Multiple Intraabdominal Cysts Could Be A Duplication Of Intestine: A Case Report And Treatment Protocol

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Citation

Abstract
Duplication of intestine is uncommon with varied presentation. We report an 8-month-old male baby with multiple intraabdominal cysts. On exploration, there were multiple cystic duplications of terminal ileum and ascending colon. Surgery was done successfully. Different types of duplication cysts, their presentation & treatment protocol are discussed.

INTRODUCTION
Enteric duplication cyst, an uncommon congenital anomaly, can occur in any portion of the alimentary tract in infants or children. Fitz first used the term intestinal duplication, but it was not widely used until popularized by Ladd in the 1930s, with further classifications by Gross in the 1950s.

Duplications of the ileum represent the most common location of all alimentary tract duplications and typically appear as non-communicating spherical cysts of varying sizes. Because of their common location at the mesenteric border, they may be easily mistaken for mesenteric or omental cysts, only to be clarified by identification of a mucosal rather than endothelial lining.

CASE REPORT
An 8-month-old male infant presented with history of mild to moderate abdominal distension for 2 months. There were no other significant gastrointestinal or urinary complaints. Antenatal history was not significant. Clinical examination: The general condition was satisfactory. The baby was well-nourished, weighing 6.5 kg and had minimal signs of dehydration on admission. Abdominal examination revealed few vague lumps in the right iliac fossa and umbilical region. There were no visible loops and peristalsis. The rest of the examination was unremarkable. The patient’s hemoglobin was 8g% and serum electrolytes were normal. X-ray of chest and abdomen was inconclusive.

Ultrasonography of abdomen was suggestive of multiple intraabdominal cysts (3 in number) in the right iliac fossa and umbilical region, suggestive of mesenteric cysts. CT scan of the abdomen could not be done, as the patient was not able to afford it. On laparotomy, there were multiple cysts present near the ascending colon and terminal ileum. The cysts near the ascending colon and ileum were resected (FIGURE 1). One cyst was opening into the caecum with tubular duplication at the terminal ileum of a length of 8-10 inches (FIGURE 2) which was removed too. Another cyst was opening into the caecum and to avoid major surgery, the ileocaecal junction was preserved and the proximal single lumen of the ileum was anastomosed to the distal two lumens. The postoperative period was uneventful. The patient is doing well at 1 year of follow-up without any complications.

Figure 1
Figure 1: Resected cyst
Histopathology was suggestive of ileal and colonic mucosa in the duplication cyst.

**DISCUSSION**

Enteric duplication cyst is an uncommon congenital anomaly that typically occurs in infants or children and has an incidence of two or three cases per year in pediatric referral centers \(^{(1,2)}\). Approximately two thirds of all intestinal duplications are discovered within the first 2 years of life, with one third identified in the newborn period. According to the definition of Ladd and Gross \(^{(2)}\), the cyst must be adherent to some part of the GI tract, contain smooth muscle in the wall, and have an internal lining of alimentary epithelium, which is not necessarily that of the adjacent segment of the GI tract. Ectopic tissue, such as gastric, squamous, transitional, and ciliated mucosa, pancreas, etc., can be found in these lesions at all levels of the GI tract \(^{(1,3)}\). The enteric cyst is named according to the location of the GI tract to which it is adjacent rather than by its lining mucosa (which can be variable).

Grossly, enteric duplication cysts are spherical cysts or tubular structures and tend to be situated on the mesenteric aspect of the alimentary canal, with which they share a common muscular wall and blood supply but have a separate mucosal lining \(^{(1,2,3)}\). The spherical cyst, almost closed, is the most prevalent type and constitutes 82% of GI tract duplications \(^{(1)}\). The tubular type, invariably communicating with the lumen, is found in only 18% of cases \(^{(1)}\). The most common site is the ileum, followed by esophagus, large bowel, jejunum, stomach, and duodenum \(^{(1)}\).

Various theories have been postulated regarding the origin of duplication cysts, like: a) abortive attempts of twinning \(^{(6)}\); b) phylogenetic reversion \(^{(7)}\); c) adhesions between endoderm and neuroectoderm \(^{(8)}\); d) persistence of embryonic diverticula; e) recanalisation and fusion of longitudinal folds.

Although many of the duplications are diagnosed incidentally, most patients present with a combination of pain and/or obstructive symptoms. These symptoms may be the direct effects of distention of the duplication or caused by compression of adjacent organs (including their associated blood supplies). In addition, abrupt hemorrhage with hemodynamic instability can be encountered in the case of a cyst lined with gastric mucosa that ulcerates and eventually erodes into adjacent organs and/or vessels.

Preoperative diagnosis of alimentary tract duplications is often difficult. Ultrasonography is also helpful to establish a preoperative diagnosis and may similarly be used as a screening tool to address the 10-20% incidence of multiple lesions. CT scanning of the chest or abdomen is useful in establishing a diagnosis and may be used to evaluate for synchronous lesions once a single duplication has been identified.

Resection of duplicated bowel is the treatment of choice \(^{(9)}\). Specifically, patients with previously undetected duplications may present in the setting of bowel obstruction or severe GI hemorrhage (i.e. ulcerating gastric mucosa within a duplication cyst). If encountered incidentally, duplications should be surgically addressed to avoid future complications. The normal and the duplicated bowel share the same blood supply, and their respective walls are most commonly fused along the mesenteric border of the bowel. On this account, it becomes difficult to separate both structures, which in most instances makes it impossible to resect the duplication alone \(^{(9)}\). Should there not be the close anastomotic relation aforementioned, resection of the duplication alone can be carried out \(^{(9)}\). However, in our case two cysts were excised and one cyst opening in the caecum was left as such and the ileum was anstomosed to the distal two lumens to avoid extensive surgery. The patient is doing well at 1 year of follow-up.

**CONCLUSION**

Duplication cysts can be easily confused with mesenteric cysts. Duplication cysts should also be kept in differential diagnosis in case of multiple intraabdominal cysts.
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