Terminal ileum	Communicating, Tubular (100 cm long)	Operation was abandoned due to technical limitations	Referred to another center
6*	1 mo	M	SAIO	Cecum	Cystic, NC	SRPA	Survived

** Investigated with only X-ray. All the other 5 patients were investigated with ultrasonography and CT scan.*

GI - Gastrointestinal, NC - Non-communicating with intestinal lumen, SAIO - Subacute intestinal obstruction, SRPA - segmental resection of bowel with primary anastomosis

There may be associated spinal and genito-urinary anomalies, although none of our cases had them. Two cases of cecal duplication presented with intestinal obstruction due to near total occlusion of the lumen, which is rare. Another case of long tubular ileal duplication presented with recurrent gastrointestinal hemorrhage. There was extensive communication with the ileal lumen. The bleeding is attributable to peptic ulcer from the presence of ectopic gastric mucosa, although this could not be confirmed histologically in our case as resection was not performed.

Ultrasound and CT scans are the primary tools of diagnosing these cysts. Sonography is especially valuable in revealing an echogenic inner margin (mucosa) and an outer hypoechoic rim (smooth muscle) - the 'muscular rim sign'.[15] Differential diagnoses include cysts of mesenteric, omental, ovarian and pancreatic origin. Segmental bowel resection is the preferred treatment of GIDC.[3]

One of our patients (case 3) had a completely isolated duplication cyst (CIDC). According to Menon a CIDC is diagnosed by the absence of any communication with the lumen of the adjacent alimentary tract, beside the typical histopathological features of a duplication cyst.[16] Only 13 cases of CIDC have been reported in the world literature.[17]

Since GIDC share a common muscle wall with the normal bowel, exclusive excision of a duplication cyst is typically not possible. The optimal procedure involves resection of the adjoining intestine along with the duplication cyst after ligating their mesenteric vascular supply. However, in one of our cases, complete enucleation of the cyst was possible as it was a CIDC.

In cases of long tubular duplications, complete excision is not advisable due to the high risk of developing short bowel syndrome. Instead, mucosal stripping by multiple incisions is recommended.[18] Untreated ED may be complicated by perforation, hemorrhage, obstruction and malignancy.

CONCLUSION

GIDC should be considered in the differential diagnosis of abdominal cystic lesions. Curative resection is possible a majority of cases. Enucleation of the cyst alone can be considered in CIDC.

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Received : 6 December 2024
Accepted : 12 January 2025

Acknowledgements : None
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : None (Retrospective reporting of routine clinical care

Citation: *Gupta R, Tomar D, Singh A, Gupta A. Intestinal duplications: our experience and review of literature. Pediatr Surg Trop 2025 (Apr-Jun); 2(2): 109-113. https://doi.org/10.70947/pst.2025.18*

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