

Gastrointestinal Duplication: 10 Years' Experience

Mazen O. Kurdi, FRCS

Department of Surgery, Division of Pediatric Surgery, Faculty of Medicine
King Abdulaziz University, Jeddah, Saudi Arabia

Correspondence

Dr. Mazen O. Kurdi
P.O. Box 80215, Jeddah 21589, Saudi Arabia
e.M: mokurdi@gmail.com

Submission: 04 Sept. 2015
Accepted: 09 Dec. 2015

Citation

Kurdi MO. Gastrointestinal duplication 10 years' experience. JKAU Med Sci 2016; 23 (2): 1-14.
DOI: 10.4197/Med. 23.2.1

Abstract

Gastrointestinal duplication is a rare congenital abnormality. Over the last 10 years, 10 cases of gastrointestinal tract duplication were managed at King Abdulaziz University Hospital, Jeddah, Saudi Arabia. Duplications were variable in their level and presentation. The experience in the management of the different levels of gastrointestinal tract duplication will be presented along with a suggested general plan for the management of gastrointestinal duplication based on our experience and the collective experience from different reports in the literature.

Keywords

Gastrointestinal tract; Duplications; Children; Management; Saudi Arabia

Introduction

Fitz^[1] in 1884 was the first to use the term duplication to describe omphalomesenteric remnants, but Ladd^[2] in 1937 was the first to apply and encourage the use of this term for the real duplication which lies on the mesenteric side. Rowling^[3] put forth the following criteria for diagnosing a gastric cyst as duplication; It should have intimate contact with the bowel, have a muscular wall and be lined with gastrointestinal epithelium. The 2nd and 3rd criteria should be fulfilled in all gastrointestinal duplications but some might not be in intimate contact with the wall of the gut^[4-7].

Gastrointestinal duplication is a relatively rare congenital malformation. An average of 1-3 cases per year might be seen in a major pediatric surgery center. Bower *et al.*^[6] in 1978 reviewed 78 duplications in 64 patients seen at the Pittsburgh Children's Hospital over a 40 year period (1935-1975).

Duplication can occur anywhere from mouth to anus, but ileal duplication was found to be the

commonest in all the reported series. Various theories were suggested to explain the embryogenesis of this phenomena, none of which could fully explain the duplication at different levels and their different features^[4-7].

In this paper, the experience in the management of 10 cases of gastrointestinal duplication at the different levels will be discussed along with a suggested general plan for the management of this abnormality based on the previous reports in the literature and on our experience in the management of 10 cases.

Patients and Methods

During the last 10 years (2005-2015), 10 cases of gastrointestinal duplications were seen at King Abdulaziz University Hospital (KAUH), Jeddah, Saudi Arabia. The duplication involved different levels of the gastrointestinal tract (GIT) (Table 1). Four patients were female and 6 were male, 6 (60%) were diagnosed as duplication preoperatively and the other 4 (40%)

Table 1. Summaries of the 10 cases.

Case Age/Sex	Presentation	Pre-op Diagnosis	Diagnostic Modality of Duplication	Intra-op Diagnosis	Type of Duplication	Epithelial Lining	Treatment
1. 3 y / F Fig. 1, 2	Recurrent chest infection	Esophageal duplication	Ultrasound - CT isotope scan	Esophageal duplication	Tubular	Gastric epithelium	Excision of the duplication alone
2. 3 y / F Fig. 3, 4	Recurrent abdominal pain	Gastric duplication	Ultrasound	Gastric duplication + accessory pancreatic tissue	Cystic	Gastric epithelium	Excision of the cyst with the adjacent wall of the stomach
3. 2 d / F Fig. 5	Neonatal intestinal obstruction	Duodenal atresia	Diagnosed at laparotomy	Duodenal atresia & duodenal duplication	Cystic	No tissue excised	Duododuodenostomy and cystoduodenostomy
4. Newborn / M Fig. 6	Neonatal intestinal obstruction	Born with high imperforate anus	Diagnosed at performing colostomy	Jejunal cystic duplication	Cystic	Intestinal epithelium	Excision of the cyst with the adjacent segment of the jejunum
5. 1 y / M Fig. 7	Abdominal pain	Duplication vs. Mesenteric cyst	Ultrasound	Short tubular jejunal duplication with autonomous blood supply	Tubular	Intestinal epithelium	Excision of the duplication alone
6. 2 ½ / M Fig. 8, 9	Abdominal mass, Rt. lower abd.	Duplication cyst	Ultrasound	Ileal duplication at the ileocecal junction	Cystic	Intestinal epithelium	Excision of the cyst with the limited ileocecal segment
7. 2 ½ y / M Fig. 10, 11	Recurrent abdominal pain with bleeding / rectum	Meckel diverticulum w/ ectopic gastric mucosa	Diagnosed at laparotomy	Multiple ileal duplication perforating to the ileum.	Multiple cystic	Gastric epithelium	Excision of the cyst with the adjacent segment of the ileum
8. 4 m / M Fig. 12	Abdominal pain	Duplication cyst	Ultrasound – CT scan	Ileal duplication	Cystic	Intestinal epithelium	Excision of the cyst with the adjacent segment of the ileum
9. 10 m / M Fig. 13	Abdominal pain and mass	Duplication cyst	Ultrasound – and CT scan	Ileal duplication at ileocecal junction	Cystic	Intestinal epithelium	Excision of the cyst with the limited ileocecal segment
10. 3 m / F Fig. 14	Passage of stool/vagina	Rectovaginal fistula	Diagnosed at laparotomy	Total colonic duplication with RVF	Tubular	Colonic epithelium	Side to side anastomosis of the distal part (rectal) of the tubular duplication with repair of rectovaginal fistula

