Annular Pancreas as a Cause of Acute Neonatal Duodenal Obstruction *

DANIEL M. HAYS, M.D.,** EDWARD M. GREANEY JR., M.D.,*** JAMES T. HILL, M.D.

From the Surgical Services of the Los Angeles Children's Hospital and the University of California Hospital, Los Angeles, California

Introduction

ANNULAR PANCREAS, when observed as a disease entity producing chronic or recurrent intestinal symptoms in adults or children, has been frequently reported and several hundred cases have been described.

Annular pancreas, as a cause of acute duodenal obstruction in the newborn, is much less commonly encountered. Among cases of neonatal intestinal obstruction described by Gross,[8] annular pancreas comprised less than one per cent of the total group, and less than 5 per cent of those with localized duodenal obstruction.

Seven such infants, in whom functional obstruction was noted from the onset of oral feedings and laparotomy performed in the initial ten days of life, have come under our observation at the Los Angeles Children's Hospital and U. C. L. A. Medical Center during the past six years (1954–59). Thirty-three additional operative cases, which have been reported during the past 15 years (1944–59) and meet these criteria, have been collected (Table 1) for comparison.

Embryology

Theories of the embryologic origin of annular pancreas have extremely practical implications warranting discussion in detail.

An analysis of pancreatic development particularly relative to this anomaly, was made by Lecco [11] whose concepts have been accepted by Smetana [21] and others.[3,16] In brief, the pancreatic anlages are two pouches, extending dorsally and ventrally from the primitive gut in the vicinity of the hepatic diverticula. The dorsal pouch elongates dorsally and to the left, ultimately forming the ductal system of the body, tail and ventral aspect of the head of the mature pancreas. The ventral pouch (which may be bifurcated into right and left pouches—Lewis [12]), intimately attached to the developing choledochus, migrates to the right and then dorsally with this structure ultimately forming the ductal aspects of the dorsal head and uncinate process. The duct formed by the ventral process becomes the proximal portion of the principal pancreatic duct (Wirsung) and forms connections with branches of the dorsal duct in the body and tail, while retaining its attachment to the choledochus. The proximal portion of the dorsal duct remains as the accessory pancreatic duct (Santorini).

The embryological explanation for the development of the annular pancreatic anomaly (Lecco) postulates a failure of the entire ventral pouch to undergo migration around the right side of the gut. Its connection with the choledochus is established; but its original attachment to the ventral wall of the gut is also retained. As the choledochus migrates to the right, dorsally, and then to the left of the prin-

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** Assistant Clinical Professor of Surgery, U. C. L. A.

*** Instructor in Surgery, University of Southern California.

TABLE 1. *Annular Pancreas Producing Duodenal Obstruction in the Neonatal Period (Collected Operative Cases, 1944–59)*

Authors	Year	Total Cases	Infants without Assoc. Major Anomalies or Prematurity			Infants with Associated Major Anomalies or Prematurity							
			Total #	Survivals	Deaths	Total #	Survivals	Deaths	Premature	Mongol	T.E.F	Imperf. Anus	Congen. Ht. Dis.
Gross & Chisholm	1944	1	1	1	0								
Burger & Aldrich	1949	1	1	0	1								
Glover & Barry	1949	1				1	0	1	1				
Ravitch & Woods	1950	2	2	2	0								
Shapiro, Dzurik & Gerrish	1952	3	3	2	1								
Wilson & Bushart	1953	1	1	0	1								
Gross	1953	8	6	5	1	2	0	2		(2?)	2	1	2
Dunavant	1954	1	1	1	0								
Hope & Gibbons	1954	3	2	0	2	1	0	1			1		
Kiesewetter & Koop	1954	4	3	1	2	1	0	1			1		
Rickham	1954	3	1	1	0	2	1	1	1				1
Small & Berman	1954	1	1	1	0								
VonEkesparre	1956	1				1	1	0		1			
Wilkinson & Simms	1957	1	1	1									
Weatherill, Forgrave & Carpenter	1958	2	2	1	1								
Hays, Greaney & Hill	1959–1960	7	2	2	0	5	2	3	2	2	2	1	
Totals		40	27	18 (67%)	9	13	4 (31%)	9	4	3	6	2	3

cipal axis of the duodenum, one segment of the original ventral pouch remains attached to it, while the other portion continues to adhere to the ventral aspect of the gut. The ductal system formed by the ventral pouch thus almost completely encircles the duodenum—running from left to right anteriorly and from right to left posteriorly. Relatively minor enlargement of the tissue of the pancreatic head would complete the encirclement.

