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Cystic duplications of the duodenum

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CYSTIC DUPLICATIONS OF THE DUODENUM

The term cystic duplication of the duodenum is a malformation which has been described as, enterogenous cysts, enteric cysts, duodenum duplex, unusual Meckel's diverticula, inclusion cysts, and duplications of the duodenum. The latter is more inclusive and is to be preferred since all of the abnormalities are embryologically associated.(1) The term cystic is retained to limit the paper to those duplications of the duodenum which do not communicate with the normal duodenal lumen. Duplications are not limited to the duodenum but may appear at any level of the gastro-intestinal tract, and have been found from the base of the tongue to the anus.(2)

ETIOLOGY:

Duplications of the gastro-intestinal tract present a a wide variation in size, shape, relation to the intestinal wall and lumen, and relation to the mesentery. No single theory as to their origin can satisfactorily explain all the cases observed. There are three different theories concerning the embryogenesis of intestinal duplications, which between them can adequately account for the gamut of variations noted clinically.(3)

An often suggested mode of origin for cystic duplications is that they result from abberations in the development of a Meckel's diverticulum. This fairly common diverticulum is the persistent vitelline duct, originally connecting the yolk sac and the intestinal tract. It has its own group of anomalies such as fistulae

or cysts, which are well understood, always recognizable by their positions and entirely distinct from duplications of the intestinal tract. This mode of origin cannot explain those cases of duplications found at the base of the tongue, esophagus, small bowel, or colon.

A second mode of origin, that has been stressed by Ladd and Gross,(4) and others, (5,6,7,8,) is based on the observation of Lewis and Thyng, (9), in 1907, who reported frequent diverticula at the various levels of the fetal intestinal tract in the pig, rabbit, and man. These diverticula are found in 20-30mm. embryo (eight to nine weeks), usually on the antimesenteric border, and normally disappear completely. It is not difficult to visualize how such a residual outpouching can become pinched off and give rise to cystic structures with the histologic components of the intestinal wall. They may be located anywhere from mouth to anus; they may be in a submucous, intramural, subserous or intramesenteric position; and they may communicate with the bowel or be completely separated fromit. However, this mode of origin will not account for the long tubular intestinal duplications that have been observed by Ladd and Chrisholm,(10) and Feingold and Shulman,(11) nor those that have a separate mesentery or more than one communication with the bowel lumen.

A third mode of origin has been described by Bremer.(12) In the 10 mm. embryo (about six weeks) just prior to the rapid lenghtening of the intestinal tract there is a hyperplasia of the

epithelial cells producing a marked reduction in the size of the lumen, or even occluding it. The solid stage may occur at any point along the intestinal tract. These epithelial cells secrete a fluid which forms small vacuoles. These vacuoles coalesce and reform the lumen of the tube. In rare cases a chain of vacuoles may coalesce but not reunite with the main lumen, producing two separate lumens. As growth progresses the two lumens become more separated and each comes to be surrounded by a submucosa and muscular layer. This, there is produced a double-barrelled tube or duplication of the intestinal tract. The duplicated portion may be long or short; it may be within the wall of the intestine or completely separate from it even to the point of having a separate mesentery; it may communicate with the intestinal lumens at both ends, at one end, or be completely sealed off.

It is very likely that most of the spherical cysts as are found in the duodenum are derived from diverticula as based on the observations of Lewis and Thyng, (9) and that most of the tubular structures originate by the abnormal persistance of the vacuoles as proposed by Bremer.(12)

PATHOLOGY:

Cystic duplications of the duodenum are spherical or elongated hollow structures closely resembling the gastro-intestinal structures, their walls being made up of a mucous membrane, smooth muscle, and serosa.