ABBR: CT scan: Computerized tomography scan; RVF: Right ventricular failure

were diagnosed intra-operatively for other surgical indications (duodenal atresia high imperforate anus, bleeding per rectum with suspicion of Meckel diverticulum, rectovaginal fistula). The level of the duplications was as follows: one was esophageal tubular duplication at gastro-esophageal junction (Figs. 1, 2) and stomach, which was excised with intact esophagus; one was gastric cystic duplication at the greater curvature of the stomach and was excised with the adjacent wall of the stomach then the stomach wall closed in two layers with excision of accessory pancreatic tissue (Figs. 3, 4); one cystic duodenal duplication with duodenal atresia was treated with internal drainage of the cyst to the duodenum (cystoduodenostomy) and duodenoduodenostomy for treatment of the atresia (Fig. 5); one cystic jejunal duplication treated by simple excision of the duplication cysts with resection of the corresponding adjacent bowel with end to end anastomosis (Fig. 6); one was short tubular jejunal duplication with autonomous blood supply and was treated by simple excision of the duplication without excision of the adjacent jejunum (Fig. 7); five ileal cystic duplications one of them with 2 cysts all treated

with excision of the cyst and the adjacent segment of the bowel (3 segment of ileum and 2 short ileocecal segment) (Figs. 8-13) and one tubular total colonic associated with recto-vaginal fistula was treated with internal drainage (Side to side anastomosis of the distal rectal part of the tubular duplication) with repair of recto-vaginal fistula according to the standard treatment for such. All duplication tissues were sent for histopathological examination except the duodenal duplication (Fig. 14). Table 1 summarizes the 10 cases.

Results

All 10 patients had successful surgical treatment of the duplication with uneventful post-operative recovery, two patients had excision of the duplication without excision of the adjacent GIT segment as they do not share the same blood supply (esophageal and jejunal with autonomous blood supply). One gastric duplication cyst treated with excision of the cyst and the adjacent wall of the stomach which needed closure of the opened stomach, six duplications were treated with excision of the duplication with the



Figure 1. Esophageal duplication (non-communicating thoraco-abdominal tubular duplication) Excision of the duplication alone with intact esophagus as esophageal duplication does not share the blood supply of the adjacent esophagus.

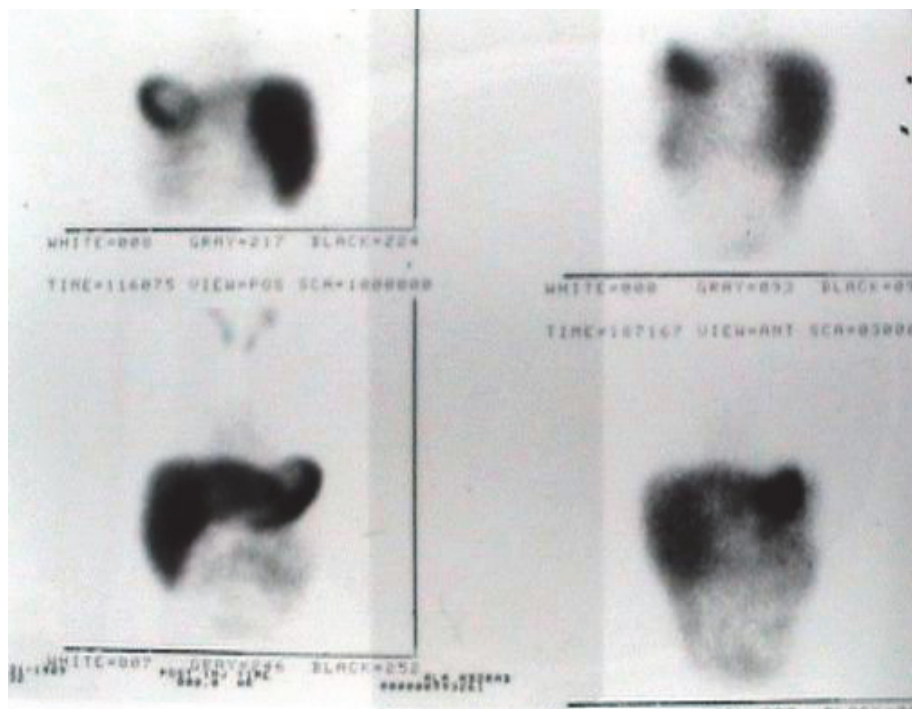


Figure 2. Isotope scan images of the esophageal duplication. Top: posterior; bottom: anterior views; Lt. early images and Rt. late images showing the tracer in the duplication (i.e., gastric epithelium).