This oversimplified explanation neglects the rotations of the gut and the elongation of the choledochus which occur concurrently with the migration of the pancreatic pouches. Of clinical significance, in this regard, is the fact that surgical division of the "ring" anteriorly is usually complicated by encountering the major pancreatic duct. Union of the two pancreatic anlages takes place between the 14 mm. and the 22 mm. stage (Lewis),[13] so that irrespective of the precise mechanism involved, it is probable that a ring of pancreatic tissue surrounds the duodenum in these cases prior to the eighth week of gestation. At this stage the caliber of the lumen of the duodenum is minute; and significant subsequent enlargement of this segment surrounded by pancreatic tissue is improbable. It thus represents a functional duodenal stenosis of a degree simulating atresia. Realization of this developmental situation makes the frequently described "associated anomaly" of "duodenal atresia or stenosis" readily understandable. It also mitigates strongly against any therapeutic concept which includes removal of the "constricting pancreatic ring" to free the duodenum, which is necessarily so structurally deformed as to make it unsuitable for function even if it were possible to separate the pancreatic tissue from its walls at operation.

Case Reports

The following seven case reports illustrate the problems presented by this anomaly in the neonatal period.

Case 1 (C.H. 110743). Birth weight 3,706 Gm. The pregnancy was uncomplicated. Vomiting of nonpigmented material by the infant was noted on the third day of life. Physical examination revealed slight upper abdominal distention and signs of dehydration. Plain x-ray films of the abdomen demonstrated apparent high intestinal obstruction. A barium enema revealed a normal rotation of the colon. Laboratory studies were within normal limits except for a hypocholoremic alkalosis, which was corrected prior to laparotomy. At operation pancreatic tissue completely encircled the second portion of the duodenum. Peritoneal bands also crossed the duodenum from the liver surface to the hepatic flexure of the colon. A retrocolic duodenojejunostomy was performed. The postoperative course was uncomplicated, and oral feedings were tolerated from the third day. The infant was discharged from the hospital on the tenth day following operation weighing 2,934 Gm.

At four months of age this child (weight 5.6 kg) was readmitted because of acute diarrhea. A gastro-intestinal series at this time showed moderate stasis in the proximal duodenal segment. Despite this finding, acceptable weight gain and general development has continued to date (3½ years).

Case 2 (U.C.L.A. 014-57-48). Birth weight 1,984 Gm. Pregnancy was complicated by polyhydramnios. Nonpigmented vomiting was noted following the initial feedings. Two meconium stools had been observed. Transfer to this hospital was carried out on the third day of life. Physical examination revealed an active, somewhat dehydrated, premature infant. The abdomen was not distended, there were no masses, and faint peristalsis was audible. Routine laboratory studies were within normal limits. A gastric tube was passed and 90 cc. of formula (nonpigmented) extracted; following which 80 cc. of air was injected into the stomach. Roentgenograms revealed two large radiolucent intestinal outlines in the upper abdomen; the characteristic "double bubble" shadow of duodenal obstruction. Laparotomy, at three days of age, demonstrated a pancreatic ring encircling the second portion of the duodenum. The gallbladder was distended. A retrocolic duodenojejunostomy was performed. The course following operation was uncomplicated, with stools passed from the fourth day. The infant was discharged from the hospital fourteen days following operation weighing 2,382 Gm.; and was known to have remained without complications for nine months thereafter.

