
Surgical Management of Alimentary Tract Duplications

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Alimentary tract duplications are unusual anomalies that may require surgical intervention in the neonate, infant, and occasionally in the older child. The clinical presentation of patients with alimentary tract duplications includes bleeding, abdominal pain, intussusception, and respiratory distress, or it may be an incidental finding on either abdominal examination or chest x-ray. A review of 96 patients with 101 duplications seen over the last 37 years is reported herein. Twenty-one duplications were confined to the thorax; three were thoracoabdominal, and 77 were abdominal. Seventy-four patients presented as infants less than 2 years of age, and 22 patients were older. Ectopic gastric mucosa was found in 21 duplications, and pancreatic tissue was found in five. Seventy-five duplications were cystic and 26 were tubular. Ultrasonography, computed tomography (CT), and myelography are helpful diagnostic tools. Ninety-four of the 96 patients underwent surgical management for their duplications. One duplication was found at necropsy, and one patient was asymptomatic and did not undergo operation. A single death occurred in a 2-day-old infant who had intrauterine volvulus and meconium peritonitis. Management was based on the age and condition of the patient, the location of the lesion, whether it was cystic or tubular and communicating with the true intestinal lumen, and whether it involved one or more anatomic locations. Generally, total excision was preferred, but staged approaches were sometimes necessary.

IN 1884, TWO YEARS BEFORE his classic description of appendicitis, Reginald Fitz used the word *duplication* to describe what he thought were remnants of the omphalomesenteric duct.¹ Thereafter, terms such as *enterogenous cyst*, *ileum duplex*, *giant diverticula*, and *unusual Meckel's diverticulum* were used by different authors to describe the congenital cystic abnormalities of the gastrointestinal (G.I.) tract. Later, Gross and Holcomb² suggested that the term *duplication* be used for all such anomalies, irrespective of their site, morphology or em-

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bryologic derivations, in order to simplify the nomenclature.

Duplications may be found anywhere from mouth to anus. They vary widely in size, are either spherical or tubular, and may communicate with the intestinal tract.^{3,4} G.I. duplications tend to be paramesenteric and share a common muscular wall, although each has its own mucosal lining.⁵ Thoracic duplications may lie adjacent to or distant from the esophagus without sharing a muscular wall.

Duplications arise from disturbances in the embryonic development of the G.I. tract. Several major theories have been proposed for the formation of duplications at various sites, including the aberrant luminal recanalization theory espoused by Bremer⁶ or the diverticular theory of Lewis and Thyng.⁷ Bentley postulated the split notochord theory to explain the formation of neuroenteric duplications and associated vertebral anomalies.⁸ Finally, Mellish and Koop⁹ suggested that environmental factors such as trauma or hypoxia in early fetal development were likely to be responsible when multiple duplications are found in association with anomalies such as malrotation or atresia.

It is imperative to understand the many surgical approaches to duplications depending on their location. Although malignant change has rarely been reported in adult duplications,¹⁰⁻¹³ duplications in children are benign lesions. The surgical procedure should not be more radical than necessary to eliminate the patient's complaints and prevent further recurrence.

Ninety-six infants and children with 101 alimentary tract duplications have been seen at The Children's Hos-

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TABLE 1. Location and Type of 101 Duplications in 96 Patients

Location	Cystic	Tubular	Total
Esophagus	21		21
Thoracoabdominal		3	3
Gastroduodenal	10		10
Jejunal	10	2	12
Ileal	25	10	35
Colonic	8	12	20
Total	74	27	101

pital of Philadelphia since 1950. Thirty-eight of these patients were previously reported in 1961.¹⁴ This report reviews our experience with all 96 patients since additional management recommendations have been made possible.

Patient Material (Table 1)

Fifty-five of our 96 patients were boys and 41 were girls. White patients outnumbered black patients by a five to one ratio (80 vs. 16). Thirty-six patients presented at 1 month of age or less, 38 patients were between 1 month and 2 years of age, and the remaining 22 patients were older. Our oldest patient was 17 years of age (Table 2). Twenty-one duplications were thoracic, 77 were abdominal, and three were thoracoabdominal. Ten duplications in the abdomen were gastroduodenal, twelve were jejunal, and 35 were located in the ileum. Colonic and rectal duplications occurred in 20 instances. Seventy-five duplications were cystic, and 26 were tubular. Nineteen duplications were lined with ectopic gastric mucosa only, three duplications had ectopic pancreatic tissue, and two duplications contained both (Table 3). Duplications were found in association with alimentary tract atresia in eleven patients.

