Annular Pancreas *

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Annular pancreas was first recognized by Ecker in 1862 in autopsy material and it was he who gave the anomaly its name. In France, 1905, Vidal,68 recognizing the annular pancreas, performed the first operation for the condition, a gastrojejunostomy with success on a three-day-old girl. From that date through 1949, 26 additional cases operated upon were presented. From 1950 through 1955, 67 further cases, including the three cases in this report, have been recorded, bringing the total cases operated upon to 93. No attempt has been made to record and tabulate cases reported in 1956. It appears, therefore, that with increasing awareness of the condition and with increasing availability of specialized roentgenologic examinations, the purported rarity of the condition is open to question. An increased index of suspicion will continue to uncover new cases.

The purpose of this paper is to report three additional cases, and to analyze our own and previous reports, especially in regard to incidence of complications occurring in the natural history of the disease; incidence of associated anomalies in adults as well as children; and correlation of the type of operative procedure with morbidity and mortality rates. The entire literature on the subject has been extensively reviewed. It is not our purpose to list all the reported cases, but rather to extract pertinent information from the series of patients operated upon.

EMBRYOLOGY AND PATHOLOGY

Two main theories have been advanced to explain the origin of an annular pancreas. The most widely accepted is that advanced by Lecco 33 and supported by studies of the duct system in annular pancreas presented by Cunningham,16 McNaught,41 and McNaught and Cox.42 This consists of a failure of the ventral anlage of the pancreas to rotate as the duodenum rotates. The ventral anlage normally makes up the head and uncinate process of the pancreas, and its duct becomes the central portion of the duct of Wirsung. The duct of an annular pancreas, in most cases, originates anteriorly and to the left of the duodenum. It then courses to the right anterior to the duodenum, thence posteriorly around the duodenum to enter the main pancreatic duct at the posterior aspect of the latter. This course of the duct within the annulus suggests that annular pancreas arises when the ventral anlage of the pancreas becomes fixed anteriorly, and the duodenum, in rotating, leaves a ring of pancreatic tissue about the duodenum.

The second theory, advanced by Tiecken, ⁶³ is the hypertrophy theory. Pancreatic tissue is believed to hypertrophy from the head of the pancreas both anteriorly and posteriorly. Evidence for this theory has been gathered from specimens of incomplete annulus where the defect is situated at the lateral aspect of the duodenum at the most distant point from the pancreas, with tongues of pancreas anterior and posterior to the duodenum. This theory does not take into account the embryologic development of the pancreas.

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TABLE 1. Congenital Anomalies Associated with Annular Pancreas (Operated Cases)

Type of Anomaly	Children (Total Cases 37; Cases with Anomalies 26) ^a	Adults (Total Cases 56)		
Duodenal atresia or stenosis	16	7		
Mal-rotation of intestine	6	0		
Esophageal atresia, with or without				
tracheo-esophageal fistula	3	0		
Meckel's diverticulum	3	0		
Congenital heart disease	3	0		
Mongolism	3	0		
Miscellaneous				
Intramesenteric hernia	0	1		
Ectopic kidney	1	0		
Imperforate anus	1	0		
Accessory spleen	1	0		

a Several children had more than one anomaly.

The pancreatic tissue in an annular pancreas is made up of normal acinar and islet tissue, and, as such, is subject to the same pathologic changes as the normal pancreas. To date, however, a neoplasm of the annulus has not been reported.

Of particular interest to us has been the high incidence of duodenal stenosis and atresia in children (43%) and duodenal stenosis in adults (12 to 19% and perhaps even greater). It seems to us more than mere coincidence that, in the six to seven week embryo, the duodenum with the ventral anlage undergoes rotation, while at the same time the lumen of the gut is evolving from its solid state. Since atresia or stenosis is believed to be an incomplete evolution from the solid state, a temporary arrest in development at this stage might explain the frequent association of the two defects. It may be argued that this stenotic band represents an inflammatory stricture, or the annulus itself in an intrinsic position. In two of our cases, however, it appeared that the annulus was separate from the intrinsic stenosis. Several authors have reported resecting the ring of pancreatic tissue only to find that the underlying duodenum was still constricted by an intrinsic fibrous band. It appears to us that this represents an associated congenital duodenal stenosis. The

remainder of the anomalies encountered are presented in Table 1 and are seen, for the most part, to be associated with faulty development of the gastro-intestinal tract. In children 70 per cent of the cases have other anomalies, while associated anomalies occur in 14 per cent of adults.

