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Our experience with unusual gastrointestinal tract duplications in infants

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ABSTRACT

Background: Classical duplications may present along any part of gastrointestinal tract (GIT) from mouth to anus. Atypical or unusual rare varieties of GIT duplications may also occur, but with different anatomical features. **Materials and Methods:** We reviewed our 5-year record (February 2008-January 2013) to describe clinical profile of unusual GIT duplications in neonates and small infants. **Results:** Three patients with atypical variety of GIT duplications were managed in our department during this tenure. Two were females and one male. Age was ranged between 11 days and 2 months. All patients presented with massive abdominal distension causing respiratory embarrassment in two of them. In all patients, the pre-operative differential diagnoses also included GIT duplication cysts. Computerized tomography (CT) scan showed single huge cyst in one and multiple cysts in two patients. In one patient the CT scan also depicted a thoracic cyst in relation to posterior mediastinum. At operation, one patient had colonic tubular duplication cyst along with another isolated duplication cyst, the second case had a tubular duplication cyst of ileum with its segmental dilatation, and in the third case two isolated duplications were found. Duplication cysts were excised along with mucosal stripping in one patient, cyst excision and intestinal resection and anastomosis in one patient, and only cysts excision in one. All patients did well post-operatively. **Conclusion:** We presented unusual GIT duplications. These duplications are managed on similar lines as classical duplications with good prognosis when dealt early.

Key words: Atresia, gastrointestinal tract, isolated duplications, neonate, segmental dilatation, unusual duplication cyst

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INTRODUCTION

Typical or classical duplication cyst is featured by an intimate contact with any part of GIT from mouth to anus, has smooth muscles in its wall, and lined by a mucosa resembling that of any part of GIT.[1-3] Atypical duplications may also occur in various forms. Isolated duplications are atypical duplications that have smooth muscles in their walls and lined by GIT mucosa, but they don't possess an intimate contact with GIT.[4-7] GIT duplications are aborted alimentary tract therefore any disease or anomaly that affects normal GIT may also, theoretically, affect duplication cyst such as atresia, perforation, inflammation, and so on thus making them unusual.[5,7] We reviewed our 5-year record to describe clinical profile of infants identified with unusual type of GIT duplications. An attempt is made to explain their formation.

MATERIALS AND METHODS

The medical record of unusual GIT duplications managed in the Department of Paediatric Surgery, The Children's Hospital and the Institute of Child Health Lahore between February 2008 and January 2013 was retrieved and information regarding demography, clinical presentation, investigations, operation, and outcome was analysed and presented.

RESULTS

There were three patients with atypical GIT duplications. Two were female and one male patient. Age was ranged between 11 days and 2 months [Table 1].

The patient no. 1 presented with massive abdominal distension causing respiratory distress. On examination, abdomen was massively distended with dilated cutaneous veins and a small umbilical hernia. Abdominal radiograph showed a mass effect with displacement of bowel to one side. CT scan

performed showed two huge cysts occupying almost entire abdomen [Figure 1]. Pre-operative diagnosis was mesenteric cyst with a differential of duplication cyst.

The patient no. 2 presented with abdominal distension and occasional non-bilious vomiting. Ultrasound and CT scan showed a huge cyst in the abdomen.

The presentation in patient no. 3 was similar to that of patient no. 2. Ultrasound and CT scan depicted two cysts in the abdomen; in addition one cyst was also present in the chest in relation to the posterior mediastinum [Figure 2].

All the laboratory investigations were within normal limits in all patients. All the three patients had preoperative differential diagnoses of GIT duplication cyst.

In the patient no. 1, there were two huge cysts in the abdomen. The proximal lesion was tubular in shape, in intimate contact with cecum, extended along ascending, transverse, and descending colon where it ended blindly. The distal lesion was cystic and present along sigmoid colon without any intimate contact [Figures 3 and 4]. Both of the lesions shared a common mesentery, originating from the mesentery of the normal colon, with a V-shaped defect in between both of these lesions [Figure 5].