Thoracic Duplications

Seven patients presented as neonates, eleven patients were between 1 month and 2 years of age, and three patients were older than 2 years of age. Nine patients presented with respiratory distress, eight were found to have asymptomatic thoracic masses on routine chest radiographs, and one had a left cervical mass. A 2-day-old infant with esophageal atresia and distal tracheoesophageal fistula was found to have a duplication of the blind proximal esophageal pouch, which was excised. Another patient

TABLE 2. Age of 96 Patients With Duplications at Initial Presentation

Age	No. of Patients
<1 month	36
1-24 months	38
>2 years	22
Total	96

TABLE 3. Location of Ectopic Gastric and Pancreatic Tissue

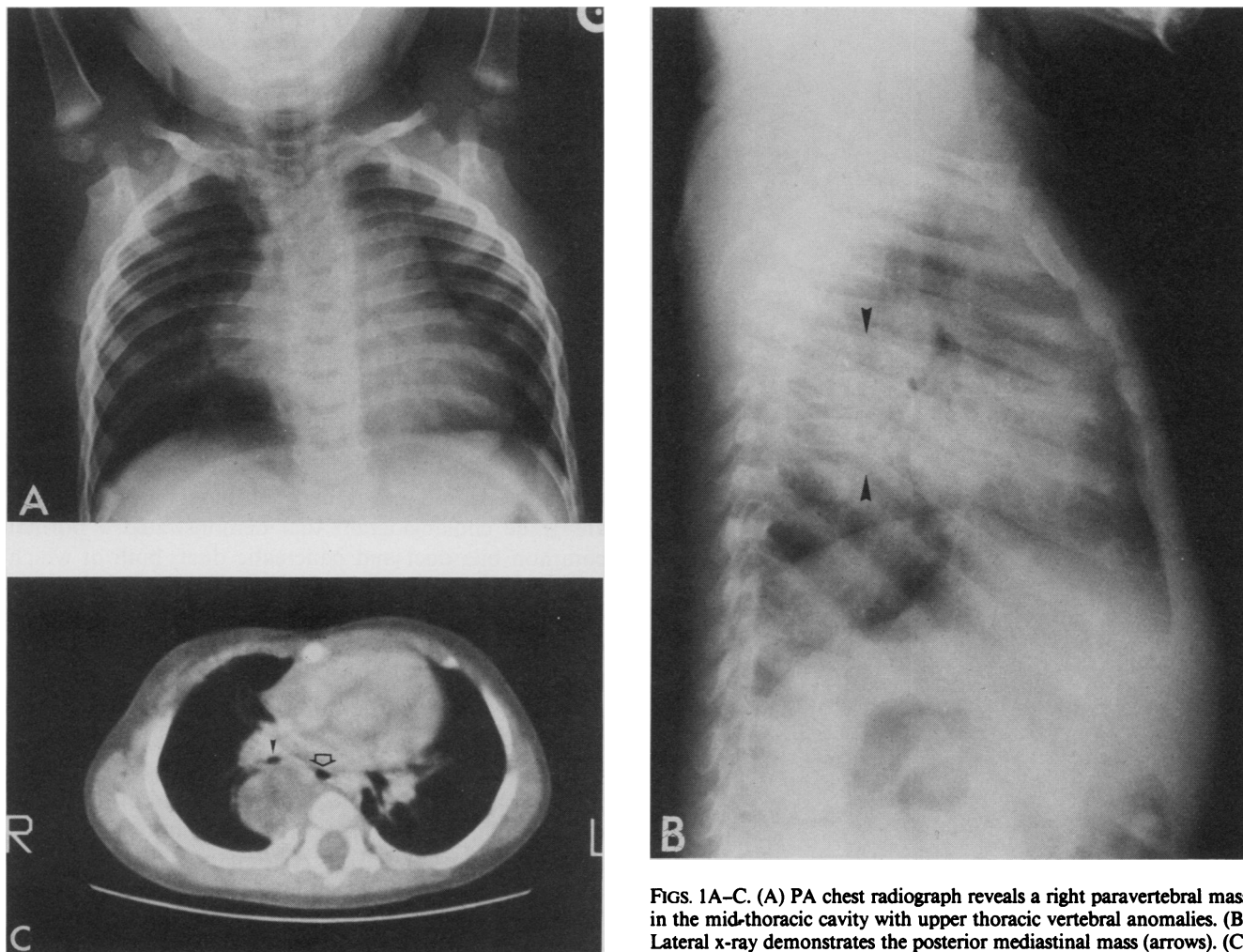
Duplications	No.	Ectopic Tissue	
		Gastric	Pancreatic
Esophageal	21	7	
Thoracoabdominal	3	1	
Gastroduodenal	10		3
Jejunal	12	1	1
Ileal	35	11	
Colonic	20	1	1
Total	101	21	5

