ANNULAR PANCREAS*

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ANNULAR PANCREAS is a sufficiently uncommon and infrequently reported anomaly so that its presence is rarely suspected. This is the more true because it is probably the only congenital anomaly of the gastro-intestinal tract which usually produces symptoms late in life. There have now been reported some 50 instances of this anomaly, most of them purely anatomic descriptions of necropsy findings. The malformation was given its aptly descriptive name in 1862 by Ecker,[10] writing in Henle's Zeitschrift. In the course of an anatomic demonstration on the body of a young man, he found "a strip of glandular substance which lay across the descending portion of the duodenum. Closer investigation showed this to be the anterior portion of a ring derived from the head of the pancreas which surrounded the descending portion of the duodenum and was formed by uninterrupted glandular tissue." This "ring formigen" portion of the pancreas contained one duct which communicated superiorly and posteriorly with the main pancreatic duct. Schirmer[20] in his 1893 dissertation on the pancreas erroneously stated that Tiedemann,[24] Bécourt,[1] and Moyse[19] had observed cases of annular pancreas and reference to these authors is still made in most discussions of annular pancreas. Review of these reports, however, shows that the writers were either discussing the comparative anatomy of the pancreas and the occurrence of annular pancreas among the birds or else were discussing carcinoma of the pancreas or other acquired encircling lesions in this area. There are 14 recorded instances of operation for annular pancreas. Most reports are of one or two cases, individual experience with a lesion of this rarity being necessarily limited. Among the most helpful discussions are those of Howard,[13] McNaught,[18] Gross and Chisholm,[12] Goldyne and Carlson,[11] and Burger and Aldrich.[4]

At operation or autopsy, one finds a thin flat band of grossly recognizable and apparently normal pancreatic tissue anterior to the duodenum in its second portion. This band is continuous with the head of the pancreas on both the convex and the concave surfaces of the duodenum, which is thus encircled by pancreatic tissue. In the instances which we have seen, there is no point of demarcation between this anterior band and the head of the pancreas with which it is continuous. On the other hand, in many of the instances previously described, and in one of our cases, the band of misplaced pancreatic tissue lies loosely upon the duodenum and may be lifted away from it. It is thus quite different from that other anomaly, *aberrant pancreas*,[8] in

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which an isolated nodule of pancreatic tissue lies buried in the wall of the stomach, jejunum, or ileum, frequently submucosally.

The anterior band of pancreatic tissue often contains a fairly large duct which has most often been reported to connect with the main pancreatic duct of Wirsung. In Cunningham's[6] case, the abnormal portion of pancreas had a duct system of its own which entered into the common bile duct on a slightly posterior plane to the entrance of the main pancreatic duct.

The generally accepted embryologic explanation for this malformation depends upon the development of the pancreas from two anlagen, dorsal and ventral, making their appearance as buds from the midgut in the 3 to 4 mm. embryo before rotation of the gut takes place. In the course of development the dorsal anlage usually forms the body and tail of the pancreas and part of the head and uncinate process. The ventral anlage contributes to the head of the pancreas. Its duct joins with the distal portion of the dorsal duct to form the duct of Wirsung. The proximal portion of the dorsal duct becomes the accessory duct of Santorini. It is postulated that, in rotation of the duodenum, pancreatic tissue from the ventral anlage may be carried with the second portion of the duodenum, subsequently fusing its free end superiorly to the head of the pancreas, creating the annular deformity. Chapman and Mossman[5] are inclined to think that the ring is produced by overgrowth of the ventral anlage around both sides of the duodenum, fusing to the head of the pancreas. Weissberg[28] found an annular pancreas in a 16-millimeter embryo (approximately six to seven weeks). At this early stage the annular ring was quite well formed. It appeared to originate from the ventral anlage and had expanded about the duodenum. On the posterior portion of the duodenum, the pancreatic tissue lay beneath the peritoneum and actually in the wall of the bowel. Weissberg feels that neither abnormal persistence of the ventral anlage nor anomalous fixation and rotation with the duodenum are responsible for the formation of annular pancreas, but that there may be simply an unusual growth of one of the ducts of the pancreas itself which the local mechanical situation may direct in this way.