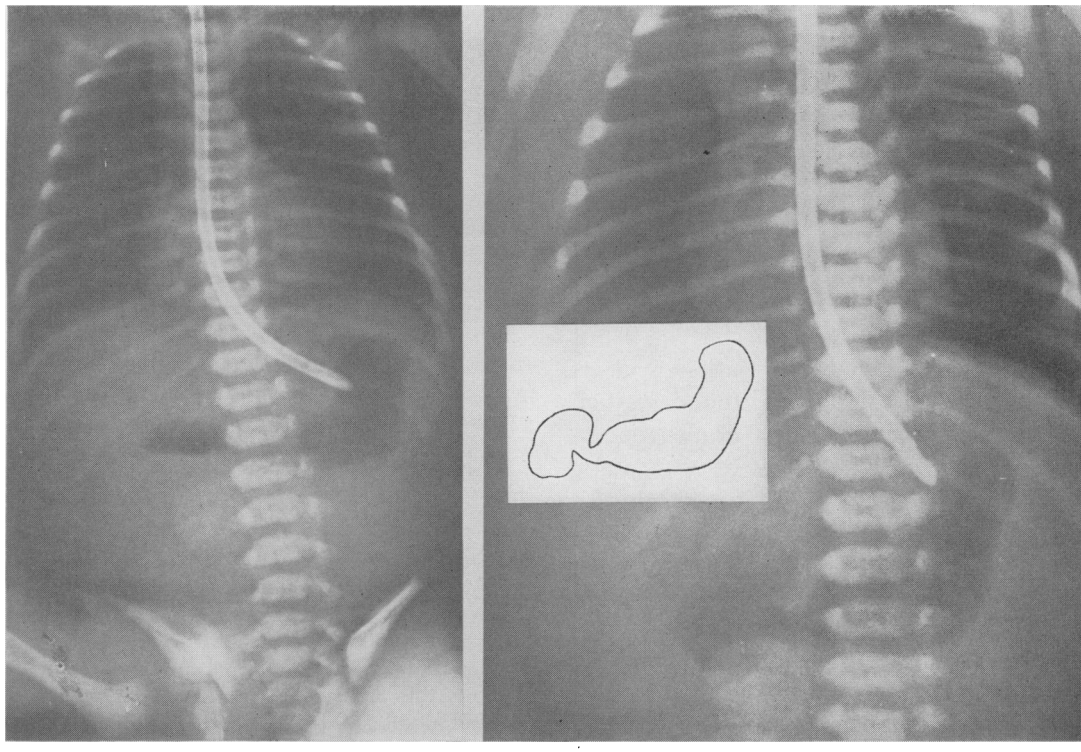

FIG. 1. Two-day-old male infant (Case 3) with history of vomiting of all water feedings. The abdomen was not distended. Studies in the upright position (left) reveal two distinct upper abdominal shadows with fluid levels, referred to as the "double bubble"; while films taken in the supine position (right) display the bi-lobed shadow of duodenal obstruction described as the "dumbbell" configuration.

Case 3 (C.H. 168232). Birth weight 3,290 Gm. The pregnancy was complicated by polyhydramnios; and delivery was by Caesarian section. All water feedings were vomited, and the patient transferred to this hospital for operation on the second day of life. Physical findings were unremarkable, with abdominal distention completely absent. X-rays revealed the stomach and dilated duodenum with a relatively narrowed pyloric area between, forming a "dumbbell" shadow. (Fig. 1). Laboratory data was within normal limits. At laparotomy (age, 48 hours) a pancreatic annular ring was encountered with functionally complete duodenal obstruction. A retrocolic duodenojejunostomy was performed. The patient tolerated oral feedings on the third postoperative day and was discharged on the twelfth day weighing 2,780 Gm. He was readmitted at age seven months (weight 8.6 kg.) for acute intestinal obstruction secondary to postoperative adhesions, which required lysis. Subsequent growth and development has been excellent (age, 1 year).