presented with abdominal pain and was found to have a thoracic mass with peptic ulceration accounting for his abdominal pain. Barium esophagram at 7 months on a patient who previously had repair of diaphragmatic hernia revealed an esophageal duplication, which was excised.

All 21 thoracic duplications were cystic; histologically all had squamous epithelium. Although five patients had vertebral anomalies, only one communicated with the spinal column. Seven of the duplications contained ectopic gastric mucosa, and ulceration was present in two. In addition, five of the patients with thoracic duplications (25%) also had separate intestinal duplications. All patients underwent excision of the thoracic duplication, except for one who underwent marsupialization. The latter was a 1-month-old infant treated in 1960 who presented with an enlarging mass in the neck obstructing the trachea and necessitating tracheostomy. After stabilization, partial excision of the duplication was undertaken, but the remaining mucosa was firmly adherent to the esophagus, so it was marsupialized. Although an esophageal fistula occurred after operation, gastrostomy and local drainage resulted in closure. No complications were seen in patients with total excision of the duplication.

Comment. Thoracic duplications are usually found in the posterior mediastinum with protrusion into either the right or left thoracic cavity beneath the parietal pleura, with the majority of the lesions in the lower half of the esophagus^{2,15} (Figs. 1A and B). Patients with thoracic duplications usually present either with respiratory distress because of airway compression due to an enlarging mass, or as asymptomatic patients with a thoracic mass found incidentally on x-ray. In the case of infants and children, such duplications should always be considered in the differential diagnosis of mediastinal masses because they are second only to neurogenic tumors as the cause of mediastinal masses in this age group.^{3,16} The association of thoracic duplication and vertebral anomalies ranging from spina bifida to vertebral fusion defects has been well-documented.¹⁷⁻¹⁹

Diagnosis can be established with a barium esophagram showing external compression of the esophagus. However,



FIGS. 1A–C. (A) PA chest radiograph reveals a right paravertebral mass in the mid-thoracic cavity with upper thoracic vertebral anomalies. (B) Lateral x-ray demonstrates the posterior mediastinal mass (arrows). (C) Intravenous contrasted CT scan of the chest shows that the cystic posterior mediastinal mass is contiguous with the esophagus (open arrow) and displacing the right mainstem bronchus (solid arrow). There is contrast enhancement of the wall of the cyst. This picture is characteristic of a thoracic duplication.

computed tomography (CT) has the advantage over conventional diagnostic procedures because it demonstrates the cystic nature of the mass and its relationship to adjacent structures in a noninvasive manner. It permits simultaneous imaging and evaluation of the spine, pulmonary parenchyma, airway, and adjacent structures. The duplication is a smooth, well-marginated circumscribed mass of homogeneous low attenuation (15–30 Hausenfield units) contiguous with the normal esophagus. There is enhancement of the outer rim of the cyst with contrast media. However, the central core does not enhance (Fig. 1C). This peripheral enhancement distinguishes this cyst from a neurogenic tumor.²⁰ Myelography should be employed for those lesions with suspected communication to the vertebral column.

In the past, duplications arising in the thorax have been treated by marsupialization followed by the destruction

of the mucosa by curettage or gauze packing. Because of the many complications involved with this form of management, it has been abandoned in favor of complete excision. In our series, all the lesions were spherical and safely separable from surrounding fissures. Patients with thoracic duplications should have abdominal ultrasonography (US) performed to identify associated intestinal duplications.

