

Intestinal Duplications

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INTRODUCTION

Duplications of the alimentary tract are unusual congenital anomalies that present a diagnostic as well as therapeutic challenge to the treating surgeon. These lesions occur infrequently and are generally not suspected until encountered intraoperatively. Due to the complicated anatomy and pathological involvement of the adjacent bowel, appropriate management requires a familiarity with the anatomy and clinicopathological characteristics of this condition.

SYNONYMS AND HISTORY

The term intestinal duplication has been assigned several names which include enterocystomas, enterogenous cysts, supernumerary accessory organs, ileum duplex, giant diverticula and unusual Meckel diverticulum.¹ Reginald Fitz first used the term intestinal duplication in 1884,² but it was not used widely until popularized by Ladd in 1937,³ and classified by Gross in the 1950s.⁴ Ladd recommended that the term 'alimentary tract duplications' be applied to those congenital malformations that arise on the mesenteric side of the involved alimentary tract and share a common blood supply with the bowel. His observations consolidated the classification of this entity and differentiated it from other cystic malformations of the alimentary tract.

DEFINITION²

Intestinal duplications consist of a group of congenital anomalies characterized by a well developed coat of smooth muscle, an epithelial lining representing mucous membrane of the normal gastrointestinal (GI) tract and frequently intimately attached to or communicating with some portion of the GI tract.

INCIDENCE

The current literature consists of scattered case reports or small case series. No centre has a large and long series to ascertain the actual incidence. In 1961 Potter reported 2 cases in more than 9000 foetal and neonatal autopsies.⁵ The small intestine is the most frequently involved, whereas gastric, duodenal, rectal and thoracoabdominal involvement is relatively rare. In one series of 28 children, thoracoabdominal duplication cysts were encountered in 20% of patients.⁶ Synchronous GI duplications occur in up to 15% of patients.⁷

AETIOLOGY

The aetiology of alimentary tract duplications has not been well established. Hypotheses have included persistence of *embryonic diverticula* during development of the alimentary tract, intrauterine *vascular accidents* and *recanalization and fusion* of the embryological longitudinal folds.⁸ *Abortive twinning* has also been proposed as a possible causative factor of extensive complete duplications of the colon and genitourinary system, which occur occasionally. A hypothesis has been proposed to explain such an occurrence as an initial developmental abnormality in the gastrulation stage which results in a *split notochord* in the foetus. During early embryogenesis, the notochord is open so that the endoderm of the yolk sac and the ectoderm of the notochord are fused and the neuroenteric canal connects the yolk sac with the amnion. As part of the development of the split notochord, an endo–ectodermal adhesion between the cord and the yolk sac has been proposed to result in the persistence of an endomesenchymal tract between the yolk sac and amnion. Thus, the endomesenchymal tract has been held responsible for the anomalies of the entire GI system. However, not all duplications are compatible with this theory.

CLINICAL PRESENTATION

Although most duplications are diagnosed incidentally, their symptomatic presentation depends upon the size, communication with the gut and location:

1. *Cervical, lingual and hypopharyngeal duplications* present with respiratory distress that may be life-threatening and may require rapid diagnosis and treatment.
2. *Thoracic and thoracoabdominal duplications* may cause respiratory distress by airway or lung compression in younger children. However, in older patients, heartburn or melaena has been reported, which is probably caused by the presence of gastric mucosa in one-third of the patients with thoracic and thoracoabdominal duplications. Haemoptysis, a very unusual presentation due to the duplication cyst communicating with a bronchus or being located within the lung parenchyma, has also been reported.⁹
3. *Gastric duplications* usually present (in children <1 year of age) with vomiting, poor feeding, failure to gain weight and a palpable mass on physical examination. Hypertrophic pyloric stenosis is a common misdiagnosis in such infants. The mucosal lining of the cysts often resembles gastric epithelium and may result in melaena or haematemesis due to acid peptic disease. A case has been reported in which the initial presenting symptom in an adult was an infiltrating adenocarcinoma arising in a duplication cyst of the stomach.¹⁰
4. *Duodenal duplications* contain ectopic gastric mucosa in up to 15% of cases,¹¹ which predisposes the patient to peptic ulcer syndrome-like features leading to painless GI haemorrhage and/or perforation. Duplications that extend into the liver¹¹ are generally diagnosed after the onset of high intestinal obstruction or haemorrhage that may be accompanied by icterus or pancreatitis. Transdiaphragmatic extensions of such lesions are known, which mandate investigation of both the chest and the abdomen in case one is found in either location. Duodenal duplications are also known to present as acute pancreatitis,

