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Diagnosis and Treatment of Jejunoileal Atresia

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A total of 116 cases of intestinal atresia or stenosis were encountered at the Yale-New Haven Hospital between 1970 and 1990. Sites involved were the duodenum (n = 61; 53%), jejunum or ileum (n = 47; 46%), and colon (n = 8; 7%). All but two patients underwent operative correction, for an overall survival rate of 92%. Challenging problems were the management of apple-peel atresia (five patients), multiple intestinal atresia with short-gut syndrome (eight patients), and proximal jejunal atresia with megaduodenum requiring imbrication duodenoplasty (four patients). Major assets in the improved outlook for intestinal atresia are prenatal diagnosis, regionalization of neonatal care, improved recognition of associated conditions, innovative surgical methods, and uncomplicated long-term total parenteral nutrition.

Atresia is the most common cause of congenital intestinal obstruction and accounts for about one-third of all cases of intestinal obstruction in newborns. The sex distribution is almost equal. Ravitch and Barton [1] in a nationwide survey of neonatal surgery, estimated the overall incidence of intestinal atresia to be 1 per 2710 live births, making this anomaly about twice as common as esophageal atresia or congenital diaphragmatic hernia and almost three times more common than Hirschsprung's disease.

Despite the relative frequency of this anomaly, the first survivor of an anastomosis for small intestinal atresia was not reported until 1911 by Fockens [2] in Holland. Evans [3] reviewed 1498 cases reported up to 1950 and found less than a 10% survival rate. The most extensive reviews on the subject were authored by Fonkalsrud et al. [4] and de Lorimier et al. [5] and are based on the collective experience of the Surgical Section of the American Academy of Pediatrics. More recently, the survival rate has risen rapidly to nearly 90% with the introduction of modern surgical techniques and the use of total parenteral nutrition [6–8]. The 92% survival rate of 118 operated cases of intestinal atresia and stenosis treated in the Newborn Special Care Unit of the Yale-New Haven Hospital between 1970 and 1990 reflects the importance of neonatal intensive care on the outcome of treating this and other major congenital anomalies.

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Classification

Duodenum

Sixty-one patients with duodenal atresia or stenosis were encountered, including 12 with preampullary duodenal obstruction based on the absence of bile in the gastric contents. A diaphragm causing partial obstruction or duodenal stenosis was found in 14 patients. An unusual cause of obstruction is complete absence of a duodenal segment accompanied by a mesenteric defect—seen in five patients. Detecting a "windsock" web is critical because there is a tendency to confuse it with distal duodenal obstruction and the frequent occurrence of an anomalous biliary duct entering along its medial margin [9, 10]. Although described, this entity was not recognized in this series. Annular pancreas represents an associated embryologic defect found with intrinsic duodenal obstruction, but it is not the primary cause of obstruction. A strong possibility exists that the true incidence of annular pancreas and biliary tract anomalies may be considerably higher than suspected, as surgeons now more carefully examine the site of obstruction prior to performing corrective surgery.

Jejunum and Ileum

Cases seen in the jejunum and ileum are equally distributed from beyond the ligament of Treitz to the ileocecal junction (Table 1). The classification used here is similar to those proposed by Louw and Barnard [11], Hays [12], and Martin and Zerella [13] but has been modified [6, 7] to include the "apple peel" lesion as a type 3b atresia and multiple atresia as a fourth type of atresia (Fig. 1).

Type 1. An intraluminal diaphragm in continuity with the muscular coats of the proximal and distal segments: 15 of 47 patients (32%).

Type 2. Atresia with a cord-like segment between the blind ends of bowel: 12 of 47 patients (26%).

Type 3a. Atresia with complete separation of the blind ends accompanied by a V-shaped mesenteric defect: 7 of 47 patients (15%).

Type 3b. Atresia with an extensive mesenteric defect with the distal ileum receiving its blood supply entirely retrograde

Table 1. Classification of anatomic types of jejunoileal atresia, Yale-New Haven Hospital, 1970–1990 (n = 47).

Anatomic site	Stenosis (no.)	Atresia, by type (no.)				
		1	2	3	4	Total
Jejunum	2	9	7	5	5	26
Ileum	1	6	5	7	3	21
Total	3	15	12	12	8	47

from a single ileocolic artery: 5 of 47 patients (11%). The distal intestine coils itself around this vessel, giving the appearance of an "apple peel" deformity. This anomaly has special significance because of frequent accompanying extreme prematurity, an unusually small distal bowel, and significant shortening of overall bowel length [14]. A hereditary predisposition of this form of atresia has been reported [15] in two of our five cases.

Type 4. Multiple atresia of the small intestine: 8 of 47 patients (17%). Combinations of small and large bowel atresia are rare, having occurred in only two of the eight cases in our series. Multiple sites of atresia from stomach to rectum is a recently recognized hereditary syndrome. Consanguinity in the families studied suggest an autosomal recessive mode of transmission. A series of five patients with this syndrome reported by Guttman et al. [16] is of special interest because of the uniformity of intraluminal calcific radiopaque densities diagnosed in utero.