Abdominal Duplications

Gastric and Duodenal Duplications

Of the eight patients with gastric duplications, one presented as a neonate, three were between 1 month and 2 years of age, and the remainder were older than 2 years of age. Five were boys and three were girls. All of the duplications were cystic and all presented with either



FIG. 2. UGI (upper G.I.) series reveals a mass effect on the antrum with extrinsic compression of the second portion of the duodenum. US documented a $5 \times 5 \times 4$ cm cystic mass consistent with a gastroduodenal duplication.

nonbilious vomiting or hematemesis. Diagnostic modalities used included barium study and US (Fig. 2).

Five of the eight duplications were completely excised.

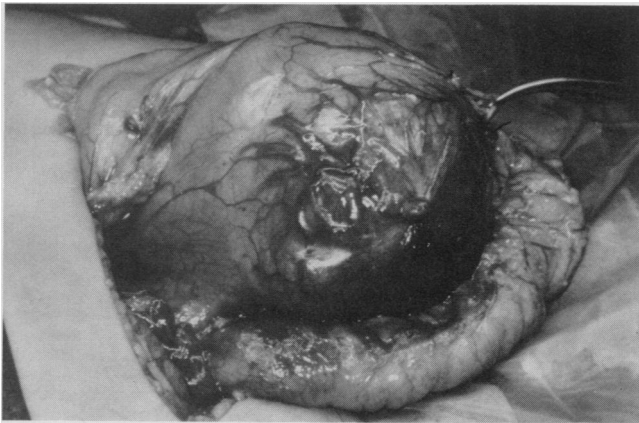


FIG. 3. Operative photograph demonstrates a large cystic duplication of the antrum. This was partially excised with mucosal stripping from the common wall. (See Figure 2 for preoperative UGI).

In two patients, the duplications were partially excised with mucosal stripping from the common wall. One duplication along the greater curvature was managed by internal drainage into the stomach. Currently, all patients are well. In two instances, there was evidence of pancreatic tissue and ducts entering the duplication, but histologically, this was interpreted as ventral pancreas, rather than independent heterotopia. The duplications and pancreatic tissue were both excised, and both patients have remained well without evidence of pancreatitis.

A 3-year-old girl and a 6-year-old boy each presented with vomiting and abdominal pain. Upper G.I. series on the former revealed a duodenal duplication, which was managed by duodenotomy, partial excision, and mucosal stripping. Barium study, US, and endoscopy in the latter patient demonstrated a duplication of the second portion of the duodenum. He underwent duodenotomy and an antegrade cholangiogram that demonstrated a normal common bile duct and pancreatic duct, both of which emptied into the duplication. The duplication was marsupialized, and sphincteroplasty of each duct was performed because the openings appeared to be stenotic. Both patients have done well.

Comment. All gastric duplications in this series were cystic, located along the greater curvature, and did not communicate with the stomach lumen (Fig. 3). Recommended management is complete excision of the duplication without violation of the gastric lumen. If this is not possible, segmental gastrectomy may be needed. Because of a large size or location near the gastroesophageal junction, partial excision with mucosal stripping along the common wall will prevent subsequent ulceration. The stomach should then be distended with air to detect any unsuspected perforation.

Pancreatic tissue and ducts were present on histologic examination in three instances. There have been six previously reported cases of gastroduodenal duplication cysts communicating with the pancreatic ductal system.²¹ In four patients, the duplications were contiguous with the stomach, and in two, the cysts were separate. In patients with contiguous cysts, all had recurrent episodes of pancreatitis. Although none of our patients had documented pancreatitis, one patient experienced intermittent abdominal pain for 18 months, and a 21-month-old had transient feeding intolerance.

Complete resection of duodenal duplications is the preferred treatment, but if this is not possible, marsupialization can be accomplished by internal drainage either into the duodenum or a Roux-en-Y loop of jejunum. Cholangiography is necessary to detect injury to the biliary system.

Jejunal and Ileal Duplications

Twelve patients with jejunal duplications and 35 patients with ileal duplications have been managed. Sixteen

patients presented as neonates, 19 patients were between the ages of 1 month and 2 years, and the remaining twelve patients were older than 2 years of age. Of those 16 patients younger than 1 month of age, 13 presented with signs and symptoms of bowel obstruction, either vomiting (nine), abdominal distention (three), or intussusception (one). Four neonates were found to have a volvulus at the site of the duplication.