The presence of the smooth muscle coat is an important constant feature of the lesion. The muscularis may be thin and attenuated or may be thick and well differentiated into an outer longitudinal and inner circular layer. It is adherent to or continuous with the muscularis of the duodenum allowing no plane of cleavage.(13) It is this coat which causes the cystic duplication to resemble the intestine to closely. Usually there is a fusion of the external musculature of the cystic duplication and the duodenal musculature with a common serosa and a common blood supply.(14) The submucosa is a thin connective tissue layer. Epithelium may form a complete lining, or be present in limited patches or even be completely absent, but in this last event epithelial cells are usually found in the contents of the cyst. The epithelium forms a thin smooth layer, occasionally possesses glands, and is most frequently columnar or low cuboidal, but it may undergo a transition to ciliated or stratified epithelium.(15)

The type of fluid which these cysts contain varies. In simple uncomplicated cases it may be clear and colorless. If the intracystic pressure becomes too great or if the cyst becomes too large, it may produce a pressure occlusion of the blood vessels within the walls of the cyst thereby causing the mucous membrane to undergo necrotizing and gangrenous changes. In such an event the aspirated fluid may be hemorrhagic.(16)

The cyst may occupy any plane in the intestinal wall: submucous, intermuscular, or subserous, and may occupy any segment of the gut periphery, whether mesenteric, anti-mesenteric or any

any intervening site. Usually, however, the duplicative cysts of the duodenum are found in the second portion of the duodenum in the intermuscular layer of the posterior wall. (See table I)

The size of these cysts vary considerably, varying from 2.1 X 1.5 cm to 12 X 15 cm. (see table I)

SYMPTOMS:

Cystic duplications of the duodenum do not give rise to any characterizing symptoms but rather mimics those seen in other forms of acute or chronic intestinal obstructions.(16) In this series(see table I) either pain and/or vomiting was the presenting complaint in all cases.

Vomiting was the most common symptom found in the cases that survived the first few days of life, being present in 73.4% of the cases.(see table I). The vomiting was due to duodenal obstruction, and became more pronounced as the obstruction increased. Dehydration, toxemia and alkalosis, and rapid intense prostation are symptoms secondary to the vomiting and may reach a marked degree if not corrected early. Mailaise is a secondary symptom and may be marked in cases of severe vomiting and dehydration. Loss of weight, or inability to gain weight is a constant feature especially in the younger patients and is the result of partial duodenal obstruction.

Pain is the only other common symptom, and was found to be present in 72.7% of the patients over the age of one and a half years.(see table I) The pain is usually in the right upper quadrant,

and the degree of pain is dependent upon the degree of duodenal obstruction. The pain may simulate that of acute cholecystitis or of a duodenal ulcer, but usually is not associated with feeding. In cases of chronic obstruction, the attacks may be separated by intervals of complete relief.

PHYSICAL SIGNS:

The most significant physical sign is the palpation of a mass in the abdomen. This sign was present in over 50% of the cases.(see table I) These masses are usually movable, may feel cystic or hard, and are usually but not always found in the right upper quadrant. The size of the mass depends upon the size of the cystic duplication which is not a constant feature.

Abdominal tenderness may be present but is not a common sign. If present, the tenderness is usually more pronounced in the right upper quadrant but this does not always obtain.

Distension is a marked feature if obstruction is nearly complete or the cystic duplication is unusually large. If caused by the former, it is due to the dilatation of the stomach and duodenum. Peristaltic waves are also noted if distension is present.

ROENTGENOLOGIC FINDINGS:

The roentgenologic studies are negative in all cases which do not have duodenal obstruction. Since the majority of the patients with cystic duplication of the duodenum have obstruction, radiologic studies will show dilatation of the

duodenum and enroachment upon the lumen of the duodenum. These findings together with delayed emptying time of the stomach are diagnostic of duodenal obstruction and are the pathognomonic findings of non-malignant lesions of the duodenum.

Dilatation shows in the X-ray film after a barium meal as a large dense shadow following the general contour and direction of the duodenum to the point of the obstruction. Fingerlike projections of the barium shadow may also be seen in the dilated crypts of Lieberkuhn.

Enroachment upon the duodenal lumen shows as an indentation of the barium shadow. It is impossible in these cases to make any positive statement as to diagnosis from this finding alone.

Although it is impossible to make a diagnosis by X-ray studies, these studies will enable the operator to limit the lesion to the duodenum, and the radiologist in most instances is able to state that the lesion is probably benign.