CLINICAL CHARACTERISTICS

In many instances, the condition is asymptomatic, and the annular pancreas is described as a curious, coincidental necropsy finding. In 14 cases—17, with the inclusion of the three herein reported—symptoms were produced of severity sufficient to warrant operation. In one of our patients (J. K.) and in the case of Lehman reported in 1942, the correct diagnosis was made before operation. Sixteen of the 17 patients developed signs of high intestinal obstruction. The seventeenth was operated upon for peritonitis found to be due to acute pancreatitis. Most of the patients were well on in years when symptoms began, and only four were in the neonatal period. Ten of the 17 patients were male. Only one patient was colored. In the older patients the appearance of indigestion, eructation, and vague epigastric pain preceded the onset of frank intestinal obstruction.

The diagnosis may be made in patients with chronic duodenal obstruction who show by roentgen ray an almost complete, smooth and sharp obstruction of the duodenum to the right of the midline. The diagnoses which have been erroneously made in the past are arterio-mesenteric ileus, cholecystitis, primary carcinoma of the duodenum, cicatrizing peptic ulcer, malrotation of the bowel, duodenal atresia, etc.

The barium meal, although rather characteristic, has often failed to lead to the diagnosis of annular pancreas. In one of our cases the roentgenologic report was "unusual dilatation of the distal third of the stomach." In Lehman's case[15] the roentgenographic diagnosis was first "a polyp of the second portion of the duodenum," and this then changed to "constricting ulcer."

In view of the rarity of annular pancreas, surgeons are justified in giving greater consideration to such rare conditions as carcinoma of the duodenum in adults and duodenal atresia in newborn infants. In 20,000 autopsies at the Johns Hopkins Hospital, annular pancreas has not been noted even once.

TREATMENT

One is dealing with a mechanical duodenal obstruction and operative interference is required for relief. Direct attack upon the constricting portion of the ring has been attempted seven times in all, including the present cases. In these instances, the ring has been divided, separated from the duodenum, and a portion resected.

Six of these seven patients survived, the seventh dying on the ninth day after operation. No autopsy was performed, but bile-stained drainage appeared from the wound. Three of the six surviving patients had persistent pancreatic drainage, and one of the three had a second operation for drainage.

In Lehman's case[15] of resection of the ring, the patient survived without fistula formation, but had persistent symptoms and roentgenograms showed persistent duodenal deformity. Similarly, one of our cases, despite the readiness with which the ring was resected, developed progressive duodenal obstruction and had to be operated upon a second time. Two of the seven patients also had a plastic procedure on the duodenum—one, the fatal case mentioned, and the other with successful outcome.

In summary, direct attack upon the annular pancreas has now been employed seven times with one straightforward cure, one death, three instances of prolonged pancreatic drainage, and two instances of persistence of symptoms. In addition, in one case,[3] annular pancreas was associated with acute hemorrhagic pancreatitis for which simple drainage was instituted. The annular pancreas was discovered at autopsy.

The second alternative, a by-pass operation, was historically the first method of treatment[27] and has now been performed ten times. There have been five gastro-enterostomies, one gastric resection in a patient who had a coincident peptic ulcer,[7] three duodenojejunostomies, and one gastro-duodenostomy. Three of the patients with gastro-enterostomy died—two of upper respiratory infections and one several hours after operation, presumably of

shock. All three of these deaths occurred prior to the days of antibiotics and multiple transfusions. One of our patients with a duodenojejunostomy had previously had a partial resection of the pancreatic ring without improvement.

CASE REPORTS

Case 1.—J. K., J. H. H. No. 317888. This was a 67-year-old man admitted to the Johns Hopkins Hospital on January 21, 1949. Fifteen years prior to admission the patient first developed intolerance to fatty food. Three years prior to admission he developed epigastric pain after meals, at first relieved by soda but later becoming more severe and persistent. In recent months he had vomited frequently and had often complained of headaches and a foul taste in his mouth. He had lost approximately 10 pounds in weight.