Colon

Colon atresia has many similarities to jejunoileal atresia but is much less common, accounting for only eight (6%) of our cases of intestinal atresia. In this series, the atresia was observed most commonly in the transverse colon alone, with atresia of the transverse colon in conjunction with malrotation and midgut volvulus in one patient and multiple jejunoileal atresia and atresia of the distal sigmoid colon in two others. The various anatomic types of atresia described for the jejunum and colon also occur in the colon, but multiple atresia of the colon is rare.

Associated Anomalies

One of the most important differences between duodenal and more-distal atresia is the obvious complex nature of the duodenal malformation and a high incidence of Down syndrome (trisomy 21) and multiple system anomalies. Trisomy 21 was detected in 19 of 61 patients (31%) with duodenal atresia in our series. Hirschsprung's disease was diagnosed at 10 days in two patients with Down syndrome and duodenal atresia. A gastric perforation was seen in two other newborns as a result of the duodenal atresia.

Congenital heart disease is the most important associated anomaly and is found in about 20% of all babies with duodenal atresia. Serious cardiac anomalies, such as endocardial cushion defects or atrioventricular canal, are seen in nearly 80% of babies with Down syndrome. Some of the other cases reported in the literature [5] have had less serious and more easily correctable lesions, such as a septal defect.

The VATER syndrome [17] is a nonrandom association of anomalies including *v*ertebral, *a*nal, *t*racheoesophageal fistula, and/or *e*sophageal, *r*enal, and *l*imb defects. The further association of duodenal atresia with this syndrome has been reported and was seen in five of our patients.

Meconium ileus must be considered in babies with jejunoileal atresia who have signs of a prenatal volvulus, meconium peritonitis, or a particularly small microcolon containing pellets of clear mucoid meconium. Meconium ileus (or microcolon) was observed in 7 of our 51 patients with jejunal or ileal atresia. The contents of the small intestine are more viscid than in uncomplicated atresia. Because the incidence of associated meconium ileus is approximately 20% in most series of jejunoileal atresia [18], a sweat test should be obtained during the postoperative period. Gastroschisis may also have associated jejunoileal or colon atresia due to ischemic injury at the abdominal wall defect. Four of eleven babies with colon atresia in one series had gastroschisis [19]. The obstruction is usually detected during operative correction of the abdominal wall defect.