DIAGNOSIS:

The fact that this condition has not been diagnosed preoperatively shows the difficulty encountered in making a definitive diagnosis. From a study of table I, the following points are pertinent. Fifty six per cent of the cases were under the age of one and a half years, the remainder ranged up to sixty-nine years of age. The lesion is found more commonly in the female, the ratio being about 11 to 7. The commonest symptoms are pain and vomiting associated with duodenal obstruction, and the most important physical finding is a mass in the abdomen. It is interesting to note the diagnosis of congenital hypertrophic

stenosis was the preoperative diagnosis in about one third of the cases. In review of the cases, the radiologic findings were most commonly reported as a non-malignant lesion of the duodenum. It would therefore behoove the surgeon to remember this lesion when the patient presents himself with the symptoms, signs, and radiologic findings as discussed in the preceeding paragraphs.

At operation the surgeon must be able to distinguish cystic duplications from adenomas, myomas, fibromas, lipomas, hemangiomas, and neurogenic tumors of the duodenum. This is easily done if one remembers that cystic duplications are hollow structures, composed of the same elements and having the same gross appearance as the adjacent duodenum to which they are attached. This description does not obtain for the above named lesions from which the cystic duplications must be differentiated. If the radiologic impression was incorrect and a carcinoma of sarcoma of the duodenum is present, these can also be distinguished by gross appearance but should be biopsied and examined histologically if any doubt is present as to the surgical procedures necessary in these cases would differ greatly from those used for the benign lesions.

TABLE I: COLLECTED CASES OF CYSTIC DUPLICATION OF THE DUODENUM

r	Sex	Age	Symptoms	Physical Examination	Preoperative Diagnosis	Location	Size	Operation	Operator	Result
0*	?	NB	.....	.....	.....		"Walnut"	.....	Sanger & Klopp	Infant died during delivery
1*	M	NB	.....	Abdomen extremely distended	.....	Third portion of duodenum	.....	.....	Roth	Infant died a few minuted after after delivery
9*	F	3wk.	Persistant vomiting beginning at 2 weeks.	Negative	Pyloric Stenosis	Submucous; extending from pyloris to ampulla of vater on medial wall	"hensegg"	none	Meyer	Died
2	F	19dys.	vomiting-abdominal pain	Firm tumor in right side of abdomen	Tumor of rt. hypochondrium	Posterior wall second portion of duodenum	"tangerine"	I. cyst evacuated & packed. II. Marsupialization.	Waugh	Died
	?	3mo.	Projectile vomiting	Hard mass in R.U.Z.	Pyloric obstruction	Second portion of duodenum	"goose egg"	Posterior gastroenterostomy attempted	Maddox	Died first hosp. day.
	F	2wk.	Vomiting	Abdominal distension mas in RUQ	Congenital pyloric stenosis	Anterior wall first portion of duodenum.	"Bantam's egg"	Biopsy pyloris dilated. Exterior drainage	-Smith	Died 7th hosp. post operative da day
	F	15yr.	Attacks of RUQ pain & Vomiting	Mass in RUQ	Choledochus cyst.	Anterior wall first & sedond portion of duodenum	About 10 cm in diameter	Enterocystostomy	Gardner & Hart	Complete recovery
	F	4mo.	Vomiting	Mass in RUQ	Cyst of abdomen	Intramuscular lateral wall, first portion ofduodenum	8 x 11 cm contents -550 cc.	Cyst drained prtial marsupialization with catheter	Pachman	Died 4 hrs. Post operatively
	F	4wk.	Vomiting	Mass in RUQ	Hypertrophic Pyloric stenosis	Anterior wall of - first portion of duodenum	2-3cm long, contents 15 cc	I. Evaluation II Gastroenterostomy 7 days later	Basman	Complete recovery
*	F	5 wk.	Regurgitation vomiting	Soft, Cystic mass above naval	Omental cyst or enteric cyst	Mesenteric border first portion of duodenum	4.5 x 3.5 cm.	Resection of cyst & first portion of duodenum with gastroenterostomy	Ladd & Gross	Protracted recovery.
0*	M	4½yr.	Jaundice Abdominal pain	Icterus mass in R.U.Q.	.....	Intramuscular; posterior wall of second portion of duodenum	12 cm diameter contents 350 cc	Excision of cyst	Gillespie & Rogers	Complete recovery
1	M	2mo.	Projectile vomiting	cystic mobile tumor in RUQ	Renal neoplasm	Pyloris and first portion duodenum	7.6cm diameter	Resection with gastroduodenostomy	Orgias	Complete recovery
1	F	14dys.	Vomiting	Tumor mass in RUQ	Pyloric stenosis	Anteriolateral to first part of duodenum	"Large"	Marsupalized Intend to anastomose at future date.	Pevaroff & Saunders	Complete recovery
5	M	26yr.	Epigastric pain, vomiting	negative	.....	Posterior wall second portion of duodenum	12 x 15 cm	Excised	Brooks & Weinstein	Complete Recovery
5	M	17yr	Intermittent attachks of nausea and vomiting	fixed mass in right epigastrium	Adenoma? Myoma? cyst of Brunner's gland?	Posterior medial wall first & second portion	2 cm diam. contents 8 cc.	Enucliation	Booher & Pack	Complete recovery
	M	12yr.	Attacks of acute pain in RUQ Vomiting	Negative	Leiomyoma of second portion of duodenum	Posterior wall second portion of duodenum.	2.1x1.5 cm.	Excision	Shallow Wagner Manges	Complete recovery
	M	17yr	Attacks of severe pain in RUQ	Negative	Tumor of second portion of duodenum	Posterior wall second portion of duodenum.	4.5 cm diameter	Partial excision. Anastomosis to duodenal lumen	Lorber Machella	Complete recovery
	F	69yr	Epigastric pain Vegurgitation	Negative	Non-malignant tumor of duodenal cap.	Anterior wall pyloris and first portion of duodenum	3 inches long 1 inch in diameter	Excision	Peple	Complete recovery