Case 4 (U.C.L.A. 018-04-61). Birth weight 2,814 Gm. Three previous pregnancies were terminated by spontaneous abortion. The present pregnancy was uncomplicated. Vomiting was described from the time of the initial feeding. Mongolism was suggested on initial examination. Roentgenograms of the abdomen revealed duodenal obstruction. This was further demonstrated by ingested barium which remained in the stomach until hospital transfer and definitive operation. Thus retention for at least 8 hours was noted Fig. 2). The patient was admitted to this hospital on the sixth day of life. At this time the abdomen was soft and undistended. There was a marked hypochloremic alkalosis. Following intravenous correction of this deficit, laparotomy was performed (age, 6 days). The second portion of the duodenum was surrounded by pancreatic tissue. A retrocolic duodenojejunostomy was performed. Gastric suction was maintained for 3 days; and feedings tolerated from the fifth day following operation. The infant was discharged from the hospital on the eighth day following admission

weighing 2,650 Gm., and the subsequent course (3 months) has been uncomplicated.

Case 5 (U.C.L.A. 015-80-56). Birth weight 2,140 Gm. Polyhydramnios complicated this pregnancy. Breech delivery was followed by neonatal respiratory distress requiring resuscitation with stimulants and a respirator. Imperforate anus and cryptorchidism were noted; but the abdomen was not distended. When respiratory function improved, an attempt to pass a gastric tube disclosed an esophageal obstruction. A radiographic study of this area using a contrast medium revealed an atresia of the esophagus; and also, incidentally, the dilated stomach and duodenum of complete duodenal obstruction (Fig. 3). On the day following birth a transverse colostomy and gastrostomy to facilitate aspiration were performed. Respiratory distress recurred and the infant expired on the third day following operation. Autopsy findings included bilateral atelectasis and bronchopneumonia as well as a cerebral hemorrhage into the parietal lobe. The diagnoses of esophageal atresia with tracheo-esophageal fistula, imperforate anus (Ladd, Type III), and annular pancreas were confirmed.

Case 6 (C.H. 92747). Birth weight 2,586 Gm. Marked polyhydramnios was noted during pregnancy. Excessive pharyngeal secretions (said to be bile-stained) were apparent following birth, and a tube could not be passed to the stomach.

