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Case Report

Intestinal Duplication Cyst - a rare cause of intestinal obstruction in neonates

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Abstract

A 2.3 kg baby boy born via vaginal route at 32 weeks' gestation with suspected intestinal obstruction and diagnosed to have duplication cyst intra operatively. Baby stayed with us for 5 months and had to undergo 4 operations as anastomotic leak was very common and inadequate weight gain. Finally, Bishop-Koop operation done and baby was discharged with good weight gain.

Keywords: congenital anomaly, duplication cyst, intestinal obstruction, neonate.

Introduction

The incidence of gastrointestinal duplication cyst (DC) is about 1 in 4500 live births and is found in 0.2% of the children, there seems to be a slight preponderance in males[1].

They can present at any age but 80% of cases present within first two years and majority within first three months of life[2]. Theories regarding etiology include the persistence of fetal bowel diverticula, vacuolization, caudal duplication, and split notochord, but the theory that has gained the most acceptance has been split notochord syndrome[3].

Case Report

Preterm baby 32 weeks' gestation was born on 9/11/18 by normal vaginal route at a private hospital with birth wt. 2.3 kg. Baby cried immediately after birth APGAR not known. Feeding was tried which the baby continuously vomited so baby was admitted and IV fluids and antibiotics were started, X-RAY done showed air fluid levels and baby was referred to our hospital on 13/11/18.

Baby was admitted in Neonatal ICU and relevant investigations sent, X ray showed dilated and fixed bowel loops and air fluid levels, so baby kept Nil by mouth and started on IV antibiotics and other supportive measures and was taken up for surgery. Intraoperative findings showed free blood and fecal matter in peritoneum, large mass arising from terminal small bowel leading to volvulus and gangrene of entire terminal ileum and perforation with extensive ischemia of small bowel. Small bowel with duplication cyst was removed and anastomosis done. There was 45 cms of small bowel from DJ flexure to ileocolic anastomosis. Baby remained stable post OP for 5 days but developed wound dehiscence with fecal fistula and was again operated on 20/11/18 and leak was contained and there was no fecal contamination of peritoneal cavity. Ileostomy was done. Continuous nasogastric

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infusion feeds were started. IV antibiotics were continued. TPN along with jejunostomy feeds were continued but baby started to have large volume ileostomy losses with no weight gain. Baby also developed cholestastic jaundice which was managed as per protocol. Blood culture sent at admission showed a growth of Pseudomonas and second culture showed candida tropicalis for which Amphotericin B and Injection colistin were added. In view of culture positive sepsis LP was done which was normal. Blood products were transfused as per need. In view of poor weight gain, electrolyte and fluid imbalances, the parents were counseled and a decision for early stoma closure was planned with the understanding of high risk for anastomotic leak and other complications. As a last resort the baby was taken up for early stoma closure on 2/1/19 and laparotomy and adhesiolysis and end to end anastomosis was done. Per-operative cholangiogram and liver biopsy was done, in view of cholestatic jaundice, which revealed patent bile ducts and Liver biopsy showed changes consistent with septicemia. Post OP baby remained stable for 4 days but developed abdominal distension on 6/1/19 so X-RAY done which showed air under diaphragm, lower GI contrast done showed leak at anastomotic site.

Baby was again taken for surgery on 7/1/19 and anastomotic leak was identified. There was fecal peritonitis and after peritoneal lavage, ileostomy and distal mucous fistula were fashioned. Baby was started on continuous feeds but developed diarrhea as feeds were reached to full, so on and off cutting of feeds were done and as baby was not gaining weight and urinary sodium was persistently low so was operated again on 13/3/19 (Bishop-koop operation done).

Baby is now stable and diarrhea resolved and was discharged on 23/4/19 (5 months of age). Discharge wt: 2.827 Kg, Length: 54 cm, HC: 36.5 cm.

Discussion

The first DC was described in 1733 by Calderin, followed by another in 1884 by Fitz. In 1937, Ladd defined DCs as having the following three properties: (a) the cyst is surrounded by smooth muscle, (b) the cyst must contain the GIS mucosa from which it takes its own origin, and (c) the cyst must have a wall in common with the anatomic region in which it is found. DCs can be found anywhere in the GIS, from the mouth to the anus[4]. They are seen most often in the ileum (30%) and the ileocecal region (30%). Their frequency in other areas of the GIS has been reported as follows: 10% in the duodenum, 8% in the stomach, 8% in the jejunum, 7% in the colon, and 5% in the rectum generally located on the mesenteric side[5].

The complications include bleeding into the cyst, intestinal obstruction, intestinal or duplication cyst perforation, volvulus, cyst torsion, cystic rupture, and malignancy (3% sarcoma, lymphangiosarcoma)[6].

The diagnosis of duplication cyst in the neonatal period is extremely difficult because the symptomatology is nonspecific. Only extremely large DCs might result in a prenatal diagnosis[7].

The imaging modalities commonly used to investigate duplication cysts are X-ray abdomen, USG, barium studies, CT scan and MRI as they are helpful in defining the anatomic borders of a

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duplication[8]. USG of duplication cysts demonstrate an echogenic inner mucosal layer and a hypoechoic outer muscular layer - "double-wall" sign which is highly indicative of enteric duplications and thus USG is considered to be the first-choice imaging modality[9,10]. CT scan and MRI are not used routinely but are quite helpful in difficult cases. Radionuclide scanning with technetium-99m sodium pertechnetate can be used in cases with suspicion of presence of heterotopic gastric mucosa.

The treatment of choice for GIT duplication remains surgical excision taking into account the location, size and benign nature of lesion. This is easily achieved in cystic variants. However, in tubular duplication cysts there is involvement of large portion of bowel which on resection might result in short bowel syndrome. Therefore, in these cases mucosal stripping is suggested thereby preventing risk of peptic ulceration or carcinogenesis[11,12].

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