te reported; other dates indicate year of operation

TABLE I (continued)

Case	Year	Sex	Age	Symptoms	Physical Examination	Preoperative Diagnosis	Location	Size	Operation	Operator	Result
19	1948*	F	6½yr.	Epigastric distress and vomiting	Abdominal distention two tumor masses over umbilicus	Multiple retroperitoneal tumors	Posterior lateral wall second portion of duodenum		I Excision II Gastrojejunostomy on fifth post operative day	Hickens Stevenson Carlquist	Complete recovery
20	1950*	F	30yr.	Mild indigestion 1 yr. Generalized Abdominal pain 3 dys.	tender a abdomen pain in RUQ	Acute appendicitis	Submucous posterior wall second portion of duodenum	4 x 8 cm	Partial excision	Gordimer Bluestone	Complete Recovery
21	1952*	F	8½yr.	Pain vomiting	.....	.....	.....	3.x4cm	Entero-cystotomy	Holcomb Gross Farber	Complete recovery
22	1952*	..	1½ yr.	pain vomiting	.....	.....	.....	5 cm diameter	Enterocystotomy	Gross Holcomb Farber	Complete recovery
23	1949	M	18dys	Vomiting	Periumbilical mass	Extrensic compression of duodenum by tumor	Second portion of duodenum	.....	Drainage by catheter	Barcot	Died one month following surgery

* Date reported; other dates indicate year of operation

TREATMENT :

REVIEW OF CASES TO DATE:

There have been 23 cases of cystic duplication of the duodenum reported in the literature. Three cases were not operated.(17,18,19) Waugh(20) in 1923, Was the first to operate on a cystic duplication of the duodenum. His patient was a 19 day old girl in which he found a retroperitoneal cyst which was adherent to the posterior wall of the duodenum. He packed the cavity of the cyst with gauze, but six weeks after the removal of the gauze it refilled. Six weeks later the child was operated again, the cyst wall being sutured to the aponeurosis of the abdominal wall, but the child died of pneumonia six days after the second procedure. Maddox (21), attempted a posterior gastro-enterostomy but the condition of the child made it impossible to complete the operation. The child died the day of operation. Smith(22), in 1930, operating on a two-week old infant with a cyst of the anterior wall of the duodenum, drained it externally. The child dies one week postoperatively, the cyst having regressed greatly in size. The first successful operation was that of Gardner and Hart(23) in 1935, who treated a 15 year old girl by opening the cyst from within the duodenum and anastomosing the cyst to the intestinal tract. In Pachman's (24) case, after removal of 550 cc of gr eenish-black thin fluid, a small portion of the cyst was removed for biopsy and a small catheter was inserted and secured to the peritoneal opening to effect partial