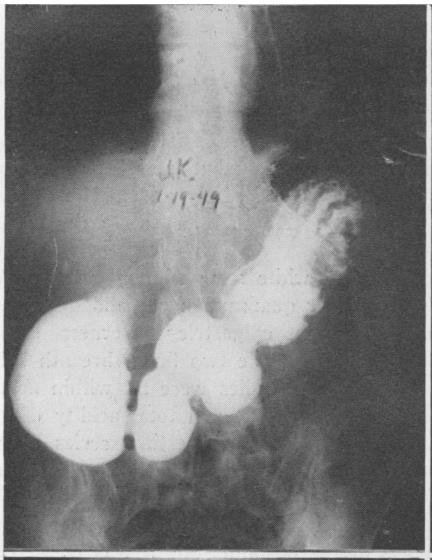
Physical examination revealed an undernourished white male who appeared chronically ill. There was a definite bulge in the right upper quadrant of the abdomen where peristaltic movements could be seen. There was slight epigastric tenderness, but no masses or muscle spasm could be felt. The liver was palpable two fingersbreadth below the right costal margin on inspiration. Blood and urine studies were all within normal range. Gastric analysis showed free hydrochloric acid of 18° and total acidity of 52°, rising to 134° and 172° respectively after histamine. A gastro-intestinal series showed what at first was thought to be almost complete pyloric obstruction with "unusual dilatation of the distal third of the stomach" (Fig. 1, A and B; Fig. 2). A small focus of calcification seen in the right upper quadrant was thought to be a calcified lymph node. In the discussion of the patient by the members of the hospital staff it was pointed out that the "dilated third portion of the stomach" seen on the gastro-intestinal series might well be duodenum. Among the diagnoses suggested were chronic duodenal ulcer, chronic cholecystitis, arterio-mesenteric ileus, and annular pancreas (advanced as a definite diagnosis by one of us).

Operation was performed (A. W., Jr.) on January 27, 1949, through a transverse incision. The first portion of the duodenum was found to be dilated and flabby. The stomach and pylorus appeared normal. The descending portion of the duodenum was mobilized. Just inferior to the point at which the common duct entered the duodenum, a broad band of pancreas completely encircled the duodenum, causing almost complete obstruction. Beyond this band the duodenum appeared normal in size. The annular portion of the pancreas was approximately two centimeters wide and one-half to one centimeter thick. It was elected to remove the anterior and lateral portions of the encircling band of pancreas. This was easily accomplished. No large pancreatic duct was identified. The cut ends of the pancreatic ring were transfixed with mattress sutures of silk. The lumen of the duodenum then appeared open, and it was thought that the obstruction was relieved. A drain was placed in the vicinity of the cut ends of pancreas and brought out through a stab wound in the right flank. Pathologic examination of the resected portion of the pancreatic ring revealed only normal pancreatic tissue.

The first week of the postoperative course was uneventful. Very little drainage was noted. On the seventh postoperative day the patient became distended and vomited, and on successive days he vomited several times. On the ninth day after operation, overnight gastric retention was found to be 400 cc., and he had a serum amylase of 518 mg. per 100 cc. A Levine tube was passed into his stomach and attached to constant suction. He was maintained on intravenous feedings. On the eleventh day the serum amylase was 756 mg. per 100 cc. The Levine tube was removed on the sixteenth day and he was begun again on a soft diet by mouth. The serum amylase thereafter was at normal levels. He was discharged on the nineteenth postoperative day, at which time he was without complaint and taking a full soft diet well.

Shortly after his return home he began again to have symptoms of epigastric fullness and he vomited on several occasions. On one visit to the hospital, one liter of

FIG. 1-A



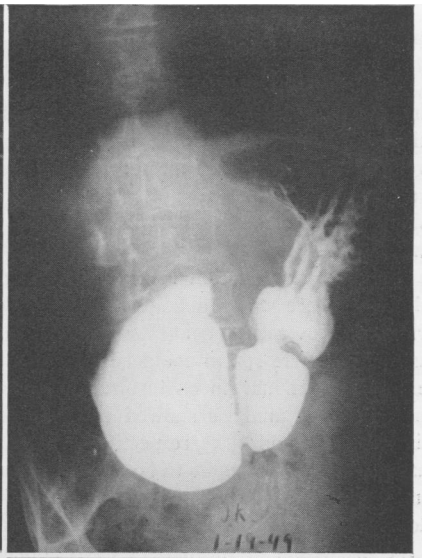
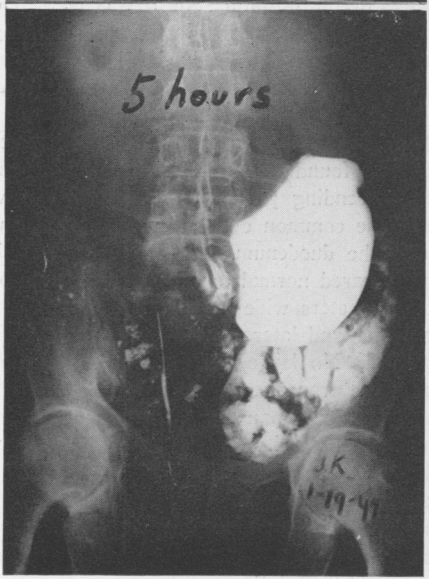