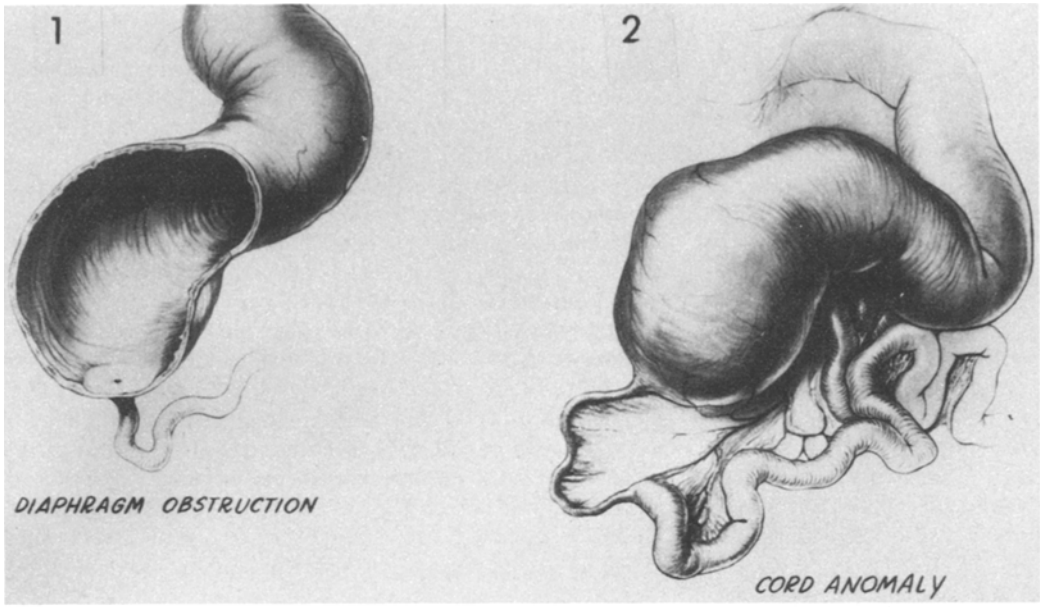
Prenatal Detection

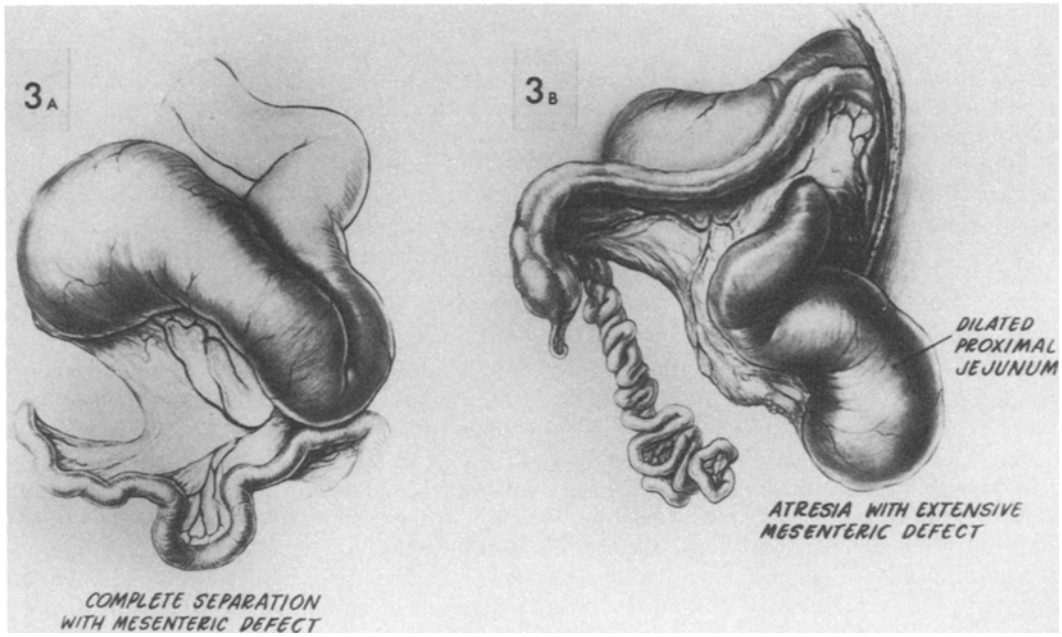
Detection of life-threatening fetal disorders by a combination of fetal screening for elevated maternal serum and amniotic fluid α-fetoprotein, structural abnormalities seen on maternal ultrasonography, and amniotic fluid and fetal blood sampling for fetal karyotype has proved increasingly important and effective, not only for decisions regarding termination of pregnancy but also for guiding the high-risk mother to obtain appropriate obstetric and neonatal care. Polyhydramnios occurs in about one-half of all newborns with duodenal and proximal jejunal atresia but in fewer babies born with ileal or colonic obstruction [20]. The finding of hydramnios is a clear indication for obtaining maternal ultrasonography, and nearly all of our patients diagnosed with duodenal atresia during the past 5 years have been diagnosed in utero. Ultrasonography as a screening procedure also provides information about cardiac and central nervous system (CNS) development, in addition to assessing the gastrointestinal tract [21]. Multiple organ malrotation syndrome (MOMS) [22]—comprising partial situs inversus, duodenal atresia, and a right-sided Bochdalek congenital diaphragmatic hernia—can be diagnosed by ultrasonography. This rare condition [22] was observed in two patients from our series.

Clinical Diagnosis

Confirming the prenatal diagnosis of intestinal obstruction is essential because not infrequently the ultrasound scan may be misinterpreted as being abnormal. For that reason, a nasogastric aspirate should be obtained after birth. Bile-colored gastric fluid or a residual volume greater than 25 ml is highly suspect for intestinal obstruction. Babies with duodenal and proximal jejunal atresia begin vomiting bilious fluid shortly after birth, whereas those with more distal atresia may not vomit until several hours or a day or two later. Babies with intestinal stenosis may present at a few weeks of age or be even older, with a history of poor feeding, intermittent vomiting and aspiration, and failure to thrive.

Abdominal distension is obvious in about 80% of babies with obstruction distal to the jejunum, and active peristalsis of the





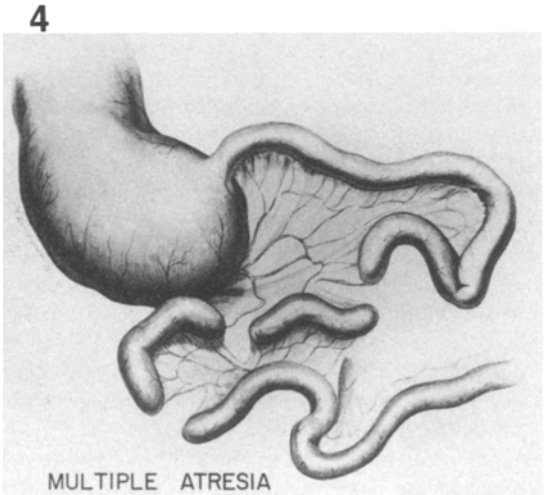


Fig. 1. Four major types of jejunoileal atresia described in the text. The distal ileum in type 3B, known as the "apple-peel" deformity, derives its entire blood supply retrograde from the ileocolic artery. (Types 1, 2, 3A, 3B: Reprinted with permission of the publisher [12].)

distended obstructed loops may be visible. Severe distension may be associated with respiratory distress due to elevation of the diaphragm. It is important to emphasize that the baby with a scaphoid or normal-appearing abdomen who vomits bile after birth must be considered to have a duodenal or proximal jejunal obstruction until further evaluation proves it not to be true.