marsupialization. The infant died four hours after operation. Basman(34), in 1936, because of the poor condition of the four week old infant, elected to simply aspirate the cyst. In seven days the cyst had refilled causing obstruction. Seven days after the first procedure, a gastroenterostomy was performed after attempting to enucleate the cyst. The infant was doing well one year after surgery.

Ladd and Gross(4), in 1940, resected the first portion of the duodenum and performed a posterior gastrojejunostomy. The five week old infant recovered after a long period. Gillespie and Rogers (25), aspirated 450 cc of the contained fluid and then excised the collapsed sac and inverted the duodenal wall. The patient recovered. Orgias(26), attempted to dissect the cyst off the posterior portion of the pyloris and first portion of the duodenum but, finding it impossible, resected the involved portion of the stomach and duodenum with subsequent end to end anastomosis, with an uneventful recovery. Pevaroff and Saunders(27), in 1941, marsupalized a cyst in a 14 day old girl. They reported the patient doing well for the past eight years but has had 10-20 cc. drainage of mucous daily. They intend to anastomose the cyst to the gastrointestinal tract in a second operation. Brooks and Weinstein(23), were able to excise a cyst located on the posterior wall of the second portion of the duodenum. Booher and Pach(29), in 1945, were able to enucleate the cyst using sharp and blunt dissection. Shallow, Wagner, and Manges (13), were able to excise the cyst from the duodenal wall with complete recovery of the patient.

Larber and Machella(30), attempted to excise the cyst but had to compromise due to the close relationship of the cyst and the orifice of the common bile duct. A partial excision and anastomosis to the duodenal lumen was performed with complete recovery. Peple(31), in 1947, was able to dissect the cyst from the duodenal wall by manipulating the cyst with a finger in the stomach. The patient had an uneventful recovery. Hickens, Stevenson, and Carlquist(16), in 1948, excised the cyst but were forced to do a posterior gastrojejunostomy on the fifth postoperative day because of acute duodenal obstruction. Gordimer and Bluestone(32), in 1950, excised the major portion of the cyst except that portion containing the termination of the common bile duct. The remaining portion of the cyst wall was sutured. The common bile duct was opened and a T tube inserted for decompression. The patient recovered completely. Gross, Holcomb, and Farber (33), reported two cases which were successfully treated by cutting a window in the cyst and allowing the contents to drain into the duodenal lumen. Barcot(35), in 1949, attempted to excise the cyst from the duodenal wall but was unable to do this, because of the intimate association of the cyst to the pancreatic duct. The patients condition became shocky and he decided to treat the cyst by drainage using a catheter. The patients condition improved with remission of symptoms in three days after surgery. A month after surgery a definitive procedure was contemplated, but the child developed a severe diarrhea from which it died.

DEFINITIVE SURGERY:

The primary consideration of surgery is to relieve the obstruction thus re-establishing the patency of the gastro-intestinal tract. The type of procedure depends upon the size, location and severity and duration of the obstruction. Six types of procedures have been used. (see fig. I)

Marsupialization or drainage by catheter has been attempted in five cases. It is by far the simplest procedure yet has only a 20% survival rate. Of the four cases that died, one died of pneumonia before the days of antibiotics. (see table I, case 4) Another died of a severe diarrhea one month after surgery. (case 23) The other two died because they were unable to withstand surgery due to their poor physical condition. (cases 6, and 8) Because there are better procedures for treating this condition, I believe that marsupialization or drainage by catheter, is not the treatment of choice. It should be considered however in the newborn or very young and those patients in extremely poor physical condition as it is the simplest procedure.