FIG. 1-B

FIG. 2



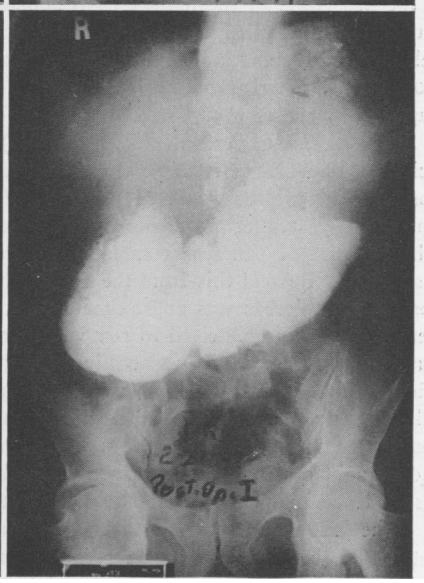


FIG. 3

FIG. 1.—(Case I) J. K., white male, age 67, No. 317888 (A and B) Antero-posterior and lateral views of the barium swallow. There is a tremendous dilatation of the duodenum ending quite abruptly at a smooth obliquely transverse obstruction in the second portion. There is no puckering or filling defect.

FIG. 2.—(Case 1) Five hour film demonstrates degree of gastric retention.

FIG. 3.—(Case 1) Barium swallow 25 days after resection of pancreatic ring—duodenum and stomach are more dilated than before.

gastric contents was removed by lavage. A gastro-intestinal series was obtained and showed the original dilatation of the duodenum plus marked additional distention of the stomach which had not been present before (Fig. 3. He was re-admitted to the hospital on March 5, 1949, and the original incision was reopened (A. W., Jr.). The duodenum was again seen to be markedly dilated in its first portion and had not changed in size. The pylorus, however, had become patulous and would admit three finger tips easily. The stomach had increased greatly in volume, and the gastric wall appeared hypertrophied. A thick band of scar tissue was found in the region of the excised annular portion of the pancreas. This thick band of scar passed anterior to the duodenum and ran from the transected end of the pancreatic ring to the lateral abdominal wall at the site of the stab wound made for the drain. The duodenum was still narrow at the site of the original obstruction, but the present obstruction seemed caused by the dense band of scar tissue. The jejunum was identified at the ligament of Treitz, and a loop approximately 40 cm. distal was brought up anterior to the colon and anastomosed to the anterior surface of the dilated first portion of the duodenum with two rows of interrupted sutures of fine silk.

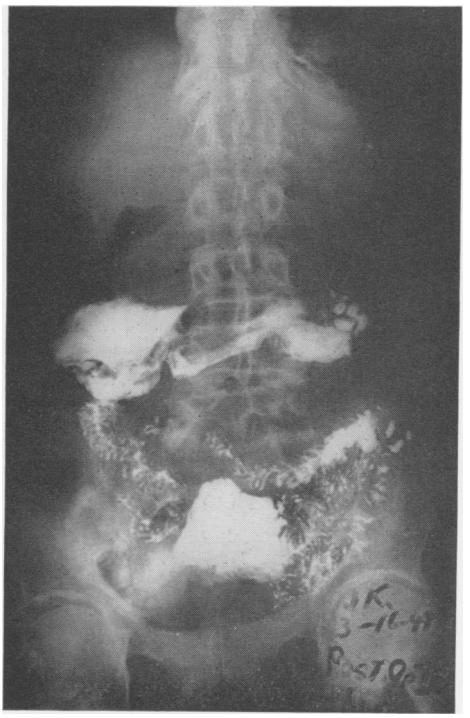


FIG. 4.—(Case 1) Barium swallow (one hour film) 11 days after second operation—duodeno-jejunostomy. The patient has remained well since.

Postoperatively, the patient was maintained for three days on intravenous feedings while constant gastric suction was applied to an indwelling Levine tube. Following this period he was fed a gradually increasing liquid diet until the tenth postoperative day when he was placed on a full soft diet. He was discharged from the hospital, asymptomatic, on March 18, the fourteenth postoperative day.