Failure to pass meconium suggests intestinal obstruction, but approximately 30% of patients with duodenal atresia and about 20% of those with jejunoileal atresia evacuate a normal meconium stool shortly after birth. The implications of these findings are obvious. Jaundice occurs in about 40% of babies with proximal atresia and in about 20% of those with more distal obstruction. Jaundice is characteristically associated with an elevation of indirect bilirubin caused by the presence of β-glucuronidase in the neonate's intestinal mucosa. This enzyme unbinds conjugated bilirubin and enhances the enterohepatic recirculation of bilirubin in babies with intestinal obstruction.

Supine, upright, and lateral radiographic views of the abdomen are essential to determine the site of obstruction (Fig. 2). The double-bubble sign, formed by the air-filled stomach and dilated proximal duodenal bulb, is diagnostic of duodenal obstruction but not necessarily atresia. Infants with duodenal stenosis may have a relative paucity of gas distal to the duodenum without characteristic signs of proximal obstruction. Careful injection of about 50 ml of air through the nasogastric tube and repeating the abdominal film in a prone position has been helpful for unmasking the obstruction and avoids the need for barium contrast.

The radiogram on babies with proximal jejunal atresia is diagnostic, showing only a few air-fluid levels and no gas in the lower part of the abdomen. The number of air-filled intestinal loops increases with more distal atresia, but on occasion the most dilated segment is fluid-filled or a volvulus forms and the segment is not clearly visible on the plain films. In addition, Hirschsprung's disease, meconium plug syndrome, and meconium ileus cause obstruction that mimics ileal or colonic atresia. Fortunately, a distinction can be made by contrast enema in most cases, and the differential diagnosis in a newborn with lower intestinal obstruction can be clarified.

Preoperative Preparation

All the standard principles of neonatal care apply to the preoperative preparation of a baby with intestinal atresia. Special emphasis, however, is placed on: (1) thermoregulation during transport and blood and radiologic studies; (2) nasogastric decompression with an adequate-sized tube to prevent pulmonary aspiration; (3) placement of an intravenous catheter and correction of any fluid losses or electrolyte imbalance. It is particularly important to assess and replace estimated losses from vomiting and fluid sequestered in the "third space" of the obstructed intestinal tract. Drainage from the nasogastric tube should be replaced volume for volume with either lactated Ringer's solution or normal saline. Babies who already have sustained major losses and are dehydrated may receive fluids equivalent to 1% of body weight per hour until adequate urine output and osmolality have been restored. In these babies, preoperative intravenous ampicillin 100 mg/kg/day and gentamicin 5.0 mg/kg/day are employed because the bowel lumen is entered to construct the anastomosis. Crossmatching of whole blood is routine, although intraoperative transfusion may not be required. Infants with peritonitis require additional fluid replacement.

Operative Method

Anesthesia is administered by endotracheal tube using halothane with muscle relaxants as necessary. Vital signs are monitored by doppler blood pressure recordings and electrocardiogram, as well as with a precordial or esophageal stethoscope. Monitoring oxygen saturation is essential. Hypothermia can be largely avoided by using radiant heat lamps during induction and by occlusive plastic drapes and a warming mattress. Despite these precautions, the core temperature must always be monitored with a rectal or esophageal probe and telethermometer.

A right-sided supraumbilical incision provides excellent exposure of the entire gastrointestinal tract. The diagnosis of *duodenal atresia* is confirmed by dividing the gastrocolic omentum, entering the lesser sac, and tracing the duodenum to the most distal point of obstruction. In most cases, the ascending colon must be reflected medially and the proximal and distal duodenum beyond the atresia Kocherized to facilitate doing the anastomosis. We prefer a simple, single-layer side-to-side duodenoduodenostomy, although a duodenojejunostomy is required when there is a wide gap between the ends of the duodenum. This situation occurred in 5 of 32 patients during the past 10 years, when a duodenoduodenostomy was not technically feasible. Following duodenostomy of the proximal segment, careful visual inspection and passage of a catheter is essential to rule out a bulging proximal diaphragmatic membrane (wind-sock web). A diamond-shaped duodenoduodenostomy [23] is an option to the traditional side-to-side duodenoduodenostomy that some authors find to result in earlier feeding and discharge from the hospital [24]. The transanastomotic feeding catheter, as advised by Wilkinson et al. [25], has been largely abandoned in favor of short-term peripheral parenteral nutrition until the anastomosis functions, a period of approximately 4 to 7 days.

In cases of *jejunoileal atresia*, the distal bowel must be examined for other sites of obstruction, which is best accomplished by instilling saline or mineral oil into the lumen just beyond the atresia and carefully milking meconium through the ileocecal valve. It is also important to ascertain that the colon is normal, as multiple atresias may obstruct both the large and small intestine. This situation was seen in two patients in this series.