COMPLICATIONS

In annular pancreas, there are four major complications which may occur. These are the manifestations of secondary changes in the gastroduodenal tract, in the pancreas, and in the biliary tract.

The first and most obvious is duodenal obstruction, and this condition brings the patient most often to the clinician. The obstruction varies in degree, being complete or almost complete in infants, and being more chronic and less marked in adults. It may be, as suggested by Beck.5 that the dilated duodenum and stomach become ptotic as time passes, and that this ptosis gradually brings on symptoms of obstruction late in life by causing angulation at that portion of the duodenum relatively fixed by the annular pancreas. The role of the intrinsic duodenal stenosis in contributing to obstruction has been commented upon.

^b Four additional cases had persistent narrowing following resection of the ring (? intrinsic stenosis).

Secondly, the prolonged stasis of ingested material in the duodenal bulb and antrum of the stomach predisposes this group of patients to peptic ulceration of the duodenum or stomach. This complication arose in 18 of 56 reported cases (32%). Occasionally, it is this factor which causes the patient to seek medical advice.

Thirdly, secondary changes in the annulus itself with development of pancreatitis, either acute or chronic, may occur. This process is usually a diffuse one involving the entire gland. It causes acute symptoms by superimposing acute duodenal obstruction upon a partially obstructed and relatively asymptomatic duodenum. The cause of pancreatitis, in nine of 56 adult patients with annular pancreas (16%), is not apparent except that the constant trauma of the partially obstructed duodenal wall may cause enough inflammatory edema as to occlude partially the pancreatic duct orifice or the papilla itself. This has been suggested as a cause for this complication.

Fourthly, secondary involvement of the biliary tract with obstruction and jaundice has been reported on three occasions.^{1, 2, 67} This is indeed rare and is secondary to pancreatitis in the cases reported.

CLINICAL PICTURE AND DIAGNOSIS

Prior to embarking upon a description of the clinical symptoms and signs of annular pancreas, it must be pointed out that many persons with annular pancreas may live their life span without ever having symptoms. Incidental annular pancreas has been noted in autopsy material on many occasions.

There are two distinct clinical types: (1) Acute and high-grade duodenal obstruction, and (2) chronic and low grade duodenal obstruction.

Concerning the former, this is the common type in infants and is early manifest as a severe disease of emergency proportions. The disease is characterized by feeding problems since birth, vomiting, dehydration, alkalosis, and weight loss. Vomitus may or may not contain bile. Rarely, jaundice has been present. Abdominal distention is not marked because of the frequent vomiting with spontaneous decompression of the stomach. These children are desperately ill and require correction of fluid and electrolyte imbalances prior to operation. X-ray findings will depend upon the degree of obstruction, but usually the stomach and duodenum are found greatly dilated. The "double-bubble" 32 sign may be helpful. This is caused by the dilated first portion of duodenum. Diagnosis in this group of patients will include annular pancreas among the possibilities, but preoperative diagnosis will be no more than an "educated guess" as pointed out by Kiesewetter.32 The necessity for operation in these patients is obvious.

Occasionally, acute pancreatitis complicating a relatively asymptomatic annular pancreas may precipitate acute duodenal obstruction in adults. The occurrence of upper abdominal pain, with evidence of peritoneal irritation, elevated serum amylase, and acute duodenal obstruction, suggests the diagnosis.

In general, however, the disease in adults follows the pattern of chronic duodenal obstruction. Here the disease is characterized by a relatively long period of lowgrade obstructive symptoms, such as postprandial fullness, belching, and intermittent vomiting. Adequate nutrition may be maintained for long periods, but usually some degree of weight loss is noted. In short, the picture at this stage is a chronic one but is slowly progressive. At any time, however, the development of a peptic ulcer, or, as noted above, pancreatitis, may suddenly transform the chronic picture into an acute clinical situation. Bleeding from ulcer has been described in many of the reported cases as the presenting symptom.

Common bile duct obstruction has been reported recently by Ayala Gonzalez *et al.*² secondary to repeated bouts of pancreatitis



Fig. 1. Case 1. GI series revealing markedly dilated duodenal bulb, so redundant that the site of obstruction at the descending limb of the duodenum is obscured.

in the annulus. This is a rare sequence of events.