On one occasion in the Out-Patient Department, he complained of epigastric fullness but a gastric aspiration at that time revealed only 100 cc. of fluid in his stomach. A gastro-intestinal series (Fig. 4) showed a satisfactorily functioning duodeno-jejunostomy, though the stomach was still dilated. He was last seen in June, three and one-half months after the second operation, when he felt entirely well and had gained 17 pounds since his discharge from the hospital.

Comments. This was a 67-year-old white male who had a 15-year history of indigestion and a three-year history of more specific symptoms. Annular pancreas was diagnosed from the roentgenograms. Division of the annulus and resection of a portion of it produced a local acute pancreatitis resulting in a dense scar obstructing the duodenum more completely than before. Duodeno-jejunostomy afforded complete relief.

Case 2.—M. G., No. A-68776. The patient was a full-term white female whose delivery was uneventful. Beginning shortly after birth she vomited thin, bile-stained material

at frequent intervals and was able to retain nothing by mouth. Physical examination revealed an alert white female infant, externally normal, and presenting no other evidence of any congenital malformation. Weight on transfer to the surgical service, when she was three days old, was 2770 Gm. Roentgen ray examination (Fig. 5) showed a large, dilated stomach, no dilated loops of intestines, and a moderate amount of gas in the large bowel. The preoperative diagnosis was partial high intestinal obstruction secondary

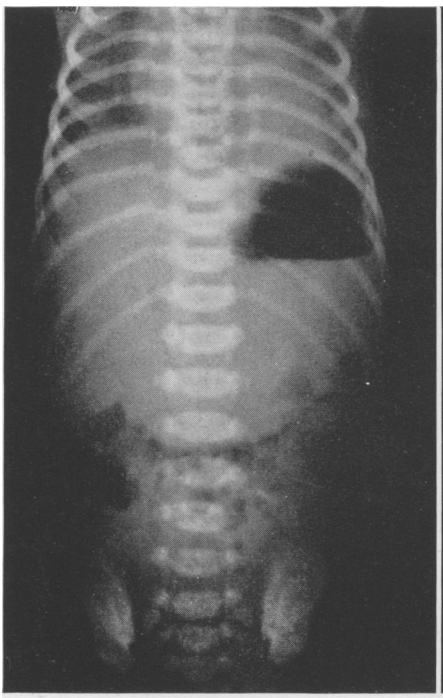


FIG. 5

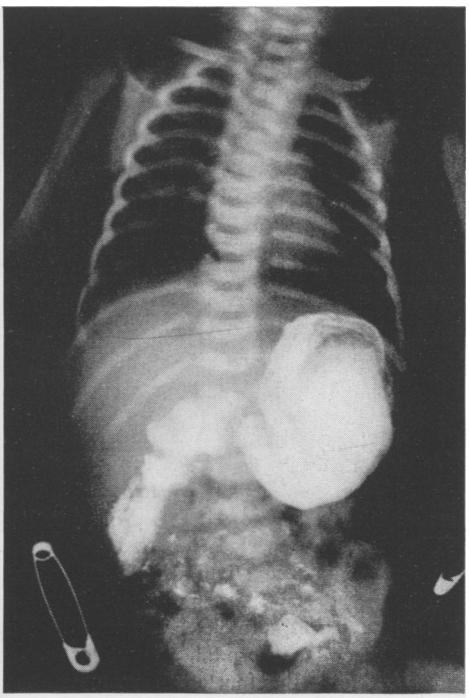


FIG. 6

FIG. 5.—(Case 2) M. G., white female, age three days, No. A-6876. Roentgenogram on third day of life showing stomach dilated with air. The presence of some air in the lower intestinal tract demonstrates the obstruction to be incomplete.

FIG. 6.—(Case 2) Roentgenogram of barium swallow taken 14 days after duodeno-jejunostomy. Barium passes at once from the still dilated duodenum into the jejunum whose plicae circulares are plainly seen.

to a partial duodenal diaphragm, malrotation, or some other congenital malformation.