pseudopancreatic cysts, obstructive jaundice and rarely with elevated carcinoembryonic antigen (CEA) levels.

5. *Small intestinal duplications*: The clinical presentation depends on the type, size, location and mucosal lining of the duplication and whether it is in communication with the intestine. Small cystic duplications can be a lead point for intussusceptions¹² or can result in volvulus, whereas long tubular duplications with proximal communication drain poorly, and retention of intestinal contents leading to massive distention can obstruct the adjacent intestine by compression and/or volvulus. Distal communication is more common and more difficult to diagnose than the proximally communicating variety because of a paucity of symptoms. Gastric mucosa in a duplication can lead to ulceration and perforation. The diagnosis is often not established before surgery.

6. Colonic duplications

i. Cystic colonic duplications are either asymptomatic or present as abdominal masses that may be accompanied by pain. Bleeding may be observed despite a lower prevalence of ectopic gastric mucosa than in other duplications. Newborns may present with volvulus or acute intestinal obstruction resulting from a weighty mass forming an axis.

ii. Tubular colonic duplications are usually asymptomatic, but may present with duplicated genitalia or sometimes a second anal orifice behind the normal one.

7. *Rectal duplications*: The presenting features of rectal or presacral duplications may include constipation, rectal bleeding, haematochezia, rectal prolapse, haemorrhoids, fistula-in-ano and perirectal abscess, besides presenting as a mass behind the anus. Such duplications have been seen in adults with malignant transformation.

In summary, intermittent vomiting, especially in infants less than 1 year of age, is seen in about 50%, abdominal pain 50%, abdominal distension 30%, palpable mass 20%, peritoneal signs 13%, blood mixed with stools 6%, fever 6% and constipation 6%.¹³

DISTRIBUTION

Approximately 75% of duplications have been reported to be located within the abdominal cavity, while the remaining are intrathoracic (20%) or thoracoabdominal (5%). Jejunal and ileal lesions are the most commonly encountered abdominal lesions (53%) followed by mediastinal (18%), colonic (13%), gastric (7%), duodenal (6%), rectal (4%), thoracoabdominal (2%) and cervical (1%).¹⁴

CLASSIFICATION

Based on physical appearance

Seventy-five per cent of duplications are cystic, with no communication with the adjacent intestine, while the remaining are tubular structures that may or may not have one or more direct communications through a common septum. Usually such communications are located at the distal end.

*Pathological classification*¹⁵

1. Parenteral cystic type (in the mesentery, cystic, most common)
2. Parenteral canal type (in the mesentery, tubular, second commonest)
3. Parietal cyst type (intramural, third commonest)
4. Enteral septum type (intraluminal due to septae, rare)
5. Solitary type (seems pedunculated, not intimately attached to the gut, rare)

*Vascular classification*¹⁶

Type 1: Parallel type, duplication is on the mesenteric border of the intestine and has an independent blood supply through the vasa recta which is separated from the straight artery of the bowel. It is not covered by mesentery. Therefore, such duplications can be resected easily without compromising the adjoining bowel.

Type 2: Intramesenteric type, the duplication is in between the leaves of the mesentery and the straight arteries arch over the duplication to reach the bowel. It is sometimes technically demanding to resect such duplications without compromising/sacrificing the adjoining bowel as the vasculature is sometimes common to both or because it is difficult to separate the vessels from the cyst.