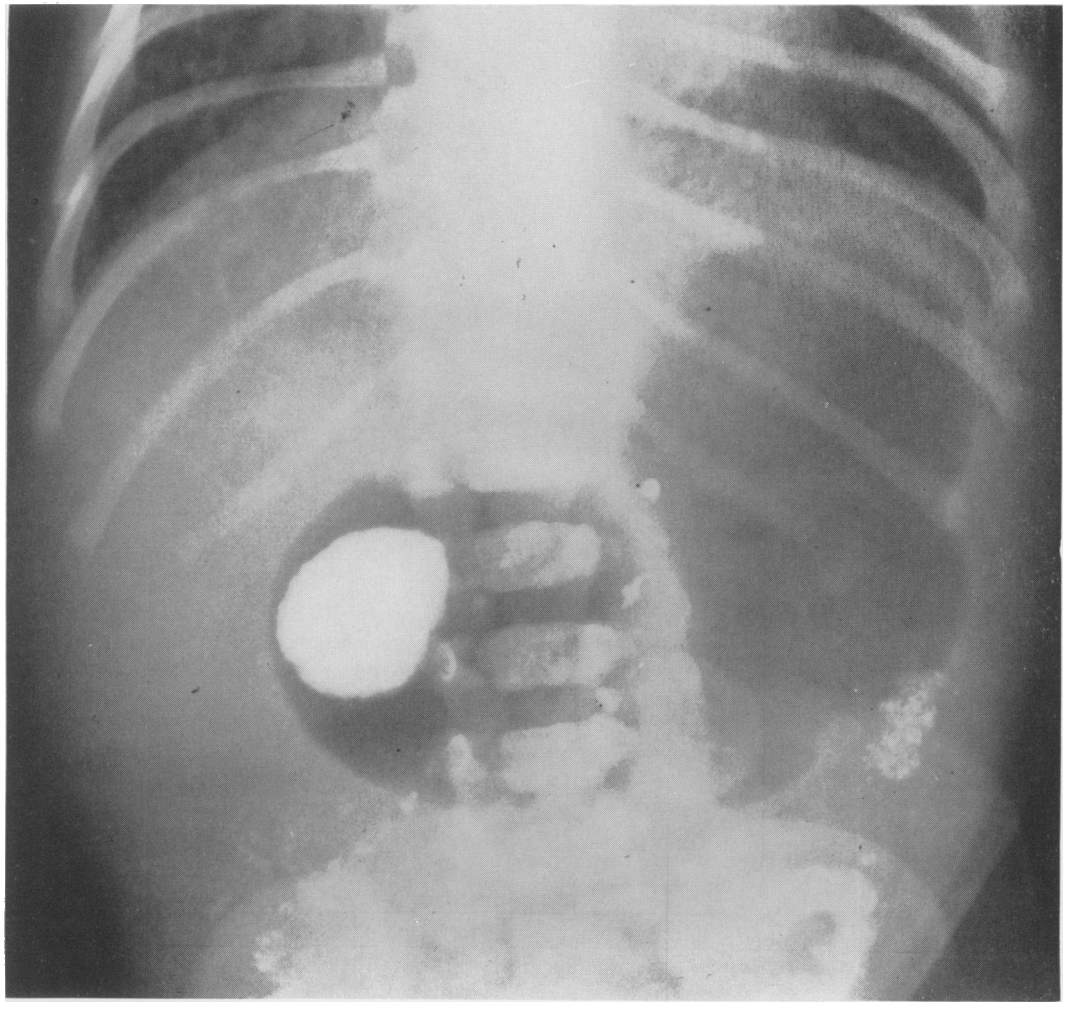

FIG. 2. Six-day-old male infant (Case 4) with history of nonbile stained vomiting since birth. Film taken in the supine position demonstrating incomplete duodenal obstruction. No significant additional information is obtained by the use of barium or other contrast media.