On February 17, 1949, under open-drop ether anesthesia, laparotomy was performed (M. M. R.) through a transverse incision in the right upper quadrant. When the abdomen was opened the small intestine was found to be collapsed almost completely, but it was not quite as small as in a true atresia. There was meconium in the colon. No abnormality was found in the jejunum. The stomach, which had been emptied by suction, was large and thick walled, and the proximal duodenum for 2 to 3 cm. was almost as large as the pre-pyloric region of the stomach (Fig. 5). Distal to this dilated portion of duodenum, the duodenum was encircled by a band of pancreas about 1 cm. wide, beyond which the duodenum was no larger than was the jejunum at the ligament of Treitz—in other words, smaller than normal. A brief attempt was made to dissect the pancreas from the duodenum, but it was so intimately attached that the operator

thought the dissection would be fraught with danger. Accordingly, a duodeno-jejunostomy was performed, bringing up anterior to the transverse colon a loop of jejunum some distance below the ligament of Treitz. A two-layer anastomosis was accomplished, using an outer continuous suture of 5-0 silk and an inner continuous suture of 4-0 catgut. The patient's condition throughout the procedure was good, and she received 100 cc. of whole blood during the course of the operation. The wound was closed with silk without drainage.

The patient's postoperative course was uneventful except for the development of edema of the epiglottis secondary to the indwelling Levine tube which was used for gastric suction for 48 hours. This difficulty vanished with the removal of the tube. The incision healed per primam. The patient began to take oral feedings well on the fourth postoperative day and was discharged from the hospital on March 8, 1949, 19 days after operation, apparently well and gaining weight satisfactorily on a standard formula. A G. I. series performed prior to her discharge (Fig. 6) showed good function of the anastomosis, though the duodenum was still dilated.

Comments. This was a thre-day-old female with incomplete duodenal obstruction due to an annular pancreas. Duodenojejunostomy gave complete relief.

Case 3.—R. L., No. A-52869. The patient was born in a doctor's office in a rural district of Maryland, in an otherwise normal delivery. The child and the mother were apparently sent home the same day. On the third day the doctor was told that the baby was vomiting and had passed no stools. The infant was found to be in critical condition, jaundiced, dehydrated, feeble, and vomiting small amounts of greenish mucus. Temperature was 102°. The baby was given a subcutaneous infusion and transferred to the Harriet Lane Home. He was then six days old. His birth weight had been 6 lbs. 9 oz. Vomiting was said usually to occur three hours after each feeding. One sibling, a girl of four, had previously been diagnosed as a Mongolian idiot. Physical examination showed the baby to a a scrawny-looking undernourished infant weighing only 1950 Gm. He was quiet, obviously ill, and deeply jaundiced. A plain roentgenogram of the abdomen showed a large collection of air in the stomach with no gas visible in either the small or large intestines (Fig. 7). Barium administered by mouth remained within the stomach for four or five hours (Fig. 8). Surgical consultation was requested at this point, and the general consensus was that the baby had a congenital duodenal obstruction which required operative relief. Exploration was performed (M. M. R.) on March 23, 1947, when the patient was 8 days old. The abdomen was entered through a transverse incision just above the umbilicus. The stomach was much enlarged and thick-walled. All the intestines were delivered, and the small bowel followed up from the cecum, which was peculiarly situated near the midline and rather high up. There was no fixation of the mesentery of the small bowel, the bowel being suspended largely by its vessels. There was no ligament of Treitz. The duodenum coiled and kinked several times on itself in the right upper quadrant, the coils and kinks being held together by what could only be described as organized adhesions. Some of these were opaque and suggested a reaction of inflammatory origin more than anything else. All of the small bowel was tiny and contained no air or meconium, while the proximal duodenum was as large as the pyloric antrum of the stomach. The distal duodenum was clamped off with a rubber shod Allis clamp about two inches below the stomach, and the segment proximal to the clamp injected with saline solution under pressure. The duodenum was distended in this manner to the size of one's little finger, until it was extremely tense. The fluid, however, did not run into the stomach, and by inverting the pyloric antrum with one's finger one could palpate the proximal end of the tensely distended saline-filled segment of the duodenum. The level at which this duodenum ended blindly was marked anteriorly by the passage over it of the portal vein. This was unmistakable and readily demonstrated. The superior and inferior mesenteric vessels joined and the