The diagnosis in the adult form of the disease is dependent exclusively upon the findings in the gastro-intestinal x-ray series. Here the typical findings consist of a dilated first portion of duodenum and a narrowed second portion. The length of the narrowed segment varies and there is no distortion of mucosal markings. A smooth dextral filling-defect, described by Lehman,³⁴ is found sometimes. Warren ⁷⁰ has pointed out that the first portion of the duodenum may not be dilated if it is the site of cicatrization secondary to ulceration in the bulb.

In illustrating the problem of differential diagnosis, we have recently had a patient in whom we diagnosed annular pancreas preoperatively because of a dilated duodenal bulb and narrow descending duodenum. At operation he had a cholecystoduodeno-colic membrane with partial ob-



Fig. 2. Case 1. At operation, dilated duodenum is suspended by sutures. At the left central portion of the picture a forceps points to the narrow annular pancreas.

struction of the second portion of the duodenum. In addition to this entity malrotation of the gut with congenital duodenal bands, neoplasms, both primary in the duodenum, or secondary from tumors of the pancreas, kidney, or retroperitoneal soft tissues, cicatrizing duodenitis, or post-bulbar ulcer with stricture, and pericholecystic inflammatory adhesions must all be considered.

The following are reports of three cases seen and treated by the authors in a 17-month period from June, 1954 to November, 1955.

CASE REPORTS

Case 1. A 66-year-old white Austrian man was admitted to Strong Memorial Hospital, Rochester, N. Y., on June 10, 1954, following the sudden passage of "about a quart" (1 liter) of old blood per rectum with associated syncope. For the past 4 years he had had intermittent episodes of heartburn, dyspepsia, post-prandial epigastric fullness,



Fig. 3. Case 2. GI series showing dilated duodenal bulb. It was possible in oblique spot views of the duodenum to demonstrate the narrowed second portion of the duodenum. This narrowing is not seen in this view.



Fig. 4. Case 2. Delayed film during GI series showing retention of barium in stomach and dilated duodenum.

and a gradual weight loss of 4.5 Kg. (10 pounds). Four years ago there had been a bout of melena which cleared spontaneously without medical attention. Constipation had been present for many years. He had had hypertension and had had a cerebrovascular accident 18 months prior to admission.

On admission his condition was satisfactory although there was evidence of recent blood loss. The physical examination was normal except for tenderness without a mass in the epigastrium, and a right hemiplegia.

Laboratory work-up revealed a normochromic anemia with hemoglobin of 11.5 grams per cent. Remainder of blood count was normal. Stools were positive for blood. Gastric analysis revealed 46 units of free hydrochloric acid and guaiac positive contents.

Gastro-intestinal x-ray series on June 11, and 16, 1954, revealed a markedly dilated and atonic first portion of the duodenum (Fig. 1) with partial obstruction in the descending limb of the duodenum. No ulcer was seen and the point of obstruction was not visualized because of the overhanging distended duodenal bulb. With a pre-

operative diagnosis of either annular pancreas, duodenal tumor, or pericholecystic adhesions, operation was performed on June 21, 1954. A narrow annular pancreas, 1.0 to 1.5 cm. in width, completely surrounding the descending limb of the duodenum, was found (Fig. 2); and, in addition, an ulcer of the anterosuperior aspect of the greatly distended duodenal bulb. Beneath the annulus there was an intramural stenosis which narrowed the lumen to 1.5 cm. diameter. The artery and vein of the annulus were branches of the middle colic vessels. Subtotal gastrectomy with retrocolic gastrojejunostomy was performed. It was felt that the duodenal stump could be closed without hazard proximal to the ring without transecting the ring. His postoperative course was complicated by transient atelectasis and auricular fibrillation which responded to specific therapy, and he was discharged on his eleventh postoperative day.

Fourteen months later the patient is doing well and has regained all his lost weight.

Comments: In this case, the occurrence of upper gastro-intestinal bleeding brought the patient to the hospital. Although a history of low-grade duodenal obstruction could be elicited, this was

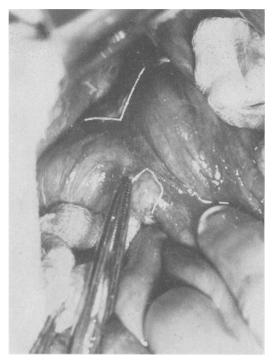


Fig. 5. Case 2. At operation, the pseudo-annular pancreas is seen. The tongues of pancreatic tissue are outlined in white. The forceps points to the gap between them, where there is constriction by fibrous bands, containing small circumferential blood vessels. This gap is present at the lateral aspect of the duodenum.