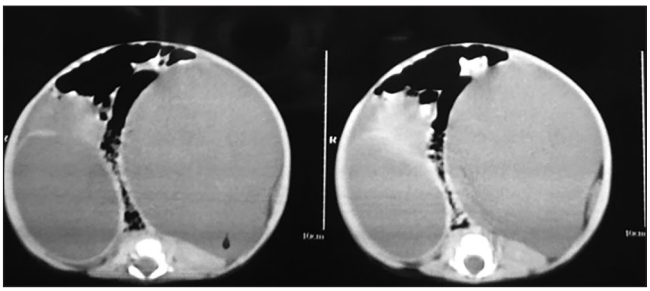


Figure 1: CT scan showing two huge cyst occupying whole of the abdomen

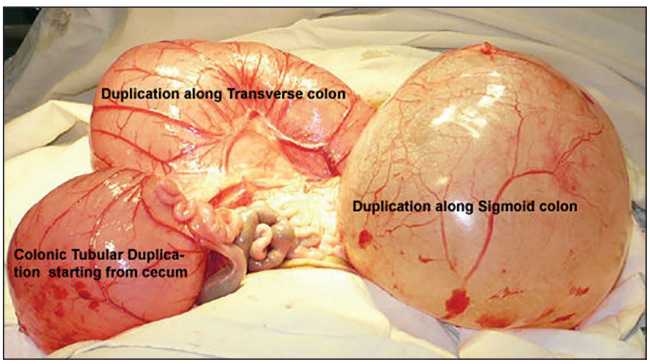


Figure 3: Tubular colonic and isolated duplications in patient no. 1

Initially, the distal lesion was excised followed by proximal lesion. While mucosal stripping at the level of cecum where it was sharing common wall, a small perforation occurred in the cecum which was managed by tube caecostomy. Histopathology confirmed both cysts as duplication cysts with colonic type mucosa lining the both.

In the patient no. 2, there was a 1.5 feet long tubular communicating duplication cyst of the mid ileum. The tubular duplication had three parts; the first part was tubular in shape, which led to a 7 cm × 8 cm sized cystic portion (second part) which again led to a small tubular portion (third part), which ended blindly [Figures 6 and 7]. All the three portions were communicating with each other and with an abrupt transition from tubular to the central cystic component. The duplication was excised with ileoileal anastomosis. The histopathology confirmed it as duplication cyst.

The patient no. 3 had two cysts in the abdomen and another cystic lesion in the chest. It was decided