An enterocystotomy has been performed on four cases (cases 7, 17,21, and 22) with a 100% survival rate. (See fig I) The argument against thisprocedure is that a diverticula is created which may cause the patient as much trouble as the cyst as occurred in the case of Gardner and Hart(23). If the cyst is small and submucous in pattern, the muscular layers of the

duodenal wall should be able to keep the diverticula empty. I think that this is the procedure of choice for cases of this type, and for those cysts which are intimately associated with the common bile duct. In the latter type, it is practically the only feasible procedure. (from Hichens et. al. 16)

Gastroenterostomy was attempted in two cases with a 50% survival rate.(cases 5, and 9) In the case that dies, (case 5), the child was unable to withstand surgery and died one hour after surgery. In the other case, this was the second procedure attempted, the first being simple aspiration. I do not believe that this procedure should be used alone and is to be condemned for two reasons. First, it leaves the cyst in the abdomen, and secondly, there are better procedures which can be used.

type surgery	Number of cases	Survival rate
excision	6	100%
excision and gastroenterostomy	1	100%
resection of cyst and gastroenterostomy	2	100%
gastroenterostomy alone	2	50%
gastrocystotomy	4	100%
marsupalization and/or drainage by catheter	5	20%

figure I. The type surgery compared to the survival rate as reported by the various operators.

Resection of the cyst and adjacent duodenum with a gastro-enterostomy has been succes fully performed in two cases. (see table I cases 10 and 12) Many surgeons, (4,29,32), believe this to be the procedure of choice because it removed the cyst and does not leave a weakened duodenal wall as may occur in excision of the cyst, or create a diverticula as does an entero-cystotomy. The advocates of this procedure fail to consider that in the majority of cases these cysts are intimately associated with the common bile duct which whould have to be transplanted if the cyst and adjacent doudenum were resected. In the latter type, I believe that this type procedure should not be performed because there are simpler procedures. When the condition of the patient and the location of the cyst is such that this procedure is feasible, the surgeon should give it heavy consideration.

Excision of the cyst followed in a week by a gastro-enterostomy was performed successfully in one case. (table I, case 19) The second procedure was necessary because the welling about the excised area had caused complete duodenal obstruction.

Excision alone was performed in six cases, (table I, cases 11, 14, 15, 16, 17, and 18), with a 100% survival rate. The surgeon has three risks using this procedure, First, he may injure the duodenal wall or leave a weakened duodenal wall.

Second, the arteries and veins of the contiguous duodenum usually course over the surface of the cyst and resection may result in necrosis of the duodenal wall. These seem more apparent than real as this did not occur in any of the seven cases treated by this method. Third, edema of the duodenum may occur and cause obstruction as occurred in case 19. In reviewing the cases treated by this procedure, all of the surgeons stated that they excised the cyst only with difficulty, but they agree that this procedure should be attempted first as the procedure of choice.

From a review of the cases, it is apparent that the procedure used must be determined by the patients age, physical condition, and the location of the cyst. There is no procedure of choice, and the surgeon should be aware of the many methods which have been successfully performed. Any cyst found as an incidental finding at surgery should be left alone if it is not causing symptoms.

SUMMARY:

Cystic duplication of the duodenum is a congenital defect probably arising from a diverticula during embryonic development. They are spherical or elongated hollow structures closely resembling the duodenum in structure, as their walls are made of a mucous membrane, and smooth muscle and serosa. The chief symptoms are those of acute or chronic intestinal obstruction. Pain and/or vomiting being the presenting symptom in all cases.

The most significant physical sign is a palpable mass, this can be found in over 50% of the patients. Other physical findings depend upon the degree of obstruction present. The radiologic findings depend upon the degree of obstruction present. The diagnosis can only be made at surgery. Treatment of this condition is dependant upon the patients age, physical condition and the position of the cyst in relation to the duodenal wall and the common bile duct.

CONCLUSION:

Every surgeon must consider the diagnosis of a cystic duplication of the duodenum in patients having symptoms of acute or chronic duodenal obstruction especially if a mass is palpated in the abdomen. Because the diagnosis can only be made at surgery, the surgeon must be able to make the definitive diagnosis at operation, and be familiar with the various methods of treating this condition.

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