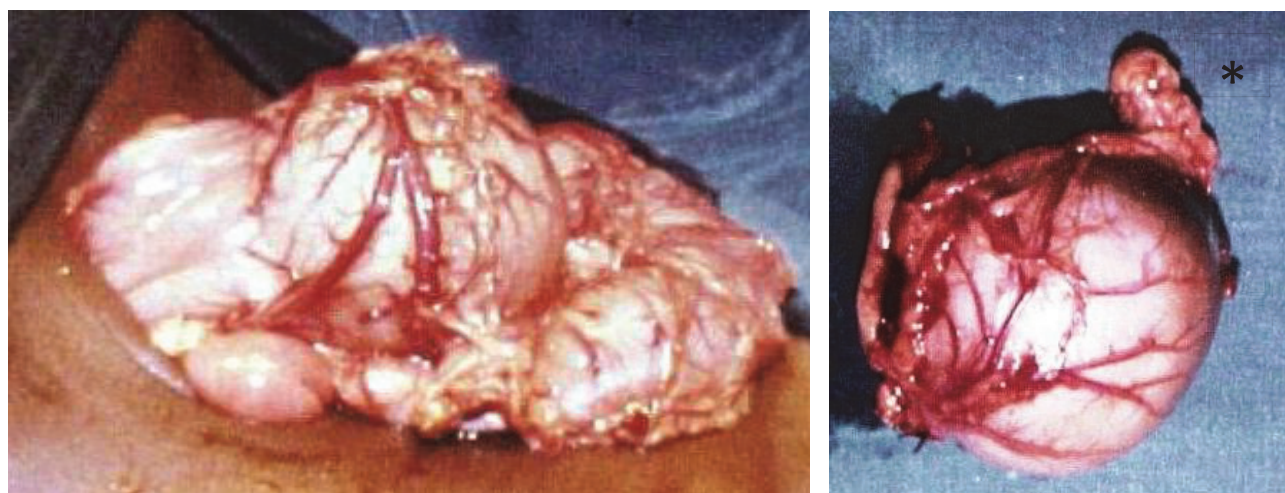


Figure 3. Gastric Duplication cyst with accessory pancreatic lobe (*) on the right. The excised part of gastric wall with the lining mucosa.

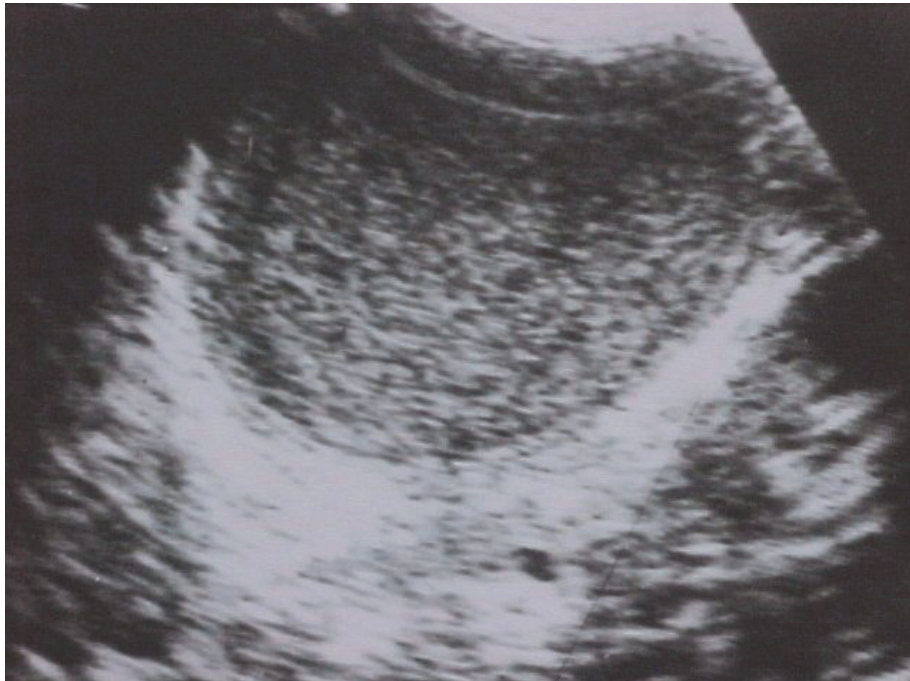


Figure 4. Ultrasound of the gastric duplication cyst with the ultrasonic feature of the duplication cyst, 3 shadows inner mucosal hyper-echoic, middle muscular echo-lucent and outer enhancement of the cyst.

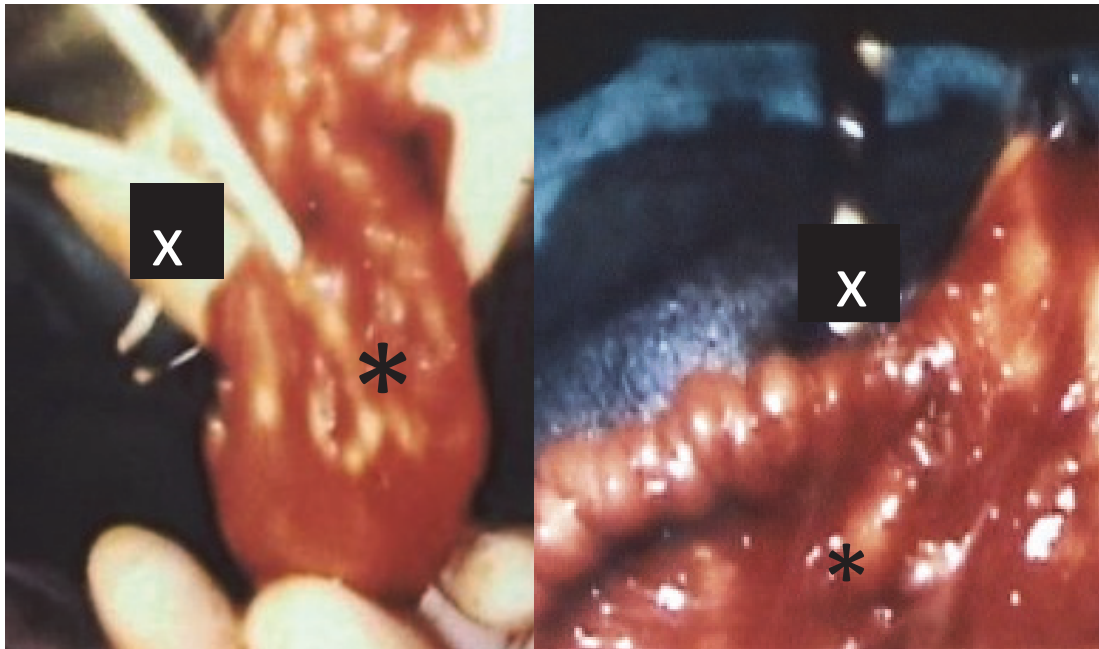


Figure 5. Patient with duodenal atresia with duodenal duplication, 2 catheters, one in the distal end of the atretic duodenum (x) and the other one in the duplication (*). Duodenoduodenostomy (x) and cystoduodenostomy(*).