Rectal bleeding and intussusception were common presentations in older patients. Twelve patients presented with symptoms of intestinal bleeding. Seven of eight patients with ileal duplications who presented with intestinal bleeding had tubular duplications lined by gastric mucosa with communication to the G.I. tract, but only one of four patients with jejunal duplications and intestinal bleeding had this association (Fig. 4). Ectopic mucosa was present in 14 patients and was gastric in twelve instances. Intestinal bleeding occurred in nine of those twelve patients, and acute abdominal pain that mimicked appendicitis occurred in two.

Four patients with ileal duplications presented with signs and symptoms of intussusception. Barium enema was unsuccessful in reducing intussusception in two patients, and the diagnosis was made upon upper G.I. series in another. All patients with intussusception had a cystic duplication as the lead point.

Associated anomalies included five patients with intestinal atresia, all of whom presented as newborns. Five of the 47 patients with jejunal or ileal duplications had separate thoracic duplication cysts.

All but one patient underwent complete resection of the duplication. This one patient underwent partial resection of an almost complete jejunoileal duplication as a neonate. Six years later, the remaining duplication was resected because of melena. At that time, the remaining normal small bowel had lengthened sufficiently to allow complete resection.

Of the entire series, the only patient who died was a 2-day-old who presented with meconium peritonitis, multiple jejunal atresias, and an associated jejunal duplication. He underwent resection of the duplication and atretic intestine, duodenojejunostomy, and gastrostomy but died in the recovery room. Three patients had complications. Two neonates developed symptoms of small bowel obstruction after resection of the duplication, but recovered without laparotomy, and an anastomotic leak occurred in a 5-day-old after jejunal resection. Anastomotic resection and reanastomosis was performed, and she recovered.

Comment. The small intestine is the most common location for alimentary tract duplications. Five of our duplications were associated with intestinal atresias. This has been noted by other authors^{10,22} and has led to speculation that some environmental stress or vascular accident may be a likely etiology for these duplications. Because most duplications involving the jejunum and ileum