Fig. 6. Case 2. One month postoperatively, GI series reveals diminution in size of the duodenal bulb, with a well functioning duodenojejunostomy stoma present at the underside of the duodenum.

not of sufficient significance to the patient to warrant his seeking medical advice. Treatment with gastric resection and gastrojejunostomy because of the complicating bleeding duodenal ulcer brought relief from the symptoms of duodenal obstruction. The annulus was not resected, and the duodenal stump was closed proximal to the annulus without subsequent difficulty.

Case 2. A 22-year-old white male sergeant in the U. S. Air Force was admitted to Valley Forge Army Hospital, Phoenixville, Pennsylvania, from an Air Force Base Hospital with the diagnosis of annular pancreas on February 25, 1955.

For 3½ years he had been having intermittentt bouts of epigastric pain, post-prandial fullness, relieved occasionally by forcing himself to vomit, and a weight loss of 10.5 Kg. (23 pounds). Prior to the present admission he had had three previous hospital admissions for these symptoms with temporary and partial relief with bland diet and antispasmodics.

Physical examination was normal except for a "gurgling" sensation on deep palpation in the epigastrium. There was no mass or tenderness on abdominal examination.

Gastro-intestinal x-ray series revealed a grossly dilated duodenal bulb (Fig. 3) with evidence of gastric retention at 3 hours (Fig. 4). The second portion of the duodenum was narrowed for a length of 5 cm. by an extrinsic mass, believed to be an annular pancreas.

On March 18, 1955, laparotomy revealed a semi-annular pancreas with a laterally-located gap about 1.5 cm. in length (Fig. 5). In the space between the unjoined tongues of pancreatic tissue there was a continuation of the circumferential blood vessels supplying the annulus. Underlying the annulus there was a narrow duodenal stenosis limiting the lumen to 1.5 cm. It was this latter condition which caused the obstruction. The first portion of the duodenum was greatly dilated. A retrocolic duodenojejunostomy was performed. The postoperative course was uneventful. One month postoperatively, gastro-intestinal x-rays revealed some diminution in size of the duodenal bulb with well-function stoma (Fig. 6). He had regained 4.5

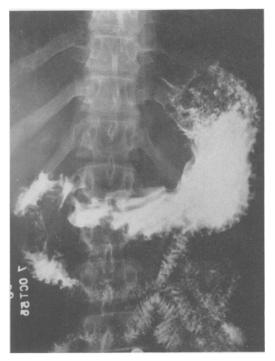


Fig. 7. Case 3. GI series showing a smooth extrinsic filling defect at the right lateral aspect of the descending limb of the duodenum. There is no evidence of obstruction. First portion of the duodenum appears normal. A benign duodenal tumor or an annular pancreas were considered the most likely possibilities in this case.

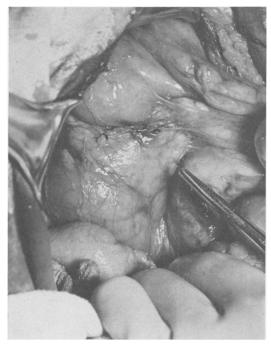


Fig. 8. Case 3. At operation, the broad annulus of pancreas can be seen. Forceps points to the annulus and two sutures can be seen on the surface of the annulus. A duodenojejunostomy was performed.

Kg. (10 pounds) at that time and 14 months postoperatively is doing full duty in the U. S. Air Force overseas, is asymptomatic, and has gained a total of 9 Kg. (20 pounds).

Comments: This case represents the problem of uncomplicated low-grade but increasing duodenal obstruction due to annular pancreas. It also represents a case of incomplete annulus with intrinsic duodenal stenosis. It is interesting that although pancreatic tissue was present only anteriorly and posteriorly, the blood vessels supplying the annulus continued around the entire circumference of the duodenum, completing the ring. Good result here was obtained with side-to-side isoperistaltic duodenojejunostomy.

Case 3. A 38-year-old unmarried Negro sergeant in the U. S. Army was admitted to Valley Forge Army Hospital September 28, 1955, for treatment of active, moderately advanced, bilateral upper lobe tuberculosis. He had been admitted to another Army Hospital on September 4, 1955, because of several episodes of hiccoughs over the 2 previous years, which appeared to have been as-

sociated with a full stomach relieved by induced vomiting. Tuberculosis had been discovered during this hospitalization and he had been transferred to Valley Forge Army Hospital. These episodes of hiccoughs sometimes lasted 2 to 3 days, and 3 hospitalizations had been necessary. Post-prandial fullness and vomiting had not been prominent although excessive belching had been present with eating, and frequently he felt as if he might vomit. Weight loss of 7.8 Kg. (17 pounds) had occurred prior to admission, but after anti-tuberculosis therapy of 6 weeks' duration, 4.5 Kg. (10 pounds) had been regained prior to operation.