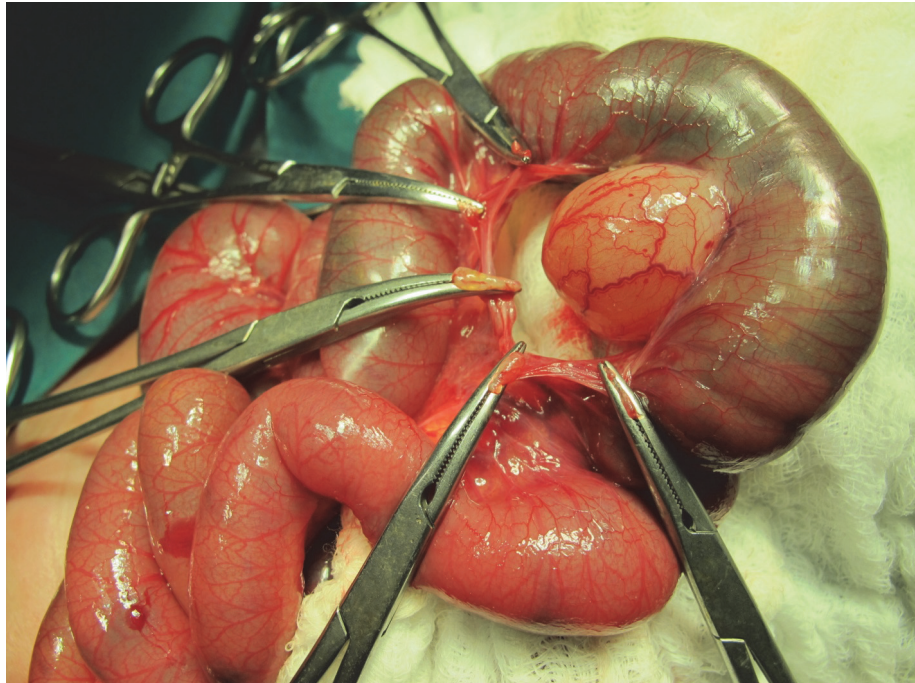


Figure 6. Cystic jejunal duplication detected intra-operatively at performing colostomy for imperforate anus, the duplication excised with the adjacent jejunum as it shares the same blood supply.

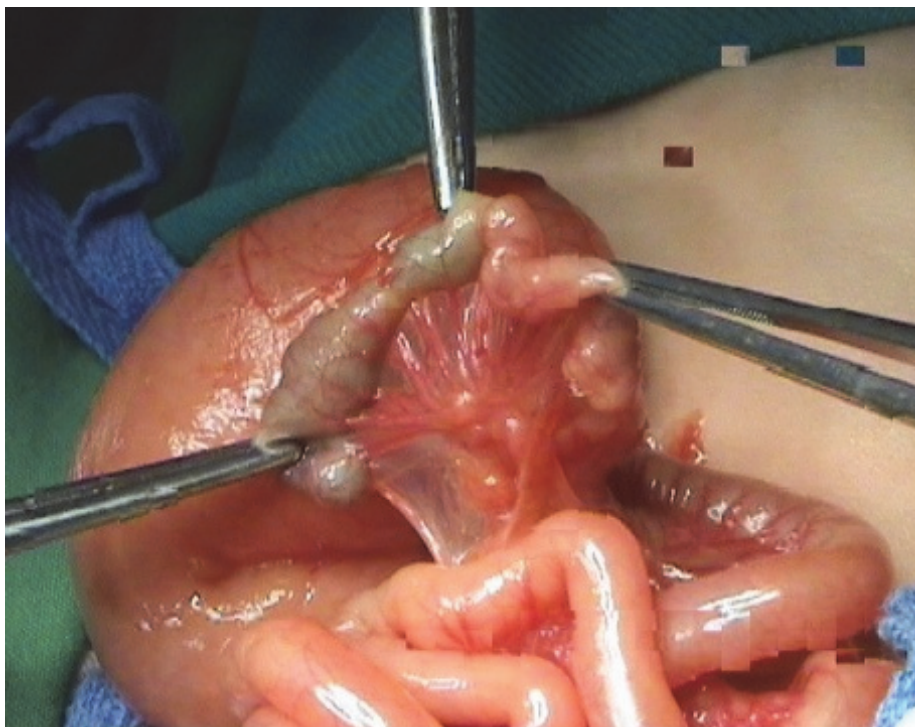


Figure 7. Short tubular jejunal duplication with autonomous blood supply.



Figure 8. Ileal duplication cyst at the ileocecal junction. Excision of the cyst with the limited ileocecal segment.