portal vein formed by their junction passed anterior to the duodenum precisely at the level of what must have been an intrinsic obstruction. At this point, too, the pancreas passed around the duodenum as an annular pancreas, the anteduodenal portion of which was just at the level of the abnormally situated portal vein. The annular portion of the pancreas connected uninterruptedly with the head of the pancreas above and below (Fig. 10A). It seemed impossible for any extrinsic obstruction to have resisted the great hydrostatic pressure induced by the injection of saline into the isolated duodenal loop below the obstruction. The loop of duodenum which was injected did not contain any bile. We were forced to the conclusion that this was a complete atresia. This feeling was bolstered by the report from the Pathological Department that the rectal discharge before operation, which was in the nature of a greenish mucous plug, did not contain any squamous epithelium or lanugal hair. The gallbladder was normal and distended with very dark bile. No attempt was made to follow up the common duct, although it appeared to enter the duodenum just proximal to the obstruction. The duodenum proximal to the obstruction was as dilated as the stomach, and could hardly be told from it. The stomach was anastomosed to the duodenum just beyond the obstruction, using

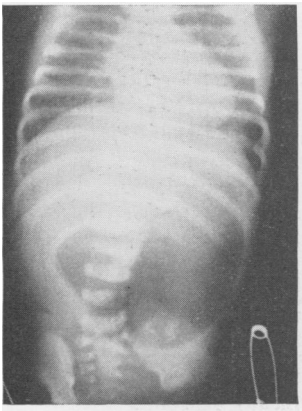


FIG. 7

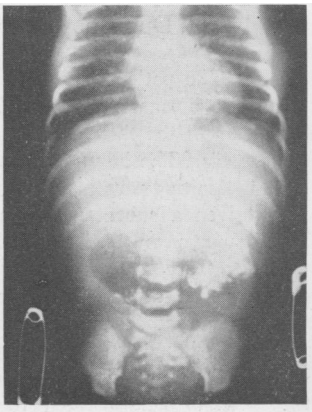


FIG. 8

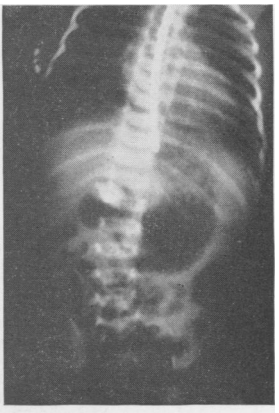


FIG. 9

FIG. 7.—(Case 3) R. L., white male, age eight days, No. A-53869. Preoperative roentgenogram showing tremendous dilatation of stomach and duodenum and absence of air elsewhere in the intestine.

FIG. 8.—(Case 3) Barium has unwisely been administered. The picture is less clear than before. The barium swallow is an unnecessary and dangerous measure in the diagnosis of neonatal intestinal obstruction.

FIG. 9.—(Case 3) Roentgenogram two weeks after gastroduodenostomy. The intestinal tract contains air throughout. A little barium persists in the still dilated duodenum. (The child is well two and one-half years after operation.)

a double row of sutures—outer of continuous 00000 silk and inner of continuous 00000 catgut.

The colon was found now to present anomalies both of rotation and of fixation. Traced upward from the pelvis the colon went straight up the abdomen on the right side, crossed over to the left, made a rather large loop and passed behind itself to emerge on the right side with little more than the cecum to the right of the vertically ascending limb first described. The colon was fixed in this position by fibrous bands which again suggested adhesions like those which held the duodenum. As these bands were released and the colon uncoiled, its mesentery was found to have no fixation at all.

Postoperatively the child was maintained on parenteral fluids. Nothing was administered by mouth, and constant gastric suction was employed for three days until serial

roentgenograms showed air passing on into the bowel beyond the level of the anastomosis (Fig. 9). This occurred on the third day; glucose was then started by mouth and the child gradually given increasing feedings. He was discharged on April 26, 34 days after operation, taking a full diet and weighing 2620 Gm. as compared to his admission weight of 1955 Gm. He has grown well in the two and a half years since operation and is apparently normal in all respects.