GROSS PATHOLOGY

All intestinal duplications contain at least one layer of smooth muscle and some type of intestinal mucosal layer lining the lumen. These cysts are often attached intimately to the adjacent segment of the normal GI tract from which they may be difficult to separate, especially if there has been an infective episode. As mentioned previously, the mucosal lining within the duplicated gut does not necessarily correspond with that of the adjacent normal intestine. It may display components of several different types of GI tract mucosa or at times respiratory or squamous epithelium,¹⁷ especially in lingual or, at times, cervical duplication cysts. Non-communicating duplications typically contain clear alkaline fluid, except in those cases where gastric mucosa is present (25%) which secretes acidic fluid. In addition, non-activated pancreatic enzymes may also be observed in those cases where ectopic pancreatic tissue is present within the duplication.¹⁸

The *histochemical pattern* was studied in a group of alimentary tract duplications ($n=12$) using special stains such as PAS, AB-PAS, and high iron diamine-AB stains.¹⁹ Eleven of these duplications had gastric mucosa in varying stages of maturation. Two had small gut and bronchial wall mucosa in addition. One intramucosal rectal cyst was lined exclusively with primitive rectal mucosa. The cysts showed a variable pattern of mucin histochemistry ranging from neutral mucin to sulpho- and sialomucins. The authors also did a correlation between the type of mucin and the age of the patients. Older infants (>7 months of age) had neutral mucins or focal positivity for sulphomucins, whereas younger infants (<1 month of age) had a mixture of sulpho-, sialo- and neutral mucins.

SITE-SPECIFIC FEATURES

Cervical duplications

These are generally duplications of the oesophagus, contain clear mucoid fluid and may be associated with vertebral anomalies.

Thoracic and thoracoabdominal duplications

As many as one-third of these lesions have a second or third duplication cyst below the diaphragm¹ which may be in continuity with or detached from the primary lesion. Therefore, diagnostic imaging should always include the abdomen (Fig. 1). Almost all patients with thoracic and thoracoabdominal duplications have associated vertebral anomalies and, in some instances, the spinal cord may also be involved.



FIG 1. Thoracic duplication of the oesophagus with anterior spinal defect in a 6-month-old male presenting with respiratory distress. Thoracotomy, excision of the cyst and repair of an anterior meningocele was done.

Gastric duplications

These are generally cystic and are located on the greater curvature. They do not communicate with the lumen of the stomach.

Duodenal duplications

These duplications generally do not communicate with the intestinal lumen. Duodenal duplications can also arise from the bile ducts or the pancreas.

Small intestinal duplications

Most small intestinal duplications are located adjacent to the distal small bowel. These may be cystic or tubular and are situated on the mesenteric border, often sharing a common muscular wall and blood supply with the native intestine. Multiple small intestinal duplications may be present.

Colonic duplications

Colonic duplication cysts may be isolated or have an external fistula to the skin, urinary tract or normal colon. Tubular duplication of the colon is often associated with

duplication of the anus or external genitalia. Kottra and Dodds²⁰ suggested a classification of colonic duplications which provides practical guidance for their management.

Type I: Duplications limited to the alimentary tract and situated above the peritoneal reflection. These could be cystic (type Ia), tubular (type Ib) or of the double-barrel type (Ic) with one or multiple communication(s) with the gut.

Type II: These are tubular colonic duplications associated with duplications of the genitourinary tract.

Ila: Tubular duplications with double genitalia, double urethra and bladder. Separate perineal ani are present on either side of the midline. There is no communication with the genitourinary system.

Ilb: An internal fistula is present between the duplication and the genitourinary system.

Ilc: Tubular duplications with imperforate anus. There is no internal fistula between the genitourinary tract and the duplication. However, the lower urinary tract may be duplicated.

Rectal duplications

Rectal duplications occur in the retrorectal space. These may present as chronic constipation or occasionally with recurrent perianal sepsis.²¹

The following *associated anomalies* are common with duplications: Approximately 15%–50% of alimentary duplications have associated anomalies.^{22,23} Vertebral anomalies are seen in mediastinal (15%) and thoracoabdominal (50%) duplications. Almost all neuroenteric cysts have vertebral defects with varying degrees of intraspinal extension. Tubular hind gut duplications, especially those extending below the line of peritoneal reflection, are associated with genitourinary malformations.