We have been satisfied with the results of primary anastomosis in the absence of ischemia, peritonitis, or a strong suspicion of meconium ileus. Our current concepts of intestinal anastomosis developed over the past 20 years are attributed largely to the efforts of Louw [26], Hays [12], and others. Their most important contribution was recognizing that the results of end-to-end repair surpassed that of side-to-side anastomosis, which had been previously advocated. Anastomotic dysfunction has largely been avoided by resecting the proximal dilated bulbous tip, sacrificing as much as 15 to 20 cm of intestine, and performing an end-to-oblique or end-to-back anastomosis [26]. Similarly, the end-to-oblique anastomosis has largely eliminated later development of the "blind-loop syndrome," often

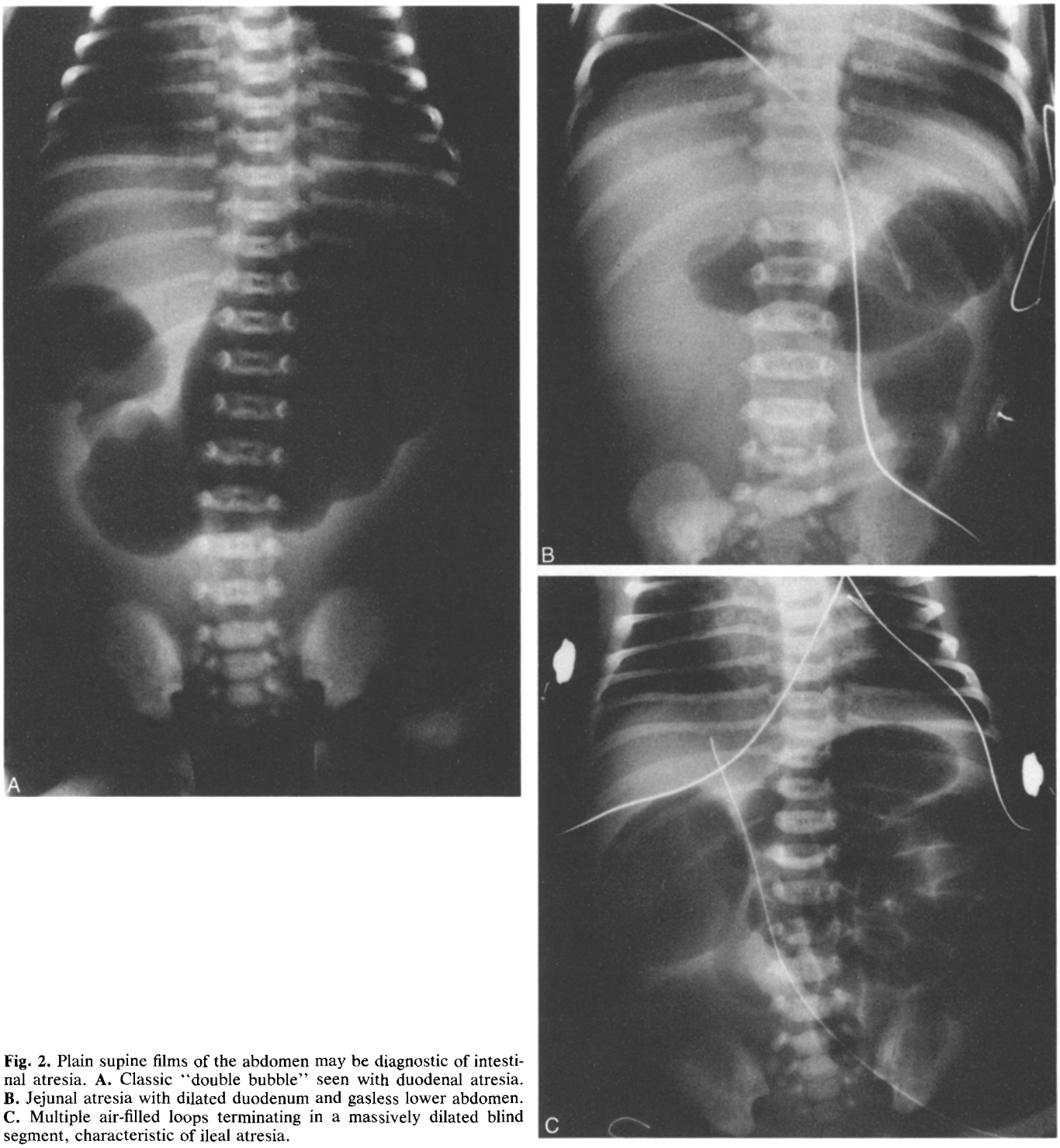


Fig. 2. Plain supine films of the abdomen may be diagnostic of intestinal atresia. **A.** Classic "double bubble" seen with duodenal atresia. **B.** Jejunal atresia with dilated duodenum and gasless lower abdomen. **C.** Multiple air-filled loops terminating in a massively dilated blind segment, characteristic of ileal atresia.

seen in conjunction with the side-to-side anastomosis. Only the tip of the distal segment should be resected, with as much of the distal intestine preserved as possible because the terminal ileum is required for fat-soluble vitamin absorption and the enterohepatic circulation of bile salts. An oblique resection at a 45-degree angle toward the antimesenteric wall with further antimesenteric extension is usually necessary to achieve an adequate diameter and make an anastomosis feasible. Our anastomosis is performed with a single layer of inverted interrupted 5-0 cardiovascular silk sutures taken through the full thickness of both the proximal and distal segments (Fig. 3). This inversion technique opposes the serosal surfaces without narrowing the lumen.