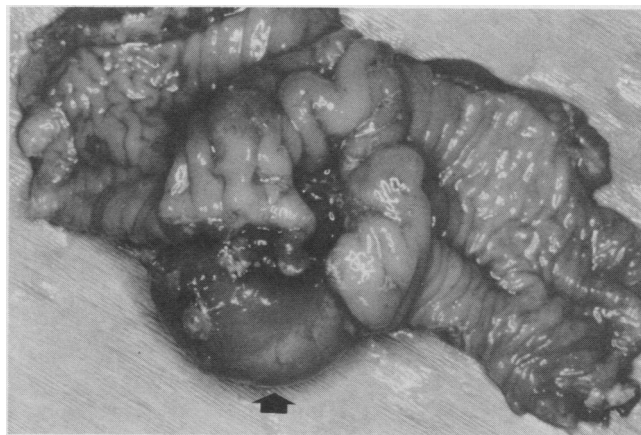


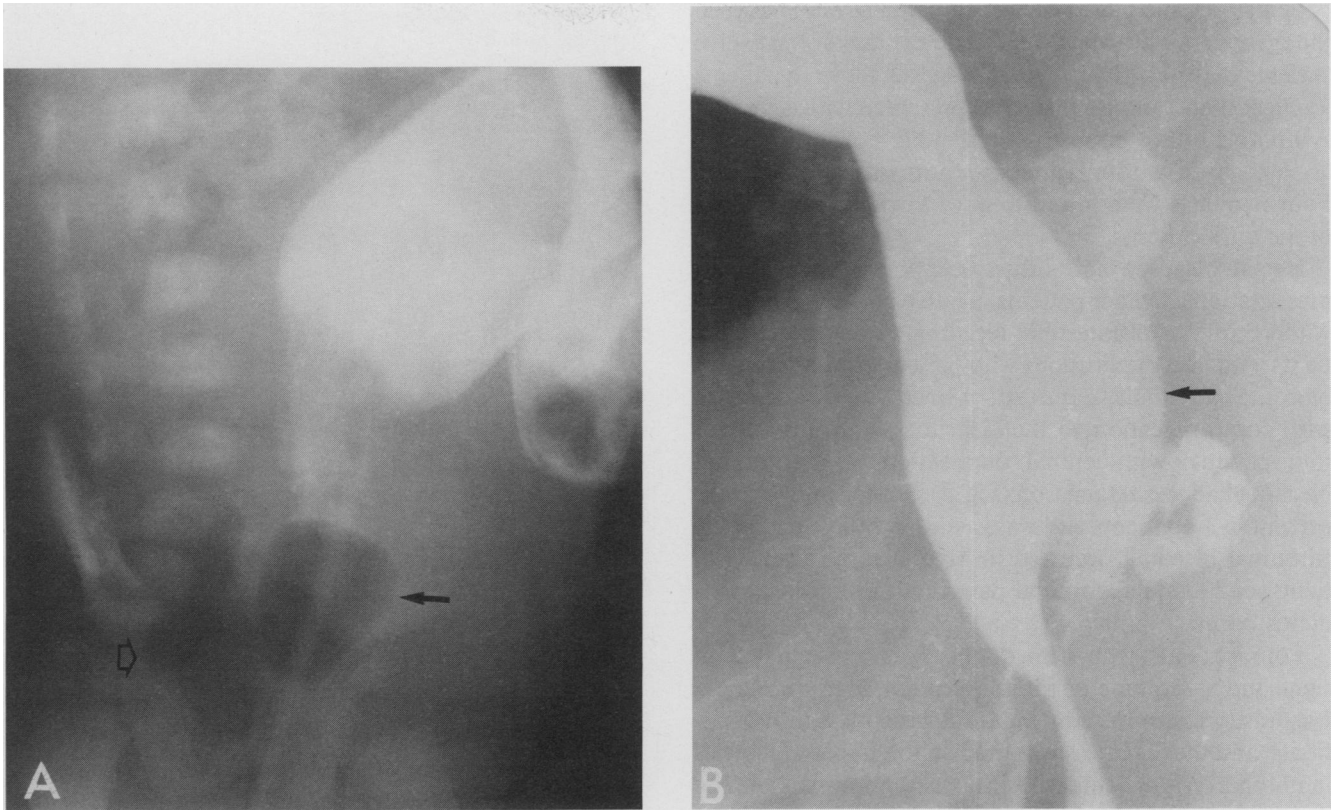
FIG. 4. Whereas most intestinal bleeding from duplications is caused by tubular duplications with communication to the intestine, this patient's bleeding was due to mucosal ulceration from an adjacent cystic duplication (arrow).

are small cystic lesions, they can easily be resected along with a portion of the adjacent intestine. In certain circumstances, such as complete duplication of the small bowel, partial resection is indicated with internal drainage at the distal end. One of our infants was treated this way, but 6 years later symptoms recurred, and the remaining duplication was then removed. This case illustrates the necessity of removing the mucosa of the duplication when complete excision is not possible (except for tubular colonic duplications, because the latter rarely contains heterotopic mucosa). When mucosa remains, acid peptic ulceration from ectopic gastric mucosa leads to bleeding and may require a second operation.

Colonic and Rectal Duplications

Twelve patients were younger than 1 month of age, six were between 1 month and 2 years of age, and two were older than 2 years of age. Fifteen patients presented with colonic duplications, and five with rectal duplications. In the colonic group, seven duplications were isolated to one colonic segment; five were cystic and two were tubular. One was in the appendix, three were limited to the cecum or ascending colon, two were in the transverse colon, and one was in the descending colon. Two other patients with cloacal exstrophy had duplicated short colons. An additional six patients had tubular duplications occupying more than one colonic segment. Two of these were in association with heteropagus conjoined twins. All but two tubular duplications communicated with the colon at one end or the other.

Patients with isolated colonic duplications had varied presentations. Two presented with vomiting and one with volvulus. One each was seen with gastroschisis, urinary retention, and at postmortem examination. An incidental



FIGS. 5A and B. (A) This newborn presented with marked abdominal distention. A #8 Foley catheter was placed in the rectum and the balloon was inflated with air (solid arrow). Posterior to the rectum and compressing it is a rectal duplication with air, indicating communication to the G.I. tract (open arrow). A colostomy was performed because of the rectal obstruction. (B) At 6 months of age, barium enema was performed in this same patient. On this lateral radiograph, there is filling of the posterior rectal mass (arrow).