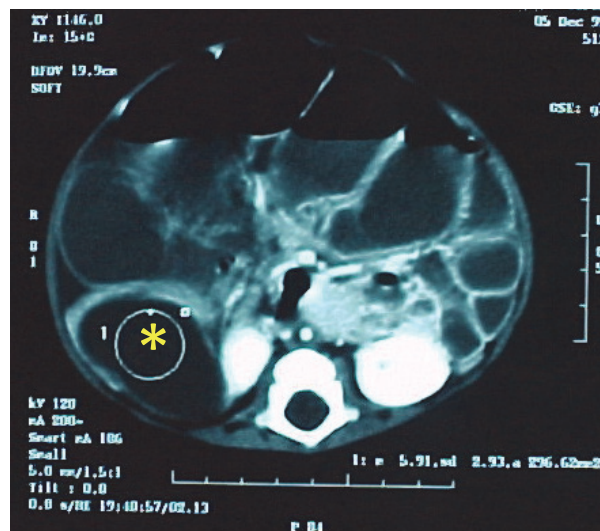
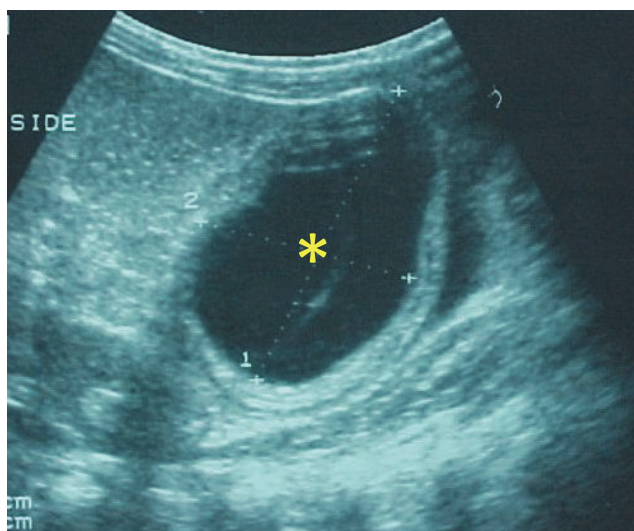


Figure 9. Ultrasound of the ileal cyst (Fig. 8. patient) with the characteristic ultrasonic features, 3 shadows inner mucosal hyper-echoic, middle muscular echo-lucent and outer enhancement of the cyst. (left). CT abdomen showing the duplication cyst * (right).



Figure 10. Resected ileal segment with the bleeding sinus leading to the duplication cyst (with gastric mucosal lining).

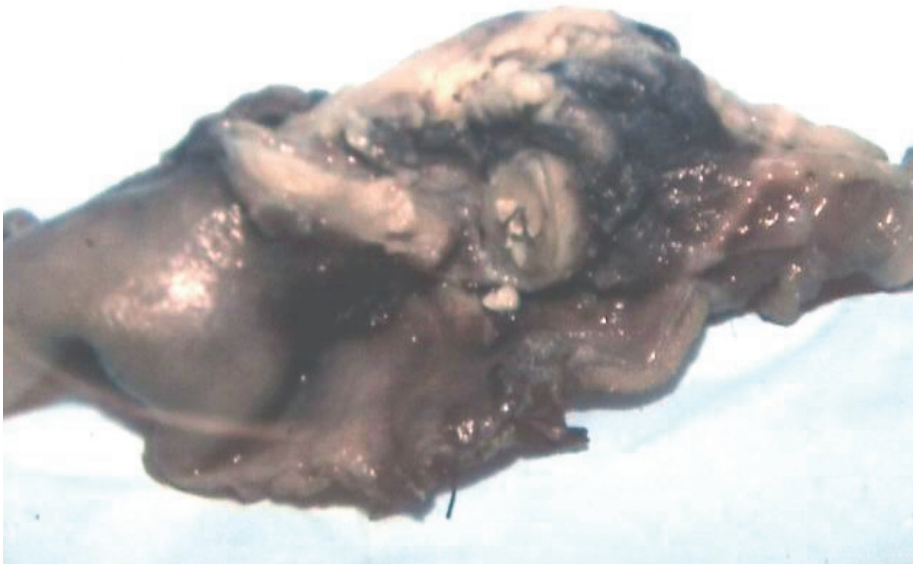


Figure 11. The back of the specimen number 10 showing second duplication cyst * (cut surface).

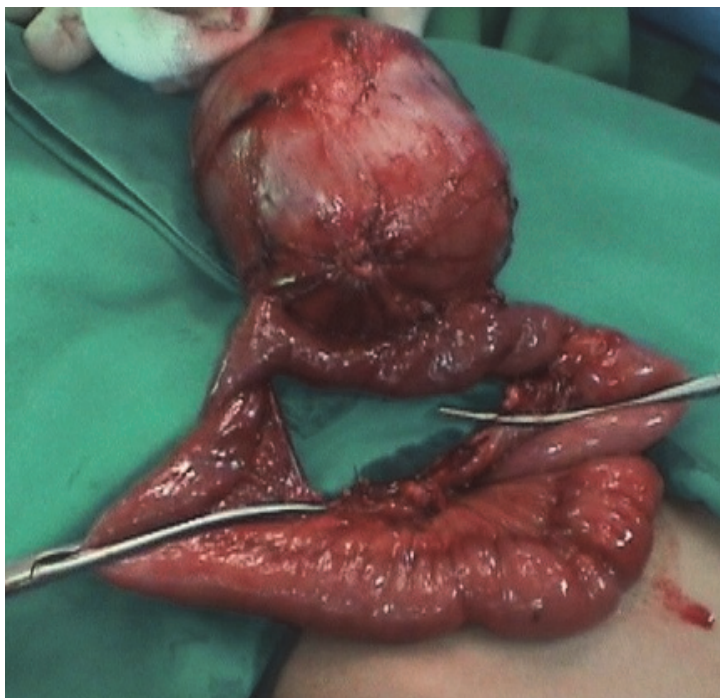


Figure 12. Ileal duplication cyst excised with the adjacent ileum as it shares the same blood supply.



