Association of Antral, Jejuno-Ileal Atresias and Biliary Peritonitis – An Unusual Case

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Pyloric and prepyloric atresias are very rare. They form only 1% of all gastrointestinal atresias. Although the lesion commonly occurs as an isolated anomaly, its association with distal bowel atresias such as duodenal, jejunoileal, ileal, ileo-caecal junction and colon has been documented. Similarly, jejuno-ileal atresias may lead to perforation of the bowel proximally with meconium peritonitis though it is rare. However, association of antral atresias and perforated jejuno-ileal atresias seem to be rare. Here, we report an instance of such an occurrence.

CASE REPORT

A full term normally delivered 2.8 kg, 12 hour old girl child was transferred from the maternity ward with a history of abdominal distension and vomiting.

During the pregnancy, mother had an evidence of polyhydramnios and she had 5 deliveries earlier. The first three children are alive and well but the last two died within 4 days after birth.

During these two pregnancies the mother had history of polyhydramnios also. The fourth baby died due to neonatal sepsis. The fifth baby had multiple congenital anomalies which included hydrocephalus, malformed ears, bilateral inguinal hernias and renal masses. The exact diagnosis of renal masses could not be made as the neonate died within 24 hours. There was no history of consanguinity.

The present newborn patient had no clinical evidence of jaundice. The abdomen was distended and the skin over it was shiny but not oedematous. There was no visible gastric peristalsis. Ascites was present.

Investigation. Hb 15.9 gm%, WBC total 17,900/cumm, blood urea 36 mg% and serum electrolytes were within normal limits. X-ray film of the abdomen in erect position revealed a single bubble and a distended contour (Figure 1). There were no calcifications in the peritoneal cavity. Ultrasonography of the abdomen confirmed the presence of ascites. Contrast study using gastrografin showed complete obstruction in the antral region. Barium enema revealed “microcolon” but there was no evidence of obstruction in the colon. The laparotomy findings are illustrated in Figure 2. The peritoneal cavity contained about 350 ml. of clear bile. The bowel was malrotated and there were multiple perforations in the jejunum. There was outpouching of the ileum, proximal to the ileal obstruction. The bowel distal to it was collapsed. The atretic portion of the antrum was excised and gastroduodenal anastomosis was performed. Also the perforated segment of jejunum was excised and jejunoileal end to end anastomosis was carried out.
Histopathological studies of the excised antral diaphragm showed fibrous septum lined by mucosa on either side with a central aperture. The jejunal septum also had such a central aperture, but the ileal septum had no such opening. Histologically the musculature of the perforated segment of jejunum contained normal muscular layers in its wall. Though the patient survived this extensive operative procedure, he succumbed to the sepsis and died on 4th postoperative day.

DISCUSSION

Clinically and radiologically, the diagnosis of gastric outlet obstruction was obvious on
Fig 2. Drawing showing laparotomy findings.
A. Antral diaphragm with central opening.
B. Common bile duct
C. Jejunal diaphragm with central opening
D. Perforated jejunum
E. Outpouching of ileum
F. Ileal diaphragm
G. Mobile colon

this neonate. The obstruction was due to the antral diaphragm with a tiny aperture at its centre which did not allow the air to pass through. In any instance when the obstruction at the gastric outlet is complete, the abdominal contour should be scaphoid and distension should be absent. However, in our patient this finding was absent, and instead the abdomen was considerably distended. This distended contour of the abdomen lead us to suspect some other associated lesion.

Extensive gastrointestinal atresias are usually familial and are characterised by presence of intraluminal calcification on the plain x-ray films of abdomen.\(^{36}\) Although there was a history of congenital malformation in one of the siblings of the present patient, the exact diagnosis was not established. Moreover, in the present patient there were no intraluminal calcifications on the plain x-ray films of the abdomen. Therefore, it is difficult to assess whether this patient falls within the spectrum of the syndrome of familial hereditary multiple gastrointestinal atresias. In our patient, the presence of large amount of sterile bile in the peritoneal cavity was due to the multiple perforations in the jejunal segment. The multiple perforations in this site could be explained on the basis of closed loop obstruction in the segment. Proximally, septum in the jejunum and distally atretic lesion in the ileum could have lead to increase in the intraluminal pressure and thus to perforation. Theoretically some decompression could have occurred through the tiny aperture in the proximal jejunal septum, but not adequate enough to prevent the distension of the closed loop. It also could be hypothesized that the continued increase in the pressure in the closed loop could have lead to the dilatation of biliary tract. But in this patient at laparotomy, biliary tract looked normal. The possible explanation is that though there could have been dilation of the biliary tract due to back pressure, it returned to normal once the bile continued to drain directly in the peritoneal cavity leading to ascites.

The clinical significance of this presentation is that when a 'single bubble' gas is present in the stomach along with a distended abdomen, a possibility of a second lesion in the intestinal tract should be considered.

REFERENCES

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L-Thyroxine Therapy for Congenital Hypothyroidism and Marfan Syndrome

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The effect of thyroid hormone on the skeleton has been a bone of contention. Recent data suggests that long-term L-thyroxine (L-T4) therapy, which is often given in supraphysiologic dosages, may predispose patients to decreased bone density in the hip, and may increase the risk of age-related bone loss. The appropriate replacement dose of L-T4 is now considered to be 1.6 μg/kg body weight. We report here a patient with congenital hypothyroidism (CHT), on long-term L-T4 therapy, who in her teens showed features of Marfan syndrome.

CASE REPORT

A 13-year-old girl was referred to us for scholastic backwardness. She was born one month postdated of a non-consanguineous union. At three months of age she was found to cry excessively on lying supine. She had prominent belly with umbilical hernia, coarse facial features, large tongue, hoarse cry, dry and thick skin. The consulting pediatrician made a clinical diagnosis of CHT. Since about five months of age she was on continuous L-T4 therapy, the daily dose ranging from 100 μg to 300 μg. Thyroid function tests were however done only during the last six years with TSH levels ranging from 0.25 to 0.9 mU/ml. About one year back she had complained of tiredness, palpitation and breathlessness on exertion. Thyroid function tests revealed T3 level of 1.9 μg/ml (N = 0.8–1.6 ng/ml), T4 levels 18.6 μg/ml (N = 50–11.5 μg/dl) and TSH 0.25 mU/ml (N = 0.5–4.0 mU/ml. L-T4 dose was reduced from 300 to 200 μg per day.

On physical examination she was a tall, thin girl with underdeveloped musculature, and body weight of 34 kg. She had arachnodactyly, hyperflexible joints with positive thumb sign and wrist sign, scoliosis, flat foot and high arched palate. Her armspan was...