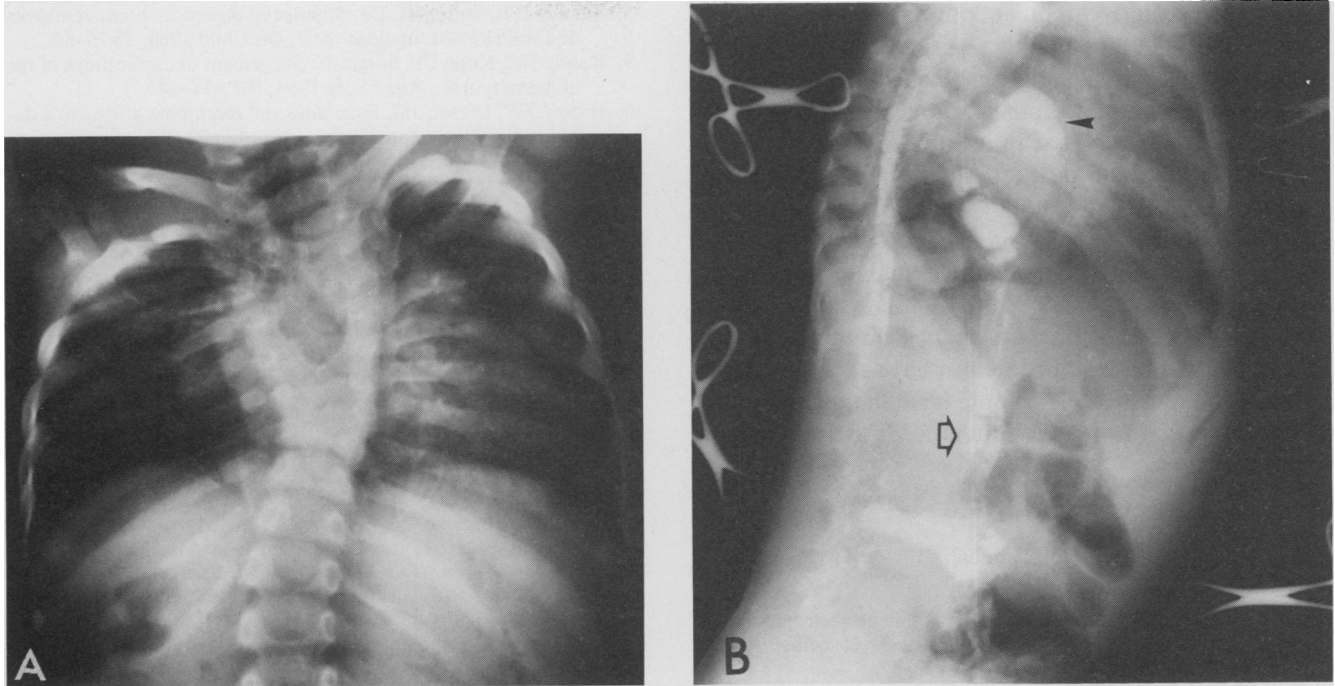
appendiceal duplication was removed at the time of ureteral reimplantation. Only two patients with long tubular duplications were symptomatic. One presented with volvulus and one with megacolon. None presented with bleeding.

Treatment varied according to the associated anomalies. For those patients with isolated colonic duplications, all five with cystic lesions underwent resection. One patient underwent colostomy, and later, internal drainage of a tubular duplication. One patient with cecal volvulus had a tubular duplication of the ascending colon left intact because it communicated at both ends. Five of the six patients with long tubular duplications underwent operation. Two were resected with primary repair. Loop colostomy of the native and duplicated colon was performed in a newborn with low imperforate anus and bladder exstrophy. She will undergo closure of her colostomy and internal drainage of her duplication at 1 year. Resection and colostomy was performed in the partial conjoined twins. In the two patients with cloacal exstrophy, one duplication was resected with colostomy. In the other, the duplicated colons, which communicated proximally, were exteriorized together as adjacent ostomies in order to preserve colonic length.

Two patients did not undergo operation. Postmortem examination of a newborn with multiple anomalies revealed a long tubular duplication of the transverse colon. An asymptomatic duplication with distal communication to the transverse colon was found incidentally upon barium study for constipation. The duplication was not believed to contribute to the symptoms, and the patient has remained asymptomatic without resection.