Figure 13. Ileal duplication cyst at the ileocecal junction. Excision of the cyst with the limited ileocecal segment

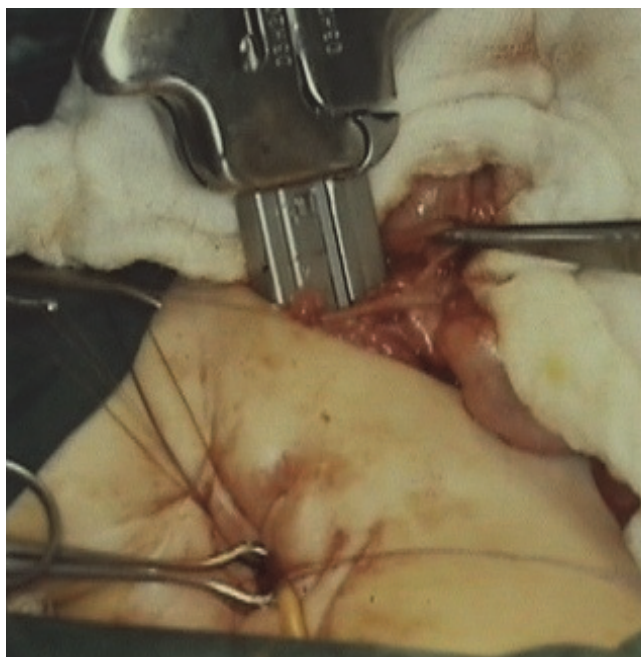
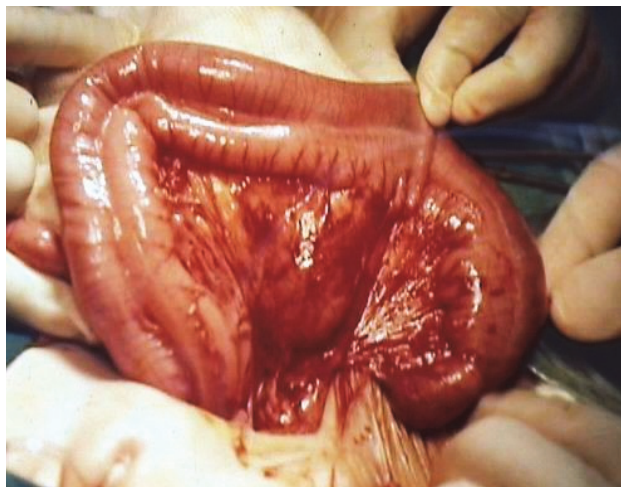


Figure 14. Total colorectal duplications in the patient presented with low recto-vaginal fistula treated Side to side anastomosis of the distal part (recto sigmoid) of the tubular duplication (right) with repair of recto-vaginal fistula.

corresponding segment of the intestine (1 jejunal cyst and 5 ileal cysts all shared the same blood supply of the adjacent intestine). Two duplications were treated with internal drainage by anatomizing the duplication to the adjacent GIT segment (1 duodenal cyst, 1 total tubular colonic duplication). The lining epithelium of all duplications correspond to their level with the exception of one ileal duplication which was lined with gastric epithelium and thus presented with bleeding per rectum for which the patient was treated initially with Cimetidine (antihistamine H₂ blocker), and one esophageal duplication which also was lined with gastric epithelium. The ages of the patients were ranged from neonate to 3 years. The period of hospitalization was 7-12 days. The post-operative follow up period was 6-24 months.

Discussion

Gastrointestinal tract duplications occur anywhere from mouth to anus. It may be either localized cystic or tubular duplications. The cystic type is the most common and usually non-communicating but peptic penetration in the gastric mucosa lined cyst can lead to acquired communication with the adjacent lumen. Seven of the cases were of cystic variety, one of them acquired the communication with the adjacent ileum.

The other three of our cases where tubular duplications might be short or long segment and are usually communicating congenitally either at the proximal or distal or at both ends. The total colorectal duplication represents long duplication which communicate at proximal ends. These morphological variations has been reported previously^[4-12].

The duplication is located on the mesenteric side of the bowel and shares the same blood supply of the adjacent segment of the bowel but exceptions have been reported by Schwartz *et al.*^[13] on the autonomously blood supplied duplication one of the cases where having autonomous blood supply and where treated by simple excision without the bowel resection; similar exception is seen in the cases of esophageal duplications which receives its blood supply from the surrounding structures and not in the form of mesenteric vascular pattern^[14] which makes it possible to excise the duplication without resection of the adjacent esophagus as what we experienced in the case of esophageal duplication.

Previous reports^[15-18] showed that duplication might be associated with other anomalies in the form of another duplication or anomaly. We had a similar experience as the 7th case showed two ileal duplication cysts, while the associated anomalies

were seen as accessory pancreatic lobe (in gastric duplication), rectovaginal fistula (in patient with total colonic duplication) and duodenal atresia (in the case of duodenal duplication). The combination of duodenal duplication with duodenal atresia has not been reported before, but a combination of duodenal atresia with duplication cyst at another location has been reported previously^[17].

Histologically, the duplication have to be lined with gastrointestinal epithelium, but not necessarily that of the adjacent level of the gut^[8-10]. The lining epithelium of duplications of the patients corresponds to the epithelium of the adjacent bowel apart from the 1st esophageal duplication lined with gastric epithelium and the 7th case of the ileal duplication with gastric mucosal lining. Pancreatic ectopic tissue and pancreatic abnormality with or without pancreatitis has been reported previously^[4,18], the 2nd case had accessory pancreatic lobe, which was excised leaving the normal pancreas. Other features of the duplication which were not seen in the 10 young children, such as malignant changes of the lining mucosa and the calcification of the duplication, has been reported previously^[19-21].