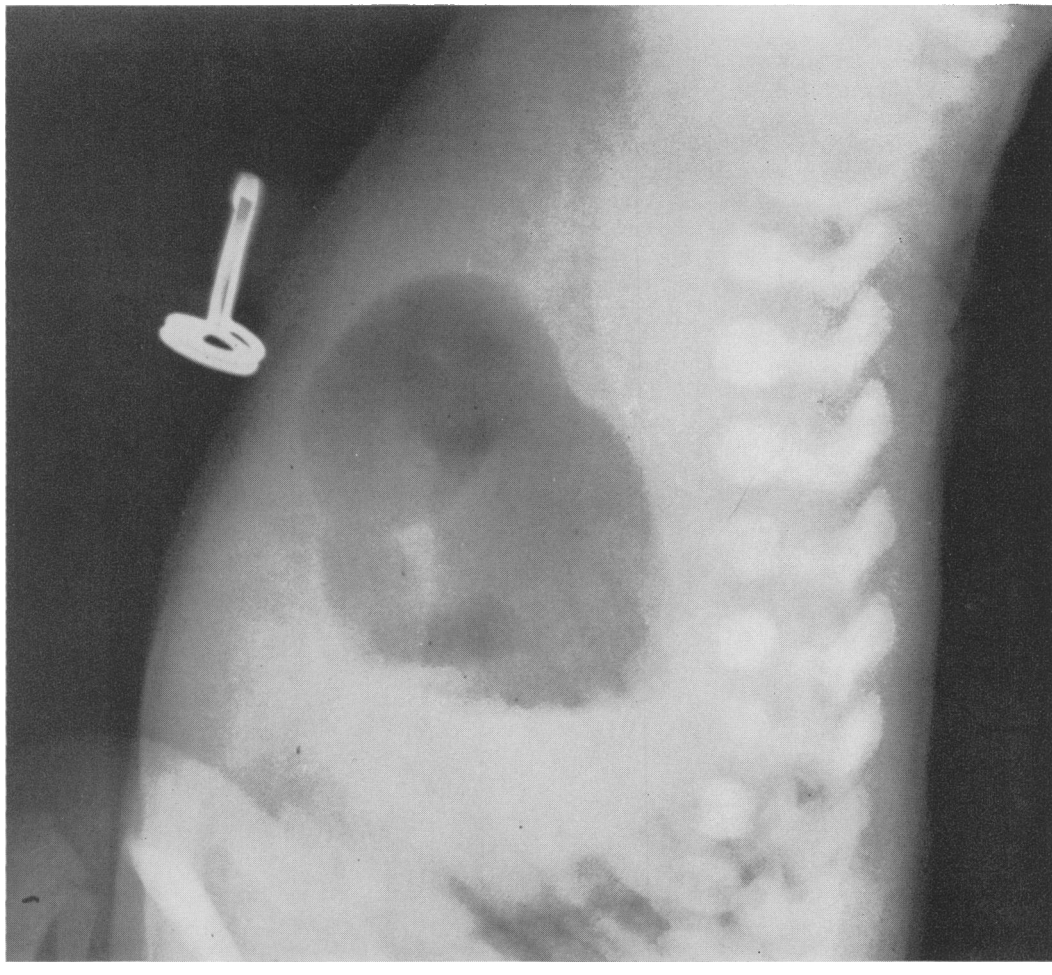

FIG. 3. One-day-old male infant (Case 5) with an imperforate anus noted at birth. X-ray exposures in the inverted position incident to the discovery of this anal anomaly revealed the detailed study of the obstructed second portion of the duodenum noted in this lateral film.

The infant was transferred from the lying-in hospital (age, 2 days) with a diagnosis of esophageal atresia. Physical signs of pneumonitis were noted over the right upper thorax. The abdomen was not distended. Roentgenographic studies revealed a dilated stomach and duodenum in addition to confirmation of the esophageal anomaly. Barium enema studies demonstrated normal colon position.

On the day of admission a transpleural division of the tracheo-esophageal fistula and primary esophageal anastomosis was performed. Under the same anesthetic, a laparotomy was carried out, revealing an annular pancreas. Duodenojejunostomy was accomplished.

The postoperative course (3 days) was characterized by excessive (bile-stained) vomiting; followed by bile-stained pleural drainage. Autopsy revealed an esophagopleural fistula from the site of esophageal anastomosis. The duodenojejunostomy was intact and apparently patent. The choledochus entered the duodenum distal to the site of the congenital obstruction.

Case 7 (C.H. 104542). Birth weight 2,650 Gm. Pregnancy was uncomplicated and no congenital abnormalities were noted at birth. Because of persistent nonpigmented vomiting, transfer to this hospital carried out on the fourth day. Physical examination at this time revealed the stigmata of Mongolism. There was no abdominal distention. Roentgenographic studies demonstrated duodenal obstruction, pneumonitis and atelectasis. There was marked albuminuria and pyuria. Operation (age, 5 days) revealed the caecum in

the subhepatic area and, on mobilizing this structure, an annular pancreas was identified. A duodenojejunostomy was performed. The postoperative course (24 days) was initially characterized by improvement with retention of all feedings; and later by generalized sepsis which failed to respond to intensive antibiotic therapy.

Autopsy findings included: (a) annular pancreas, incomplete at one point for several millimeters (the surgeon thought this completely annular), (b) duodenal atresia, which extended for at least one centimeter longitudinally, (c) a patent, intact duodenojejunostomy, (d) focal acute hepatitis, (e) multiple brain abscesses and internal hydrocephalus, (f) pyelonephritis, (g) bronchopneumonia and (h) malrotation of the colon and Meckel's Diverticulum.

Summary of Case Reports

Four of the seven gestations associated with this anomaly were complicated by polyhydramnios. A fifth had a maternal history of habitual abortion. Two infants weighed less than 2,500 Gm. at birth (No. 2 and No. 5).

In three infants, annular pancreas was the only significant congenital anomaly; and all of these survived, despite the prematurity (1,984 Gm.) of one. Mongolism was apparent in two patients. Other significant congenital defects and their frequency in this group of infants (7) were as follows: esophageal atresia with tracheoesophageal fistula (2), imperforate anus (1), duodenal atresia * (1), malrotation of the colon (1), cryptorchidism (1), and Meckel's Diverticulum (1).