Three of the five patients with rectal duplication presented as newborns, and the remaining two patients were younger than 6 months of age. All duplications were cystic. Only one communicated with the rectum, and this was associated with a duplex vagina (Figs. 5A and B). In four instances, the mass was adherent to the rectum and had a common muscular wall, but could be excised with preservation of the anal canal. Emergency colostomy was performed in one newborn because of marked colonic distention. He is 6 months of age and is awaiting excision via a posterior sagittal approach. All patients with colonic and rectal duplications who underwent operation recovered without complication.

Comment. Occasional small cystic duplications can be excised without colonic resection. However, resection and colocolostomy are usually necessary in managing patients



FIGS. 6A and B. (A) Chest x-ray of a 3-year-old demonstrates a right paravertebral mass. There is a large anterior defect in the vertebral bodies of the upper thoracic spine. (B) A myelogram shows contrast in the neuroenteric cyst (solid arrow) with extension subdiaphragmatically into the distal small intestine (open arrow).

with colonic duplications of this sort. A long tubular duplication is frequently not entirely resectable. If the blind end is above the pelvic floor, distal internal drainage can be established by excising the common wall for a short distance to allow re-entry into the single distal colon. No patient with a tubular colonic duplication presented with bleeding, and ectopic gastric mucosa was not noted in such duplications. Internal drainage of tubular colonic duplications is an effective alternative when complete resection is not possible.

The typical rectal duplication is cystic and presents as a gradually enlarging mass adjacent to but not involving the anal canal. They usually present with signs of intestinal obstruction and may resemble the more common sacrococcygeal teratomas. The term *Middledorf's tumor* is incorrectly applied to presacral teratomas, because Middledorf himself actually described an enteric cyst.^{23,24} Complete duplication of the colon and rectum can be accompanied by doubling anomalies of the urethra or bladder, exstrophy of the bladder, spina bifida, and omphalocele.³ This association between colonic duplication and genitourinary anomalies was present in four patients.

Thoracoabdominal Duplications

Three patients presented with combined thoracoabdominal duplications, and five children presented with separate thoracic and intestinal duplications. In the former group, the duplications communicated in the abdomen with the stomach, ileum, and jejunum, respectively. All

three patients had thoracic vertebral anomalies, although a connection with the spinal canal occurred in only one instance. A 7-day-old patient presented with vomiting and a left upper quadrant mass. Barium study revealed anterior displacement of the stomach. A thoracogastric duplication was completely resected along with the greater curvature of the stomach in one stage. In a second patient, a myelogram was performed on a 3-year-old girl with a right thoracic mass and thoracic hemivertebrae that revealed a cystic component in the spinal canal in continuity with the intrathoracic cyst, and contrast was seen to extend subdiaphragmatically, confirming the diagnosis of thoracoabdominal duplication (Figs. 6A and B). A three-stage procedure was performed, beginning with right thoracotomy and followed by ileal resection 2 weeks later. Emergency thoracic laminectomy (T2–T4) for excision of the intraspinal portion of the neuroenteric cyst was performed because of increasing spasticity and paresis in the left leg. This child recovered without incident.

The third patient was a 6-week-old infant in whom chest x-ray demonstrated hemivertebrae as well as air in a right thoracic mass, suggesting the diagnosis of duplication with communication with the intestinal tract. The thoracic duplication was resected first, and 4 weeks later, the subdiaphragmatic segment joining the jejunum 10 cm distal to the ligament of Treitz was removed.

Comment. Eight of the 24 children with thoracic duplications had either separate or incontinuity abdominal duplications. For this reason, patients with esophageal

duplications require careful abdominal examination; US is particularly helpful in this respect. In those patients with abdominal duplications, evaluation of the chest by x-ray is necessary. If a thoracic duplication is then suspected, an enhanced CT scan is the next logical step. Although total excision is the desirable approach, staged thoracic resection followed by abdominal resection of thoracoabdominal duplications is also satisfactory. The latter approach has been used more frequently in neonates in the past. Our experience with the child who had a neuroenteric cyst who required emergency laminectomy suggests that patients demonstrated to have such lesions on CT scan and myelography should have thoracotomy and laminectomy performed for simultaneous excision of both components.

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