Clinical presentation of the duplication varies according to the level, the morphological and histological features of the duplication which was reflected on the frequency of presenting features in the different reports^[14,15,22], but the common symptoms and signs of abdominal pain, abdominal mass and GIT bleeding were the presenting features in 6 (60%) of our patients. Patients with duplication might present with intestinal obstructive symptoms which might be due to the pressure on the lumen or segmental volvulus or intussusception or due to the associated atresia as in our case of duodenal duplication with the duodenal atresia. Thoracic duplication might present with respiratory symptoms or feeding problems as in our esophageal duplication where the patient presented due to repeated chest infection. Other less frequently reported presentations include the pyloric duplication mimicking congenital pyloric stenosis^[23] or entero urinary fistula^[22]. Our 10th case was total colorectal duplication with low rectovaginal fistula which presented due to passage of stool per vagina.

Awareness of gastrointestinal duplication and its clinical presentation is important for the suspicion of the diagnosis. In these cases the diagnosis of a duplication was established preoperatively in 60% of our patients while the other 40% was not diagnosed

preoperatively due to the mode of presentation which suggested a different diagnosis.

The conventional modalities such as plain x-ray abdomen or contrast studies have a minor role in the diagnosis of the gastrointestinal duplication, but the ultrasonic characteristics of the duplication cysts which were reported by Kangaroo and others^[24-27] (hyperechoic inner mucosa, middle echolucent muscle layer and outer hyperechoic posterior enhancement of the cyst) were helpful in establishing the diagnosis with some certainty of our 6 preoperatively diagnosed cases. On the other hand the technetium isotope scan can help in the diagnosis of the duplication which is lined by gastric mucosa as in the 1st and 7th cases^[28-29]. Computerized axial tomography (CT) scan and magnetic resonance imaging (MRI) have a role in suggesting the diagnosis of cystic nature of the duplication particularly in the deep cyst (*i.e.*, pelvic) but it does not have specific diagnostic features of duplication^[4].

Suggested Plan of Treatment

There is no uniform pattern of treatment of gastrointestinal duplication, but the options should be within the solid surgical roles taking into consideration the length of the segment involved and its blood supply, presenting problem, *i.e.* bleeding, obstruction, malignant changes, the level of the duplication and any associated abnormalities such as atresia or rectovaginal fistula. From our experience and other reports, the following are general outlines for the treatment of duplication.

Excision of the Duplication

This line of treatment is suitable, firstly in the cases of esophageal duplication as the duplication can be excised leaving a normal esophagus as in case 1. Secondly, in the cases of a gastric duplication, this duplication can be excised with a rim of the stomach and primary closure of the stomach (as in case 2) or by near total excision of the cyst with stripping of the mucosa of the remaining strip of the cyst. This second technique has been reported by White to improve the surgical outcome^[30]. Thirdly, the cystic or short tubular intestinal duplication which should be excised with the adjacent bowel as it shares the same blood supply^[22] with the exception of the loop which has autonomous blood supply^[13] which can be excised with preservation of the adjacent bowel.

Internal Drainage

This option is suitable in the long tubular duplication (as in the 10th case of the total colorectal duplication) or where important structures might get involved in the dissection such as bile and pancreatic duct with duodenal duplication (as in the 3rd case). The internal drainage is achieved by wide anastomosis to the adjacent bowel or by cystoenterostomy to loop of bowel^[31] or Roux-en-Y manner^[32].

Stripping of the Ulcerogenic Mucosa

This technique is applicable when the duplication is lined with ulcerogenic gastric mucosa and can't be excised^[33], such as the duplication which cannot be excised due to technical difficulties (posteromedial duodenal duplication), or involvement of long segment of small bowel such as in the case of Jewett of the total small bowel duplication^[10]. An antihistaminic H₂-blocker, *e.g.*, Cimetidine or hydrogen pump inhibitor *e.g.*, omeprazole can be used in the duplication with ulcerogenic mucosa to control the ulcer symptom in conjunction with the internal drainage or for control of the symptom (pain and bleeding) or as emergency treatment. Any associated anomalies should be treated in the standard way of treatment of such anomalies.

Conclusion

The management of the duplication is variable due to the wide variation of the anatomical and histological features, the location of the duplication and the associated abnormalities.

Presented here is our experience in the management of 10 cases of GIT duplication with different levels and presentations with suggested general plan for the management of the different types of the GIT duplication based on the management of the 10 cases and the previous reports.

Conflict of Interest

The author has no conflict of interest.

Disclosure

The author did not receive any type of commercial support either in forms of compensation or financial for this study. The author has no financial interest in any of the products or devices, or drugs mentioned in this article.

Ethical Approval

Obtained.