Vomiting was noted following the first or second feeding and was free of bile pigments in all infants.** Two patients (No. 1 and No. 7) had clinical icterus; the latter probably secondary to primary hepatic disease. When examined on admission all demonstrated varying degrees of dehydration and an absence of significant abdominal distention.

A diagnosis of complete or almost complete duodenal obstruction could be made from examination of roentgenograms of the abdomen without contrast media in all cases. In one, this technic was aided by the intragastric instillation of air. No barium or other contrast media was given orally following admission. Barium enema was employed in two instances in an attempt to demonstrate colon malrotation.

Exploration was performed in most instances through a transverse upper abdominal incision. The initial survey of the abdominal contents revealed a dilated (1–4 cm. diameter) intestinal loop in the subhepatic and epigastric areas; while the intestine distal to the ligament of Treitz was minute (3–4 mm. in diameter). The pyloric ring was often patulous. Mobilization of the hepatic flexure permitted a clear visualization of the entire duodenum, eliminating the possibility of transduodenal bands as a source of the obstruction. Pancreatic tissue was found to completely encircle the duodenum, either in the second portion or at the junction of the second and third portions, in six of the infants. In the seventh (at autopsy) the ring was found to be incomplete by several millimeters.

Six infants were treated by duodenojejunostomy. A retrocolic anastomosis employing the most proximal jejunum which could be easily brought to the wall of the dilated duodenum through the mesocolon, was employed. No attempt to remove, dissect or divide pancreatic tissue was made in any of these cases.

The standard "open" anastomosis was formed by an external row of 5–0 inter-

* This was believed to represent primary atresia rather than a secondary effect of the annular pancreas because the absence of duodenal lumen extended longitudinally for approximately one centimeter.

** Case number six was said to have had bile-stained mucus in the pharynx prior to the duodenal anastomosis. In this, as in the other two infants upon which autopsy studies were carried out, however, the choledochus entered the duodenum distal to point of obstruction making this observation unlikely.

rupted silk sutures,† and an internal continuous chromic gut suture joining the mucosal surfaces. It lay either transversely, vertically or obliquely, depending on the position of the dilated duodenum and the minute jejunum.

In four cases, the postoperative course was uncomplicated. Death in the other three infants was the product of complicating factors, i.e., atresia of the esophagus with tracheo-esophageal fistula and Mongolism with generalized sepsis.

Discussion

Annular pancreas is usually clinically associated with chronic or intermittent upper intestinal obstruction in adults; or pathologically, with interesting necropsy findings in individuals dying from unrelated causes. However, it may also be the etiological factor in acute neonatal obstruction of the duodenum. In this situation it simulates the far more prevalent conditions of (a) malrotation of the colon with transduodenal bands and (b) intrinsic duodenal atresia or stenosis.

The incidence of maternal polyhydramnios associated with fetal annular pancreas, four of seven cases in this series, is similar to the incidence (65.7%) reported by Lloyd and Clatworthy [14] for obstructive lesions of all types found in this portion of the intestinal tract. The duodenal obstruction probably represents a cause, rather than an effect,[14] of the maternal complication.

Intestinal obstruction in these infants is acute and vomiting begins when feedings are attempted at 24 to 48 hours of age. Abdominal distention is absent entirely or well localized to the upper abdomen. Simple roentgenograms, if necessary supplemented by the introduction of air into the stomach, are diagnostic of functionally complete obstruction. The presence of two, and only two, visible loops (often with fluid levels) in the upper abdomen, i.e., the "double bubble," is the classic and usual roentgenographic finding. Demonstration of the obstructed and dilated proximal duodenum may be even more striking, producing a "dumbbell" shaped shadow (Fig. 1). Contrast studies of the upper gastrointestinal tract contribute little. No more accurate preoperative diagnosis is possible or necessary in cases of annular pancreas. The differential diagnostic possibilities, i.e., malrotation with duodenal bands, intrinsic stenosis, and intrinsic atresia, are conditions which also demand immediate exploration of the duodenal area. A barium enema adds no vital data; but may be helpful in planning the management of infants with esophageal atresia combined with duodenal obstruction of an unknown type.