PREOPERATIVE EVALUATION

Imaging studies

1. Plain X-rays of the chest in different views may reveal a space-occupying lesion in the mediastinum, deviation or distortion of the air column in the trachea, and oesophageal shadows and vertebral defects (Fig. 2). Thoracic duplications are often discovered incidentally on chest X-rays done for some other complaint. They have a characteristic enhancing ring that can be detected by contrast-enhanced CT or MRI scan.
2. Contrast studies are helpful in demonstrating the mass effect and displacement of normal alignment of adjacent structures.
3. In cases of GI bleeding, heterotopic gastric tissue can be detected by radioisotope-labelled technetium scans.
4. Ultrasonography is increasingly being used for the diagnosis of abdominal duplications. It has the added benefit of revealing associated genitourinary or other intra-abdominal anomalies. Recently, endoscopic ultrasonography has been used

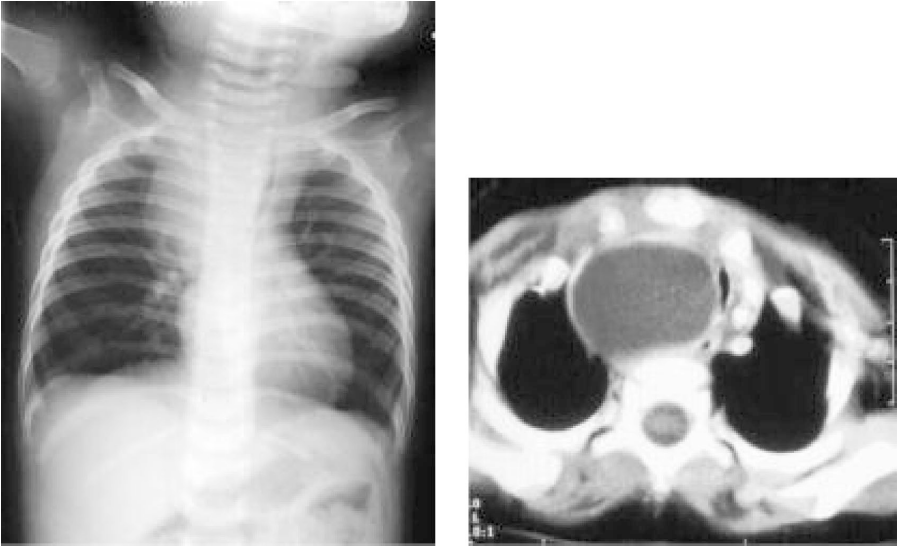


FIG 2. Plain X-ray showing upper mediastinal widening and vertebral defects. CT scan thorax shows an enhancing rim of an oesophageal duplication cyst. This was excised by right thoracotomy.

to accurately delineate the lesion especially when dissection of the oesophagus is suspected.

5. CT scan: Multidetector CT of the chest or abdomen is useful in establishing a diagnosis of alimentary tract duplication during the preoperative work up and may be used to evaluate synchronous distant lesions once a single duplication has been identified.
6. MRI: It is preferred over CT scan as it avoids radiation to young patients. It may be necessary if neurological symptoms of spinal cord compression/involvement exist or a vertebral anomaly is seen which may be indicative of intraspinal cysts. In young patients it may be difficult to perform, as the child needs to be heavily sedated to avoid movement artifacts.
7. Magnetic resonance cholangiopancreatography (MRCP): It is useful in differentiating a duodenal duplication from a choledochal cyst and in planning further management.
8. Genitogram and/or cystourethrogram may be indicated in hind gut duplications with associated anomalies.
9. Prenatal ultrasonography occasionally detects these enteric cysts, especially those that are large in size. These have been treated successfully by aspiration to facilitate delivery.

Endoscopy

1. Endoscopic retrograde cholangiopancreatography (ERCP) is useful for duodenal duplications. However, this facility may not be available for children in a large

number of centres. It has now been largely replaced by MRCP.