Primary anastomosis in cases of distal duodenal and proximal

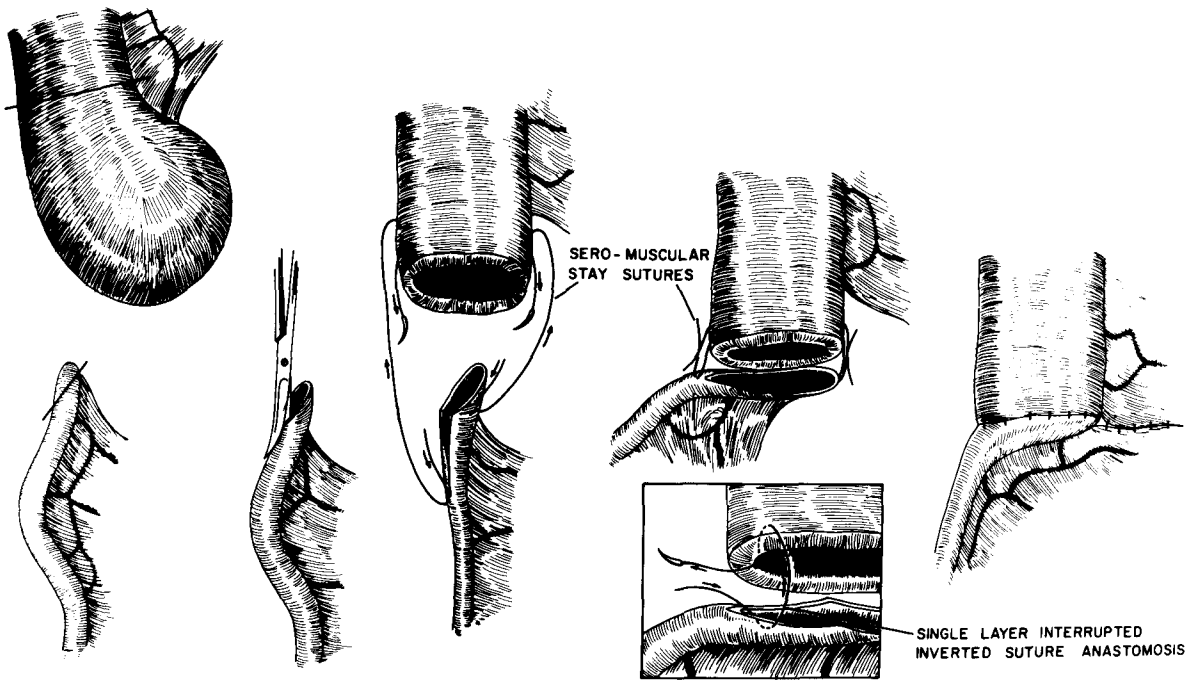


Fig. 3. Important principles of end-to-oblique anastomosis for jejunoileal atresia include (1) transverse resection of the dilated bulbous tip of the proximal segment; (2) an oblique resection of the distal segment; (3) further spatulation of the distal lumen by opening the antemesenteric wall; (4) an end-oblique anastomosis using a single layer of interrupted, inverted 5-0 silk sutures; (5) closure of the mesenteric defect.

jejunal atresia pose a specific challenge because of the marked hypertrophy and dilatation of essential intestine. Tapering the proximal jejunum and duodenum with the GIA autostapling device [6] or plicating by imbrication the intestine [27] reduces the circumference of the obstructed bowel and enhances the return of peristalsis and anastomotic function. We favor the intestinal plication procedure, which has the advantage of preserving the mucosal absorptive surface, which is important in cases of potential short-gut syndrome (Fig. 4) and has been employed effectively in the past four cases of proximal jejunal atresia.

"Apple peel" jejunal atresia [14, 15] poses a particularly challenging problem for reconstruction because of the dilated jejunum and noncontiguous small distal bowel. In these patients the proximal jejunum must be tapered and an end-to-end repair attempted [15, 28]. This approach has resulted in successful resumption of gastrointestinal function in four of five cases in our series. The other patient eventually succumbed from long-standing anastomotic dysfunction, failure of venous access, and sepsis. An alternate procedure is a side-to-end "chimney" jejunojejunostomy, as described by Santulli and Blanc [29]; it allows complete decompression of the proximal jejunum, trans-anastomotic feeding with a Silastic catheter, and subsequent closure of the enterostomy [30].

Babies with multiple sites of intestinal atresia may be predisposed to the short-gut syndrome if all the intervening intestinal segments, often described as "sausage links," are resected. To prevent this complication, multiple anastomoses are preferable to long-term home parenteral nutrition. The seemingly unavoidable kinking at the anastomoses has been prevented by stenting the bowel with a Silastic catheter, introduced via a tube gastrostomy and exiting the anus. The stent is removed after confirmation of complete transit without obstruction or leak seen with a contrast study.