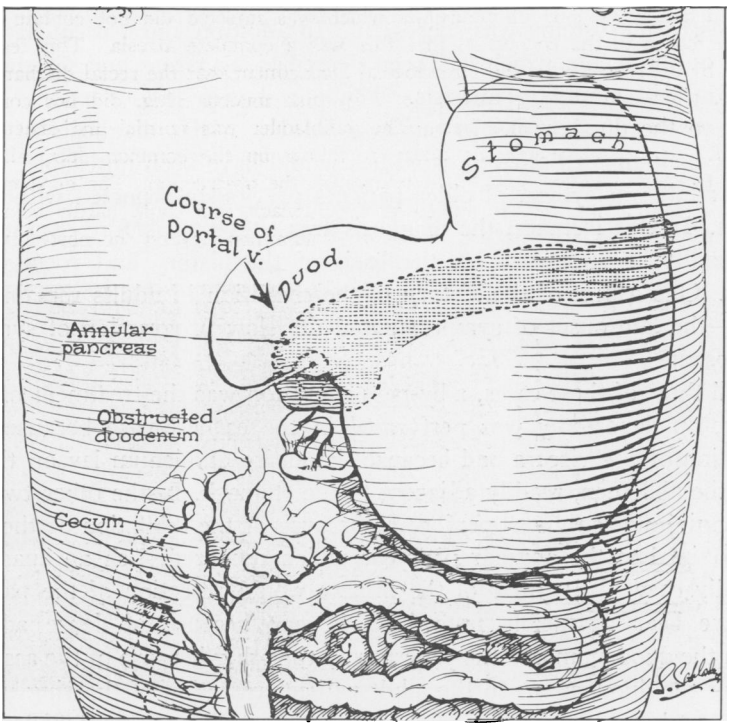


FIG. 10.—(Case 3) R. L., white male, age eight days, No. A-53869. Operative findings: The stomach and first portion of the duodenum are tremendously dilated. There is an annular pancreas surrounding the duodenum at a level coinciding with a complete intrinsic duodenal obstruction. (The portal vein passed anterior to the duodenum just proximal to the annular pancreas, but the artist has omitted this for simplicity.) The duodenum does not pass over to the left behind the superior mesenteric artery and is repeatedly kinked on itself by adhesions. There is a malrotation of the colon. After the peritoneal bands which held it in this position were released the colon was found to swing freely from a long mesentery attached only at its root.

Comments. This was an eight-day-old infant in whom deep jaundice complicated the picture of complete duodenal obstruction. Jaundice, presumably due to back pressure, is not rare in such situations. An annular pancreas was associated with malrotation of the intestine, complete duodenal atresia, and passage of the portal vein anterior to the duodenum. Gastro-duodenostomy relieved the obstruction, and division of mesenteric and peritoneal bands corrected the malrotation.

DISCUSSION

These three cases provide fairly characteristic instances of annular pancreas. The two infants were typical of patients with neonatal duodenal obstruction, whatever the cause. One infant presented a combination of malformations—namely, an annular pancreas and atresia of the duodenum—previously described by Vidal[27] in 1905 in an operation on a newborn in whom gastroenterostomy was successful. In addition, our patient had a malrotation of the colon, which has been described by Gross and Chisholm[12] in connection with annular pancreas. The portal vein in our patient crossed anterior to the duodenum. In our two infants, a flat diagnosis of annular pancreas excluding all other possibilities would have been untenable.

The adult was typical of older patients previously reported with annular pancreas. In this instance, the diagnosis was suggested before operation and could readily be supported on the basis of the history and roentgenologic findings. One aspect of the history of the condition in adults remains unexplained—the late onset of symptoms and the slow progression of symptoms, once present.

In all three of these cases, a by-pass operation was the treatment of choice. A gastro-duodenostomy was performed in the earliest case because of the malrotation of the viscera and because the entire duodenum lay on the right side of the abdomen, readily adjacent to the stomach. In the other two cases, a duodenojejunostomy was performed because of the dilatation of the duodenum proximal to the annular pancreas which rendered such an anastomosis technically easy. In neither of the infants would resection of the pancreatic ring have been a feasible procedure, in one because of dense adherence between the duodenum and the pancreas, in the other because of the associated-atresia of the duodenum. In the adult patient, resection of the ring was performed first but without relief of the obstruction. The presence of scar tissue exactly at the site of the previously resected ring gives credence to the suggestion that leakage of pancreatic secretions stimulated the dense scar formation. Prompt and immediate relief from obstruction was obtained in all three patients with the formation of short circuiting anastomoses.

SUMMARY

Three cases of annular pancreas are reported, two infants and an adult. Operation was performed in each instance with success. Division of the pancreatic annulus introduces the hazards of pancreatic fistula, or of pancreatitis. Duodenojejunostomy is probably the procedure of choice.

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