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Multiple Atresias of the Bowel with Reference to Tandler's Theory of Embryopathogenesis

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Atresia of multiple segments of bowel has been reported with an incidence ranging from 6-29% in different series(1). Association of duodenal atresia with multiple atresias of the small and large bowel is very rare. No satisfactory theory has been put forward to explain the etiopathogenesis of multiple atresias of bowel. In the present case of multiple atresias of bowel, histopathological evidence supporting Tandler's solid cord theory of embryogenesis of atresias of bowel is evident.

Case Report

A two-day-old premature male child, born after 34 weeks of gestation was brought with history of repeated episodes of bilious vomiting. He had not passed meconium. History of maternal polyhydramnios was present.

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The child weighed 1600 grams and was hypothermic and dehydrated. His abdomen was flat with absence of bowel sounds. The anus and rectum were normal. Plain *X*-ray of the abdomen revealed gas and fluid levels only in the stomach and proximal duodenum. A clinical diagnosis of duodenal atresia was made.

After nasogastric decompression and improving the general condition of the child, he was taken up for surgery. At laparotomy, done through a right transverse supraumbilical incision, there were multiple atretic segments with skip areas distributed over the entire length of bowel. It was decided to resect the atretic segments and anastomose the ends of the bowel in stages because of the poor general condition of the child.

At the initial surgery, the atretic fourth part of duodenum and upper 5 cm of jejunum, 4 cm of atretic proximal ileum and 5 cm of atretic midileal segments were excised and end to end anastomoses performed between the ends of the patent bowel (three anatomoses). Postoperatively, the patient was kept on total parenteral nutrition (TPN).

After one week, during which time the child had gained 150 g in weight, a second laparotomy was done and a 5 cm segment of atretic terminal ileum and ascending colon were excised. An ileostomy was performed by bringing out the distal most part of the patent ileum. The lumen of the distal colon was hardly 0.5 cm in diameter. It was brought out separately as a stoma. TPN was continued postoperatively, but the child did not pass meconium even after 10 days. Plain *X*-ray abdomen showed gas only upto the jejunum with a small air fluid level.

Laparotomy done for the third time revealed an obstruction at the jejunal anastomosis. The anastomosis was dismantled and 2 cm of jejunum was excised and jejunojejunostomy was redone. However, the child developed septicemia after the third surgical procedure and died one month after admission to the neonatal surgical intensive care unit.

Histopathological examination of atretic segments of the small intestine and colon revealed linear channels (*Fig. 1*) lined by low columnar and cuboidal epithelium with glands

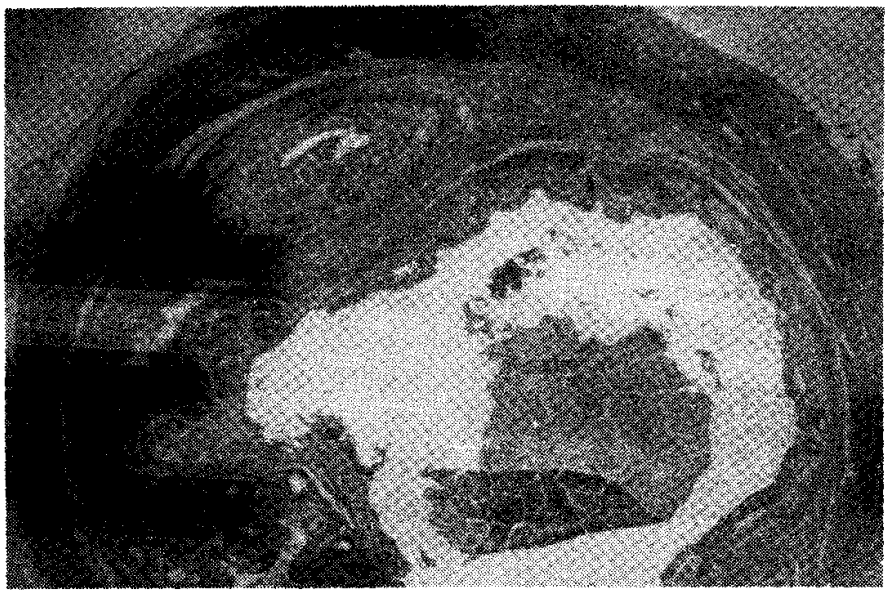

Fig. 1. Photomicrograph of excised jejunal atretic segment showing multiple mucosa lined channels instead of a single column.