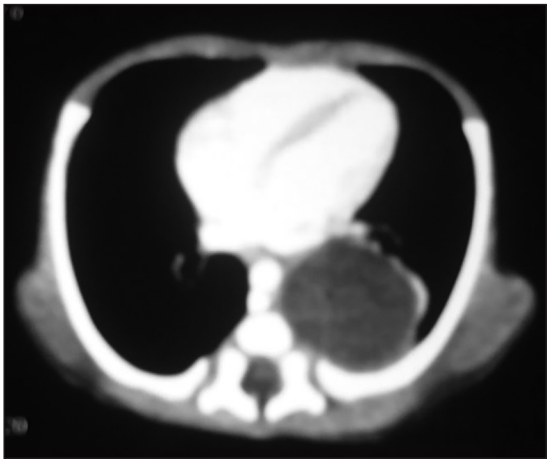


Figure 2: Cystic lesion in the chest in patient no. 3

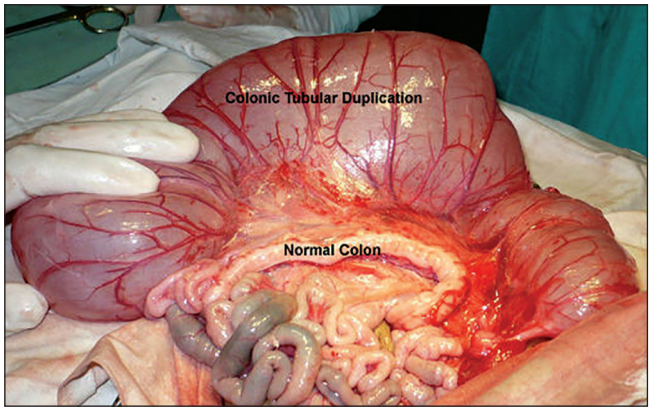


Figure 4: Patient no. 1 after excision of the isolated duplication

to address abdominal lesion first followed by chest lesion after few weeks. At operation, there were two huge cysts that did not have any intimate contact with any part of the GIT [Figure 8]. Both of the lesions had their separate narrow mesentery arising from the retroperitoneum. Both of the lesions were excised. The histopathology revealed smooth muscles in the wall, and gastric mucosa lining both of the cysts.

All the patients had uneventful postoperative course. In patient no. 1, the tube caecostomy was removed after 2 weeks and patient discharged in good clinical condition. The patient no. 3 lost to follow-up despite counselling about intra-thoracic cystic lesion. Table 1 describes the summary of these patients.

DISCUSSION

We presented three cases of unusual GIT duplications. The presentation is similar as that of typical duplications[5-8] however certain anatomical features differentiated them into atypical category. In the first case, there were two duplications; one was typical tubular duplication extending from the caecum to the level of descending colon and second was isolated cystic duplication along sigmoid colon both shared common mesentery with a gap between two lesions. Figure 5 is

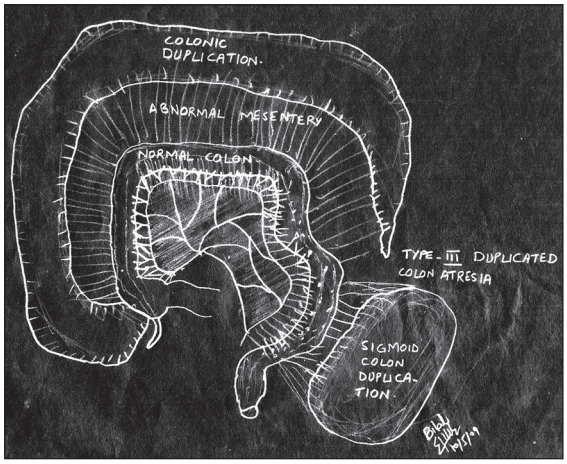


Figure 5: Schematic diagram of operative findings in the patient no. 1

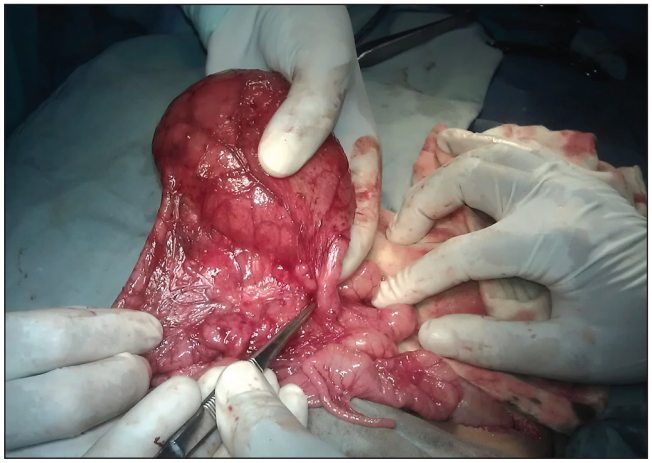


Figure 6: Complex duplication cyst in patient no. 2

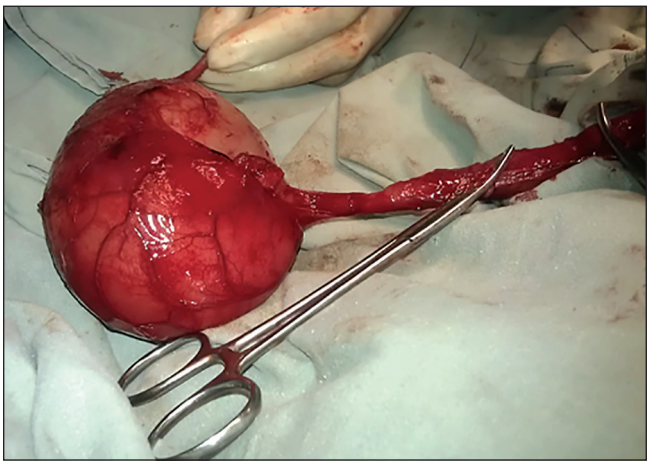


Figure 7: Segmental dilatation of the ileal tubular duplication (after excision)