Physical examination had been within normal limits except for a small epigastric hernia.

A gastro-intestinal x-ray series revealed a dextral filling-defect in the second portion of the duodenum which appeared to be due to an intramural tumor or an extrinsic mass (Fig. 7). There was no evidence of obstruction and the first portion of the duodenum showed no dilatation. Although annular pancreas was considered as a possibility, a benign tumor of the duodenum was felt to be the most

TABLE 2. Complication and Mortality as Related to Type of Operative Procedure

	Children					Adults					
		Complications ^a	Per Cent	Mor- tality	Per Cent		Compli- cations ^a		Mor- tality	Per Cent	
Resection or division of											
annulus	1	0	0	0	0	12	8	67	0	0	
Resection or division of annulus followed by											
by-pass procedureb	2	1	50	1	50	2	1	50	0	0	
Division of annulus plus									_	-	
duodenoplasty	1	1	100	1	100	7	2	29	1	14	
Duodeno-jejunostomy	24	9°	38	9	38	6	1	16	0	0	
Gastro-enterostomy	4	2	50	2	50	10	3	30	3	30	
Duodeno-duodenostomy	2	1	50	1	50	1	0	0	0	0	
Gastro-duodenostomy	2	0	0	0	0	0	0	0	0	0	
Gastric resection without											
division of annulus	1 ^d	0	0	0	0	11	1	9	0	0	
Gastric resection with											
division of annulus	0	0	0	0	0	4	2	50	0	0	
Miscellaneous (vagotomy + gastro-jejunostomy, pyloro-											
plasty, and pyloropexy)	0	0	0	0	0	3	0	0	0	0	

a Includes deaths.

likely diagnosis. The absence of evidence of obstruction in the roentgenograms was felt to favor the latter. On November 30, 1955, exploratory laparotomy revealed a complete annular pancreas, of uniform width of 2.0 cm. (Fig. 8). There was no intramural duodenal stenosis. The first portion of the duodenum and the stomach were not dilated, but the wall of the duodenum was moderately hypertrophic. A retrocolic isoperistaltic sideto-side duodenojejunostomy was performed. It was felt that clinically there had been minimal intermittent obstruction, and that such an operative procedure would prevent further or progressive obstructive symptomatology. There was no peptic ulcer and there were no other anomalies. The epigastric hernia was also repaired.

Postoperatively there were no complications and he was transferred to the Tuberculosis Service on the twelfth postoperative day.

Seven months following operation, he states there has been no recurrence of hiccoughs nor post-prandial distress whatsoever. He has gained an additional 6.8 Kg. (15 pounds) since operation.

Comments: This is a case of complete annular pancreas without duodenal stenosis in a patient who had never had any complications of his anomaly except vague and minimal obstructive symptoms. Postoperatively, however, he has stated

that many of his preoperative symptoms, to which he had become accustomed and considered almost normal, are now absent. Exploration in this case was prompted, however, by the possibility of a duodenal tumor more than by the clinical picture of partial duodenal obstruction. It should be mentioned parenthetically that the smooth dextral filling defect noted in this case has been described as typical of annular pancreas.

TREATMENT

Eleven operative procedures have been recommended in the surgical treatment of annular pancreas. Results following each are depicted in Table 2.

Of the numerous procedures advised, three main categories are noted: (1) Resection or division of the pancreatic annulus; (2) bypass procedures; and (3) gastric resection.

At the present time, the frequent occurrence of postoperative pancreatitis and of pancreatic fistula after procedures upon the pancreatic annulus leads us to decry its use. In addition, if an associated duodenal steno-

b This group represents resection of annulus failures, but were not included in statistics of that category.

^c One of these had simultaneous resection of ring.

d Duodenojejunostomy failure; not included in duodeno-jejunostomy cases.

sis is also present, obstruction may not be relieved by division of the ring alone. Table 2 graphically demonstrates the high incidence of complications with resection or division of the annulus with 11 of 21 adult cases having postoperative difficulties. The complications consisted of pancreatitis, pancreatic fistula, and failure to relieve the obstruction. Only one death, however, is recorded. In children, two of four patients died.