2. Bronchoscopy may be done to evaluate the trachea in cervical oesophageal duplications, especially those presenting with haemoptysis.
3. Genitoscopy and cystoscopy are indicated in hind gut duplications with associated genitourinary malformations.

WORK UP IN A SUSPECTED CASE OF DUPLICATION

Children with space-occupying lesions in the chest and an associated vertebral defect, with obscure GI bleeding or recurrent subacute intestinal obstruction (recurrent volvulus) should be suspected of having intestinal duplications (Figs 3 and 4). CT scan and technitium pertechnetate scan are the most useful preoperative investigations.^{24,25}

Recently, laparoscopy has been advocated as a useful diagnostic and therapeutic tool in patients with mediastinal duplication with transdiaphragmatic extension to rule out or excise any intra-abdominal cyst, or in the evaluation of obscure GI bleeding. (If duplication cyst/Meckel diverticulum is found to be the cause, it can be treated by laparoscopy.)²⁶



FIG 3. Cystic ileal duplication causing intestinal obstruction



FIG 4. Long tubular duplication of the small intestine presenting as melaena in a 2-month-old infant.

SURGICAL TREATMENT

Surgical excision or mucosectomy is the treatment of choice for all types of duplication, especially in view of increased reports in the literature about malignant degeneration of this malformation.²⁷⁻³⁰ Irrespective of the location, total excision is the preferred method of treatment for alimentary tract duplications. If this is considered unsafe due to anatomical considerations then one can either do a mucosectomy or internal drainage (to be avoided) provided the gastric tissue, if present, has been removed.

Most duplications share a common blood supply with the normal bowel and, therefore, it may be necessary to perform a segmental resection of the adjacent bowel. In the absence of this, it may be possible to excise or 'shell out' the cyst if an adequate plane of separation exists between the cyst and the mesentery with its blood supply. Mucosal stripping after opening the cyst has also been described for cysts in which attempted excision may endanger vital structures. If excision is not possible, for example, due to proximity to the biliary or pancreatic ducts, an internal drainage procedure commonly employing a Roux-en-Y loop may be done. However, if this is planned, one must determine preoperatively whether gastric mucosa is present (with technetium scan) and, if present, it must be excised to prevent future ulceration. In colonic and rectal duplications below the peritoneal reflection, endoscopy of the urogenital tract is done to rule out associated anomalies.

Site-specific approaches

Cervical oesophageal duplications. Surgical treatment involves excision of the cyst. However, if the cyst cannot be excised, total mucosectomy can be done.

Thoracic and thoracoabdominal duplications. Such duplications occupy the posterior mediastinum. A posterolateral thoracotomy (preferably muscle sparing) and excision of the cyst is desirable. It is a simple procedure and postoperative recovery is usually uneventful. Care should be taken to avoid injury to the oesophagus while shelling out the duplication. In case of difficulty, leaving behind a segment of common musculature is acceptable. In cases where the anatomy is obscure, the cyst is opened and the mucosa stripped; this may be difficult and can be associated with loss of blood. Combined thoracoabdominal cysts comprise <2% of all duplications. If staged excision is planned, or only the cyst in one area is excised, the remaining duplication should be removed early to avoid distension due to reactionary accumulation of secretions/haemorrhage which, at times, can develop very rapidly with life-threatening consequences, especially if within the chest.

Gastric duplications. In most cases, resection can be accomplished without entering the stomach by peeling the cyst off the stomach or resecting the shared wall between the stomach and the duplication. Gastric resection is generally not required. A limited excision of the common wall of the stomach may be indicated in cases of difficult dissection. If the lumen is entered accidentally it can be closed carefully with non-absorbable sutures.

Duodenal duplications. As mentioned earlier, surgical resection is the treatment of choice. However, drainage of duplication cysts into the duodenum or into a Roux-

en-Y limb of jejunum is an acceptable alternative if there is any risk of injury to the biliary or pancreatic ductal system. Preoperative ERCP, percutaneous transhepatic cholangiography (PTC), MRCP or intraoperative cholangiography (more commonly used) can help delineate the exact anatomy, should it be necessary to evaluate the involvement of the biliary/pancreatic ducts. These studies may also distinguish between a duodenal duplication and a choledochal cyst. Excision should be done if gastric mucosa is present to avoid later ulceration. Marsupialization and external drainage, a method of the past, are mentioned to be condemned.