in the submucosa. Smooth muscle was seen in only a few section.

Discussion

Although multiple atresias of the bowel have been reported with a varying incidence of 6-29%, only four cases of multiple atresias of bowel involving stomach, duodenum, jejunoileum and colon (in various combinations) have been reported(2,3). Of these Guttman *et al.*(2) reported two cases with multiple septae in the stomach, duodenum and rest of the bowel. Mishalany and Der Kaloustem(3) reported the other two cases of duodenal and multiple jejunal atresia in siblings.

The etiology of multiple atresias is as obscure as it is for single atresia. For many years the most convincing explanation was presumption of failure of recanalization following a stage in development when the intestinal lumen was obliterated(4). Tandler(5) described the development of the duodenum during the second month of gestation. In embryos of 8.5 and 14.5 mm there was such proliferation of the epithelium that the lumen was plugged. This was followed by an increase in the diameter of the surrounding mesoderm, formation of vacuoles in the epithelium and the coalesence of these vacuoles around the 20 mm stage to reform the lumen. However, there is considerable doubt whether the occurrence of atresias, except in the duodenum takes place before 8 weeks of gestation as the bowel distal to ileal atretic segments are known to contain bile stained meconium, squamous epithelial cells and lanugo resulting from bile secretion and swallowing after the 11th and 12th weeks of gestation, respectively.

Volvulus of a part of the small intestine resulting in the twisting off of a segment of gut may also cause atresia(6). The review by de Lorimer, Fonkalsrud and Hays(7) showed evidence of intrauterine bowel infarction in 42% of 449 patients. Atresia due to pulling or nipping off of a loop of intestine within the sac of an exomphalos or hernia was described by Nixon(8) and intrauterine intussusception by Tadoni, Tabuchi and Tanaka(9).

The so called vascular theory, which postulates that failure of the blood supply to the gut may cause intestinal atresia, was evolved over seventy years ago. Koga and associates(10) were also able to produce atresia of the small bowel in fetal puppies following vascular insults.

However, multiple atresia of both small and large bowel with an intact mesentery and no evidence of vascular insufficiency have been described in families as mentioned earlier. Rittenhouse and his colleagues(11) suggested that since the septae were both transverse and longitudinal and had their own muscularis mucosae, they were unlikely to be the result of epithelial plugging but possibly due to an intrauterine ulcerative process.

In the present case, atretic segments of the small intestine and colon showed the presence of linear channels suggesting that the formation of intestinal lumen was arrested in the stage of coalesence of vacuoles after complete plugging of the lumen with epithelial cells had occurred. This is in contrast to the widely accepted vascular theory of embryogenesis of small bowel atresias. It is possible that atresias of bowel may result in more than one way. Multiple atresias involving different parts of the bowel may result due to failure of recanalization following a stage of complete epithelial plugging of the intestinal lumen early in the process of development of the gastrointestinal tract. On the other hand, atresias involving only either the jejunoileum or colon may

be a result of vascular accidents occurring late in the gestational period.

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NOTES AND NEWS

TRAINEE FELLOWSHIP IN PEDIATRIC GASTROENTEROLOGY

The Department of Pediatrics, Maulana Azad Medical College proposes to conduct a trainee fellowship in Pediatric Gastroenterology for a period of six weeks in October-December 1993. Upto two candidates, deputed by Medical Colleges, will be taken up for this programme.

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