The simplest procedure which will relieve obstructive symptoms, and which is attended by the fewest number of complications, is the side-to-side isoperistaltic duodenojejunostomy performed either anterior or posterior to the colon. Table 2 reveals 0 per cent mortality in adults and 36 per cent mortality in children. In the latter group, the operative risk was necessarily greater due to prematurity and other anomalies. Gastrojejunostomy, although performed most frequently early in the collected series. has the obvious disadvantages of not adequately decompressing the dilated duodenal bulb and of producing a situation conducive to the development of marginal ulcer. The high mortality for this procedure, noted in Table 2, occurred early in the series and was due to three cases of postoperative bronchopneumonia. Duodenostomy would seem to be theoretically the ideal bypass procedure, but only three cases are available for study. Since, however, from the practical point of view, this procedure offers no more advantages than the technically more simple duodenojejunostomy, we continue to favor the latter.

Gastrectomy has been performed, for the most part, when associated peptic ulcer has been present. Gastric resection with gastrojejunostomy was studied in the collected cases in order to discover any increased morbidity in closing the duodenal stump proximal to a constricting annulus where the annulus was not disturbed. The series is admittedly small (11 cases), but there were no instances of duodenal stump leak-

age. Pancreatic fistulae developed in two of four cases, in which the ring was divided to protect the duodenal stump from back pressure. These two cases 48, 70 were successfully treated for these complications. The duodenal stump has been protected by Monsaingeon 43 and by Baker 3 by bringing up a limb of efferent jejunum and performing end-to-side duodeniejunostomy proximal to the annulus. This apparently worked well in difficult situations. At the present time, from the small series available and from personal experience with one case, we feel that the annulus should not be divided when gastroectomy is performed. The duodenal stump should be closed routinely. Usually this is done easily because of the dilated and thickened duodenal bulb. If the constriction beneath the annulus is marked and back pressure upon the healing duodenal stump is feared, duodenojejunostomy, anastomosing the end of the duodenum to the side of the efferent loop of jejunum, may be used.

The surgical treatment of annular pancreas with recurrent pancreatitis is the least standardized of the operative procedures, just as is chronic relapsing pancreatitis without annular pancreas. Duodenojejunostomy has been performed under these circumstances in order to correct abnormal mechanics in the duodenal wall with the hope that recurrent pancreatitis will be prevented, and this should be done as a first step. In view of the fact that the annular duct empties through the major papilla in the majority of cases, sphincterotomy might be considered. There is no reported case in which this has been done. If pancreatitis is limited to the annulus, complete resection of the involved annulus might be considered, but we feel that this represents the only possible indication in which resection is justified.

If jaundice is present, decompression of the common bile duct with T-tube, plus duodenojejunostomy to correct the duodenal obstruction, is recommended. There has not been, however, sufficient experience with the complication of pancreatitis and obstructive jaundice to allow any standardization of procedures in these cases. A good initial result was obtained by Ayala-Gonzalez with resection of the annulus, and the use of a long arm T-tube through the ampulla. No long-term result is available in this very interesting case.

On the grounds that much of the obstructive symptoms late in life may be due to ptosis, Beck ⁵ has recently advocated pyloropexy or duodeno-pexy in selected cases. He has performed this operation on one such case with good initial result.

SUMMARY

- 1. A brief review of the theories of development of annular pancreas, its complications, and the clinical picture has been presented.
- 2. The literature concerned with the cases operated upon through 1955 has been reviewed, and the experience in these 93 cases has been analyzed as regards incidence of other anomalies, incidence of associated peptic ulcer, or pancreatitis, and results with various methods of treatment. An attempt has been made to standardize the surgical therapy of this condition. Duodenojejunostomy is recommended as the treatment of choice when duodenal obstruction only is present. Gastrectomy without division of the annulus with gastrojejunostomy is recommended in those cases having associated peptic ulcer. In cases where the annulus is the isolated site of pancreatitis, resection of the ring is suggested. It is emphasized that in cases where the entire pancreas, including the annulus, is the site of chronic relapsing pancreatitis the treatment is not standardized. This latter situation represents the biggest problem in the surgical therapy of annular pancreas.
- 3. Attention is again drawn to the frequent co-existence of duodenal atresia or stenosis and annular pancreas, and a pos-

sible explanation of this co-existence is offered.

4. Three personal cases are presented.

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