Small intestinal duplications. Segmental resection along with the adjacent intestine is the preferred treatment for small cystic or short tubular duplications. Unresectable cystic duplications (in the absence of gastric mucosa) may be drained into a Roux-en-Y limb. Long tubular duplications that cannot be resected or cannot be separated because of their length can be managed by mucosal stripping through multiple incisions (Wrenn technique),³¹ or diversion into the stomach (Fig. 5).



FIG 5. Mucosal stripping of tubular duplication and resection of the cystic end having intestinal communication. Histopathology showed the presence of ectopic gastric tissue causing haemorrhage.

Cystic colonic duplications. In smaller cysts, a complete excision of the duplication and its attached normal colon is preferred. However, because long colonic duplications generally have a communication with the normal colon, creating another internal communication by excising a small part of the common wall to permit re-entry from the duplication into the normal colon to prevent accumulation of debris, is sufficient.

Tubular cystic duplications. If the duplication has a common wall with a normal rectum and perineal opening, the duplication can be connected transanally by excising part of the common rectal wall through the anus using a stapler. Smaller duplications may also be treated by excising the intervening fistula wall (common wall between cyst and rectum) and making it part of the common rectum. Tubular duplications that extend below the peritoneal reflection and are associated with abnormalities of the genitourinary tract should be evaluated by preoperative cystoscopy and vaginoscopy/hysteroscopy to identify duplication, fistula or other abnormalities of the bladder, urethra or genital tract.

Rectal duplications. The general approach involves transanal exposure of the cyst by incising the posterior rectal mucosa to drain the cyst, and stripping the mucosal

lining. Total excision may also be done through the transanal, posterior sagittal or transcoccygeal (Kraske) approach.

Minimal access surgery

As with many types of surgical interventions, several reports^{32,33} have been published promoting the use of laparoscopy/thoracoscopy for the definitive diagnosis and treatment of alimentary tract duplications. These modalities are particularly helpful in managing transdiaphragmatic thoracoabdominal enteric duplications.³⁴

CONSERVATIVE TREATMENT

The preferred treatment of GI duplications is excision. However, in colonic duplications that have a communication proximally and distally between the duplication and the normal colon, administration of stool softeners and enemas can improve symptoms.

In complex colonic duplications associated with duplications of the genitourinary system, a conservative approach should be taken to maintain an unobstructed lumen of the colon, genitourinary tract and duplication.

COMPLICATIONS AND LONG TERM OUTCOME

Complications include bowel obstruction and haemorrhage. As most intestinal duplications are cystic and appear adjacent to the ileum requiring a limited resection, complications related to surgical intervention are non-specific and include postoperative bleeding, infection and bowel obstruction. However, in patients with large tubular duplications, injury to the normal intestine with resultant short bowel syndrome must be avoided. Other complications include scattered reports of intestinal carcinomas found within duplication cysts.

The long term outcome in a majority of duplications is good. Complex colonic duplications and tubular duplications where a long segment of small bowel has to be excised have a poor outcome as such children are likely to develop short bowel syndrome.

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EDITORIAL COMMENT

Duplications of the intestine are congenital problems. Though commonly seen in infants and children, these may also present in adulthood. The symptoms produced by these lesions are related to their size and nature of the lining epithelium (e.g. gastric mucosa causing bleeding or pancreatic tissue causing pancreatitis). The epithelial lining of the cyst can undergo malignant transformation and hence a complete excision should be done. If it cannot be done safely, all the lining mucosa must be removed. These duplications have been reported in some unusual sites such as the tongue. Awareness of this condition can help make an appropriate diagnosis. With the use of ultrasonography, these lesions can be detected antenatally and managed before delivery or during the neonatal period.