Babies with *colonic atresia* have had a temporary end-colostomy with resection of the dilated proximal segment. Anastomosis of the colon is done at about 1 year of age, or earlier if the caliber of the proximal colon returns to normal. An attempt at primary anastomosis may be appropriate in carefully selected cases, but a marked discrepancy in the caliber of the proximal and distal unused colon is not unusual and makes the procedure technically difficult.

Postoperative Management

The key to postoperative care is adequate nasogastric decompression and total parenteral nutrition (TPN) in premature infants, those with malabsorption because of reduced absorptive surface, or when anastomotic "ileus" delays feeding for more than 5 to 7 days. We begin peripheral parenteral nutrition in all babies on the first postoperative day, but there is a direct correlation between the need for prolonged TPN and the type of jejunoileal atresia encountered, the severity of postoperative malabsorption, and the length of residual intestine [13]. In our experience, only 20% of patients with types 1 and 2 atresia require TPN, whereas all patients with extensive mesenteric defects, apple-peel mesentery, or multiple atresia have received TPN for 10 days to several weeks. Great patience must be exercised before deciding to reoperate for anastomotic "obstruction," as the discrepancy in the size of the lumen may cause a prolonged ileus. Stricture angulation or leak of a

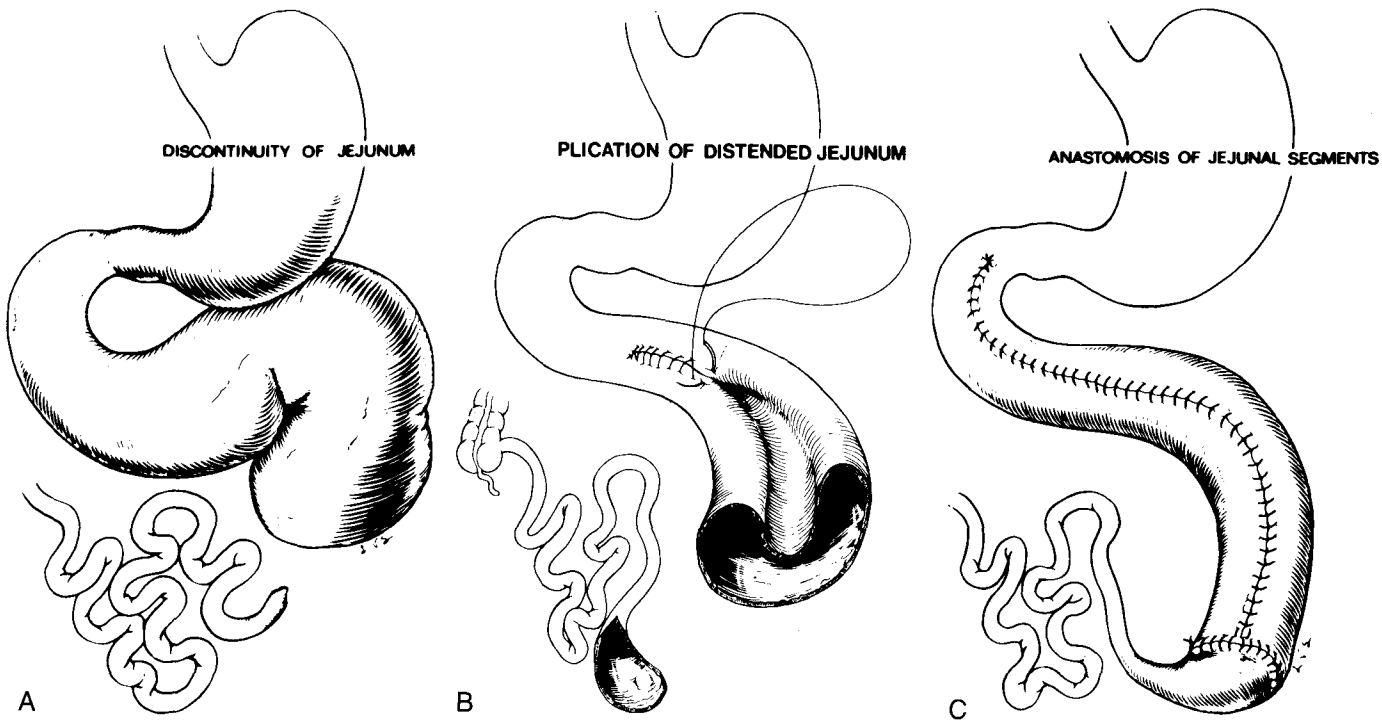


Fig. 4. A. Typical appearance of jejunal atresia with enormous distension of the jejunum and duodenum. **B.** Jejunum and duodenum have been mobilized to the right of the superior mesenteric vessels. The excessively dilated duodenum and jejunum are plicated to a more normal diameter, with a running seromuscular suture placed at 1.0-cm intervals. **C.** Following the plication, an end-to-oblique anastomosis completes the procedure. A U-shaped suture is placed between the plicated proximal jejunum and the distal bowel to prevent leakage of this site. (Reprinted with permission of the publisher [27].)