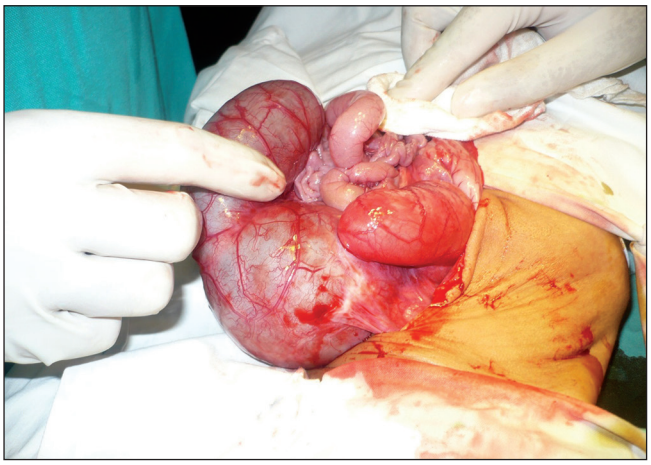


Figure 8: Two isolated cyst in the patient no. 3

Table 1: Summary of all patients

Patient no.	Age	Presentation	Duplication	Procedure	outcome
1	2 months	Abdominal distension, respiratory distress	1 tubular colonic and other isolated	Excision, tube caecostomy at the site of perforation	Alive
2	25 days	Abdominal distension, vomiting	Ileal tubular with segmental dilatation	Excision and ileoileal anastomosis	Alive
3	11 days	Abdominal distension, respiratory distress	2 isolated cysts	Excision	Alive

a sketch of the operative findings. It can be speculated that initially the distal isolated lesion might have been the part of proximal lesion and an *in utero* vascular event would have resulted in a type IIIa atresia of the duplication resulting in isolation of the distal lesion. As histopathology commented the same colonic mucosa in both of the lesions, therefore, our assumption seems the most plausible explanation for sequestration of the distal lesion. Pant *et al*.[7] and Norris *et al*.[5] have reported similar cases where a typical duplication coexisted with an isolated duplication and proposed that a vascular insult could have led to the isolation.

In the second case, the duplication cyst was interesting and complex one having two (afferent and efferent) tubular parts and a central cystic part. It appeared as combined tubular and cystic duplication cyst. The transition from tubular components to the cystic component was abrupt and without tapering. This kind of lesion is reported in the normal GIT as segmental dilatation and may involve small and large bowel. It appears that in our second case there was a segmental dilatation of the communicating tubular duplication of the ileum. A perusal of literature did not reveal any similar GIT duplication with segmental dilatation.

In the third case, we encountered two isolated duplications. There was another cystic lesion present in the chest that may have differentials as duplication cyst, bronchogenic cyst, or sequestration. In presence of two duplications in the abdomen it can be presumed as duplication cyst along the oesophagus. Both of the cysts could be presumed initially the part of the cyst in the chest and during formation of the diaphragm, these cysts could have isolated in the abdomen. However, multiple lesions in the abdomen are difficult to explain. Both the cyst had gastric mucosa is also in favour of our opinion. If the patient would have operated for thoracic lesion, it could be possible to further look into the aetiology of isolation of the duplications in the third case.

CONCLUSION

Atypical GIT duplications have similar presentations, investigations, and operation as typical duplications. Certain structural differences make these unusual. These unusual duplications open further avenue of exploring the aetiology and pathogenesis of duplications. These cases endorse our belief that duplications cyst may be affected by any lesion that may affect normal GIT.

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