References

- [1] FITZ RH. Persistent omphalomesenteric remains: their importance of the causation of intestinal duplication, cyst formation and obstruction. *Am J Med Sci* 1884; 88: 30.
- [2] LADD WE. Duplications of the alimentary tract. *South Med J* 1937; 30: 363.
- [3] ROWLING JT. Some observations on gastric cysts. *Br J Surg* 1959; 46(199): 441-445.
- [4] Macpherson RI. Gastrointestinal tract duplications: Clinical, pathological, etiological and radiological considerations. *Radiographics* 1993; 13: 1063-1080.
- [5] Pintér AB, Schubert W, Szemlédy F, Göbel P, Schäfer J, Kustos G. Alimentary tract duplications in infants and children. *Eur J Pediatr Surg* 1992; 2(1): 8-12.
- [6] Kuo HC, Lee HC, Shin CH, Sheu JC, Chang PY, Wang NL. Clinical spectrum of alimentary tract duplication in children. *Acta Paediatr Taiwan* 2004; 45(2): 85-88.
- [7] Olajide AR, Yisau AA, Abdulraseed NA, Kashim IO, Olaniyi AJ, Morohunfade AO. Gastrointestinal duplications: experience in seven children and a review of the literature. *Saudi J Gastroenterol*. 2010; 16(2): 105-109.
- [8] Bower RJ, Sieber WK, Kiesewetter WB. Alimentary tract duplication in children. *Ann Surg* 1978; 188(5): 669-674.
- [9] Agha FP, Gabriele OF, Abdulla FH. Complete gastric duplication. *AJR Am J Roentgenol* 1981; 137(2): 406-407.
- [10] Jewett TC Jr, Walker AB, Cooney DR. A long term follow-up on a duplication of the entire small intestine treated by gastroduplication. *J Pediatr Surg* 1983; 18(2): 185-188.
- [11] Soper RT. Tubular duplication of the colon and distal ileum: case report and discussion. *Surgery* 1968; 63(6): 998-1004.
- [12] Sharma S, Yadav AK, Mandal AK, Zaheer S, Yadav DK, Samie A. Enteric duplication cysts in children: a clinicopathological dilemma. *J Clin Diagn Res* 2015; 9(8): EC08-11.
- [13] Schwartz DL, Becker JM, Schneider KM, So HB. Tubular duplication with autonomous blood supply: Resection with preservation of adjacent bowel. *J Pediatr Surg*. 1980; 15(3): 341-342.
- [14] MELLISH RW, KOOP EE. Clinical manifestations of duplication of the bowel. *Pediatrics* 1961; 27: 397.
- [15] Wiczorek RL, Seidman I, Ranson JH, Ruoff M. Congenital duplication of the stomach: case report and review of the English literature. *Am J Gastroenterol* 1984; 79(8): 597-602.
- [16] Sherman NJ, Morrow D, Asch M. A triple duplication of the alimentary tract. *J Pediatr Surg* 1978; 13(2): 187-188.
- [17] al-Salem AH, Khwaja MS. Three faces of midgut duplication. *Int Surg* 1990; 75(1): 47-49.

- [18] Spence RK, Schnauffer L, Mahboubi S. Coexistent gastric duplications embryogenesis and treatment. *J Pediatr Surg* 1986; 21(2): 68-70.
- [19] Orr MM, Edwards AJ. Neoplastic change in duplications of the alimentary tract. *Br J Surg* 1975; 62: 269.
- [20] Hickey WF, Corson JM. Squamous cell carcinoma arising in a duplication of the colon: case report and literature review of squamous cell carcinoma of the colon and of malignancy complicating colonic duplication. *Cancer* 1981; 47(3): 602-609.
- [21] Omojola MF, Hood IC, Stevenson GW. Calcified gastric duplication. *Gastrointest Radiol* 1980; 5(3): 235-238.
- [22] Ravitch MM. Duplications of gastrointestinal tract. In: *Pediatric Surgery*. Welch KJ, Randolph JG, Ravitch MM, Mustard WT, eds. Chicago, IL: Year Book Medical, 1986. 911-920.
- [23] Grosfeld JL, Boles ET Jr, Reiner C. Duplication of pylorus in the newborn: a rare cause of gastric outlet obstruction. *J Pediatr Surg* 1970; 5(3): 365-369.
- [24] Kangaroo H, Sample WF, Hansen G, Robinson JS, Sarti D. Ultrasonic evaluation of abdominal gastrointestinal tract duplication in children. *Radiology* 1979; 131(1): 191-194.
- [25] Teele RL, Henschke CI, Tapper D. The radiographic and ultrasonic evaluation of enteric duplication cysts. *Pediatr Radiol* 1980; 10(1): 9-14.
- [26] Moccia WA, Astacio JE, Kaude JV. Ultrasonic demonstration of gastric duplication in infancy. *Pediatr Radiol* 1981; 11(1): 52-54.
- [27] Lamont AC, Starinsky R, Cremin BJ. Ultrasonic diagnosis of duplication cysts in children. *Br J Radiol* 1984; 57(678): 463-467.
- [28] Ferguson CC, Young LN, Sutherland JB, Macpherson RI. Intrathoracic gastrogenic cyst-preoperative diagnosis by technetium pertechnetate scan. *J Pediatr Surg* 1973; 8(5): 827-828.
- [29] Rodgers BM, Youssef S. "False positive" scan for Meckel diverticulum. *J Pediatr* 1975; 87(2): 239-240.
- [30] White JJ, Morgan WW. Improved operative technique for gastric duplication. *Surgery* 1970; 67: 522-526.
- [31] Gardner CE Jr, Hart D. Enterogenous cysts of the duodenum; report of a case and review of the literature. *JAMA* 1935; 104(20): 1809-1812.
- [32] Leenders EL, Osman MZ, Sukarochana K. Treatment of duodenal duplication with international review. *Am Surg* 1970; 36(6): 368-371.
- [33] WRENN EL JR. Tubular duplication of the small intestines. *Surgery* 1962; 52: 494-498.

التضاعف المعدي المعوي: خبرة ١٠ سنوات

مازن عمر كردي

قسم الجراحة، شعبة جراحة الأطفال
كلية الطب، جامعة الملك عبدالعزيز
جدة - المملكة العربية السعودية

المستخلص. يُعتبر التضاعف المعدي المعوي تشوهاً خلقياً نادراً. على مدار العشر سنوات الأخيرة، تمت معالجة ١٠ حالات تضاعف الجهاز الهضمي لدى مستشفى جامعة الملك عبدالعزيز بجدة، المملكة العربية السعودية. واختلف كل تضاعف في مستواه في اعراض ظهوره. في هذا البحث، سيتم عرض خبرة معالجة مختلف مستويات تضاعف الجهاز الهضمي إضافة إلى اقتراح خطة عامة لمعالجة التضاعف المعدي المعوي بناء على خبرتنا والخبرة الجماعية الواردة في تقارير العديد من الأبحاث الأخرى.