Table 2. Percent survival of patients with intestinal atresia, Yale-New Haven Hospital, 1970–1990 (n = 116).

Site of atresia	No. of patients	Operative survival
Duodenum	61	54/59[a] (91%)
Jejunum and ileum	47	43/47 (90%)
Colon	8	8/8 (100%)
Total	116	105/114[a] (92%)

[a]Two patients with severe cardiac anomalies died prior to operation.

properly constructed anastomosis is a rare occurrence; however, a contrast study is required to exclude these possibilities.

Sugar-water feedings are begun when gastric drainage diminishes and becomes clear or brownish and after several normal meconium, or "starvation," stools. We offer an elemental formula (Nutramingen or Pregestimil) or breast milk to babies with duodenal obstruction or jejunoileal atresia because of its small curd size and better absorption, and to minimize the risk of an obstruction at the anastomosis. Feedings are begun with dilute formula, and the volume and concentration are then sequentially increased over several days. The baby can be weaned onto a proprietary formula following discharge. Four patients with short-gut syndrome required home TPN for as long as 1 year; they have all been weaned from TPN and have survived. Critical to this success was scrupulous care of the catheter, cycling of the TPN to a 12- to 14-hour nighttime schedule, gradual administration of an elemental diet, and monitoring both weight and metabolic parameters.

Outlook

Common causes of death in babies with duodenal atresia are prematurity with chronic lung disease or associated major anomalies, particularly cardiac lesions. Two of our patients with severe cardiac anomalies died prior to operation. Morbidity accompanying functional intestinal obstruction due to anastomotic dysfunction that leads to severe nutritional and fluid maintenance problems should be kept to a minimum. The reported survival rate in jejunoileal atresia has risen from about 68% in 1968 [10] to 92% in this series (Table 2). The major reasons for the overall improvement in operative survival includes the following: (1) Prenatal diagnosis of almost all babies with duodenal atresia and most of those with jejunoileal atresia. In most instances the mother should be referred to a maternal high risk unit affiliated with a newborn special care unit and a pediatric surgery service. (2) The supportive care available in a regional neonatal center by specially trained personnel, including neonatologists and primary care nurses. (3) The early recognition of major associated anomalies and appropriate timing of operation in premature babies. (4) Primary repair using modern surgical techniques that minimize anastomotic complications and the blind-loop and short-gut

syndromes. (5) The use of long-term total parenteral nutrition in cases of anastomotic dysfunction or prematurity. The morbidity and mortality previously associated with chronic malnutrition has largely been eliminated. Postoperative deaths are unusual and, when reported, are usually the result of severe cardiac anomalies, such as an endocardial cushion defect with trisomy 21, ventilator-dependent chronic lung disease, or pulmonary aspiration.

Growth and development remain normal, although some patients with short-gut syndrome may exhibit "catch-up" growth after a lag phase while recovering from surgery. In these patients normal growth patterns may be achieved during the latter part of the first year of life.

Résumé

Cent vingt cas d'atrésie ou de sténoses intestinales ont été traités entre 1970 et 1990 à l'Hôpital de Yale-New Haven. Ces lésions avaient comme localisation le duodénum dans 62 cas (51%), le jéjunum ou l'iléon dans 51 (43%) et le côlon dans 8 (6%). Tous les patients sauf deux ont été opérés; la survie globale a été de 92%. Les problèmes thérapeutiques les plus ardus ont été celui du traitement de l'atrésie en "pelure de pomme" (5 patients), des atrésies multiples associées à un intestin court (8 patients) et de l'atrésie jéjunale proximale associée à un mégaduodénum, nécessitant une duodénoplastie (4 patients). Interviennent dans l'amélioration du pronostic des atrésies intestinales, le diagnostic prénatal, la reconnaisance des lésions associées, l'innovation des méthodes chirurgicales de réparation et la réalisation d'une alimentation parentérale à long terme, sans complications.

Resumen

Se registró un total de 116 casos de atresia o estenosis intestinal en el Hospital Yale-New Haven en el período 1970–1990. La ubicación incluyó el duodeno, 61 casos (51%), el yeyuno o el ileon, 51 casos (43%) y el colon, 8 casos (6%). Todos los pacientes, excepto dos, fueron soemtidos a corrección quirúriga con una tasa global de sobrevida de 92%. Los problemas principales etuvieron representados por atresia en cáscara de manzana ("apple peel, cinco casos), atresia intestinal múltiple con síndrome de intestino corto (8 casos) y atresia yeyunal proximal con megaduodeno que requirió duodenoplastia por imbricación (4 casos). Factores favorables que significan un mejor pronóstico incluyen el diagnóstico prenatal, la atención neonatal regionalizada, la debida identificación de entidades asociadas, métodos quirúrgicos innovativos y nutrición parenteral a largo término y no complicada.

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