The principle, well-documented, operative cases of annular pancreas reported during the period 1944–1959—which meet the criteria previously noted—are outlined (Table 1). The addition of seven infants in this report, raises the total during this period to 40 cases. Sixty-eight per cent of these infants were the products of full-term pregnancies and free from associated anomalies of significance. In this group of 27 apparently otherwise normal infants, the mortality of operative correction of annular pancreas was 31 per cent.

The remaining 13 infants (32%) had the major complicating features outlined in Table 1. The mortality of operative repair in this group was high (69%).

It is a tribute to the influence of Gross,[7] that only two of these 40 infants were subjected to pancreatic dissection (with fatal results); while prior to his published advocacy of duodenojejunostomy (1944) such dissections were usual. Division of the annulus has occasionally been successful in the adult type of this disease; but in the newborn it has lead to catastrophy. Among the 40 collected cases noted in Table 1, four were treated by gastro-

† In Cases #1 and #7 anastomosis was made with a single layer of interrupted silk sutures.

jejunostomy without a recovery (two had associated esophageal atresia). Four were subjected to what was described as "gastroduodenostomy" (2) or "duodeno-duodenostomy" (2) with three recoveries. Duodenojejunostomy was performed as the sole procedure in 29 infants with 19 survivors (66% recovery).

Thus the current series, as well as the collected data, supports the efficacy of the indirect operative approach to annular pancreas and suggests that duodenojejunostomy is preferable to gastroenterostomy as a "bypass" procedure. Cases of annular pancreas in the newborn associated with hyperinsulinism (Beck) have not been reported in this country; and cases in which the biliary tract was significantly obstructed by the annulus, only by Ravitch[17] (1 case).

Annular pancreas was associated with esophageal atresia and tracheo-esophageal fistula in two infants in the present series, and has been noted four times in previous reports. None of these infants has recovered, although the mortality for each of these conditions occurring separately is steadily improving and should be regarded as less than 50 per cent. The recommended sequence of surgical procedures in this situation is: (a) primary esophageal repair plus gastrostomy (which is immediately placed on suction), followed in 48 hours by (b) duodenojejunostomy (or division of transduodenal bands if malrotation is discovered). Reversal of this sequence, i.e., performing the duodenal operation initially as advocated by McGarity,[15] has the following inherent disadvantages: (a) the tracheo-esophageal fistula will be subjected to the pressures of the obstructed gastric contents for several days (our duodenojejunostomies have not functioned adequately for two to three days following anastomosis), or (b) gastrostomy will be performed at the time of duodenojejunostomy and may restrict subsequent upward mobilization of the distal esophagus. In this series one such case was treated by initial laparotomy and a second by performing both anastomis at a single procedure. These terminated fatally.

When imperforate anus is added to this combination of anomalies (annular pancreas and esophageal atresia) as in Case 5, a transverse colostomy must be included in the second procedure.

Summary

Seven cases of acute obstruction of the duodenum in the neonatal period secondary to annular pancreas are described. Duodenojejunostomy was performed in six instances, with survival in four. Three fatalities (two following duodenojejunostomy) were associated with complications attendant to the management of other major anomalies.

Analysis of the results of operative treatment of 40 collected cases of annular pancreas producing intestinal obstruction in the neonatal period suggests that: (a) operative dissection or division of the pancreatic annulus is contraindicated from both theoretical (embryological), and practical considerations; (b) duodenojejunostomy has proved a more effective "bypass" than gastroenterostomy; and (c) the association of this condition with esophageal atresia occurs with sufficient frequency to warrant the development of a plan of management of this dual condition, which has to date been uniformly fatal. A two-stage procedure with initial esophageal anastomosis and secondary abdominal exploration seems advisable.

Maternal polyhydramnios was noted in the majority of these infants. Among the collected cases, 32 per cent had additional major anomalies or were premature by weight. When this group was excluded, the mortality of operative repair of neonatal annular pancreas was reduced to 31 per cent.

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