

CASE SERIES

Duplication Cyst with Intestinal Volvulus Causing Intestinal Atresia/Stenosis in Neonates

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ABSTRACT

Duplication cysts of small bowel seldom present in newborns and usually represent the development of complications. In utero complications may lead to mesenteric vascular accidents and thus resulting in intestinal atresias. We report three neonates with duplication cyst of small bowel causing localized intestinal volvulus, leading to small bowel intestinal atresia/stenosis. The neonates underwent excision of the duplication cyst and resection anastomosis of the small bowel. Post-operative recovery was uneventful in all three of them.

Key words: Atresia; Duplication cyst; Stenosis; Volvulus

INTRODUCTION

Volvulus due to duplication cyst is a rare cause of neonatal intestinal obstruction [1]. In utero volvulus due to duplication cyst may also produce intestinal atresia [2,3]. Delayed diagnosis of volvulus is known to contribute to a high rate of morbidity and mortality. Herein, we report three neonates having duplication cyst, leading to localized intestinal volvulus resulting in intestinal atresia or stenosis.

CASE REPORTS

Case 1

A 2-day-old male full-term baby, weighing 2.8 kg, presented with abdominal distension, bilious vomiting, and failure to pass meconium soon after birth. He was a product of non-consanguineous marriage and born at home by spontaneous vaginal delivery (SVD), attended by lady health visitor (LHV). On examination, the neonate was dehydrated and had abdominal distension; he passed mucous on rectal stimulation. He had 40 ml of bilious drain on nasogastric intubation. Laboratory investigations were within normal ranges. Erect abdomen X-ray showed air-fluid levels; ultra-

sound of abdomen showed dilated bowel loops. After adequate resuscitation, the patient was operated with suspicion of intestinal atresia. At surgery, a 3 cm × 5 cm sized non-communicating cyst was encountered with 3–4 twists causing localized volvulus. Further, exploration after detwisting showed type III-A ileal atresia (midileum); the cyst was attached to ileum distal to atresia (Figure 1). The cyst with small portion of distal ileum and few cm of dilated ileum proximal to atresia was resected and ileoileal end-to-end anastomosis was performed. The post-operative recovery was uneventful, and he was discharged on full feeds on the 8th post-operative day. The histopathology of the cyst was consistent with ileal duplication cyst with ectopic gastric mucosa.

Case 2

A 9-day-old female baby (full term, SVD, 2.5 kg) presented with neonatal intestinal obstruction since birth and was being managed in a local hospital with intravenous fluids and antibiotics. The antenatal scan had shown polyhydramnios and a 3.5 cm × 2.7 cm sized cystic structure in the abdomen. Abdominal examination showed upper abdominal fullness and a cystic mass in the midabdomen. X-ray abdomen revealed 2–3 air-fluid levels. Ultrasound abdomen

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showed hypoechoic cystic mass in the midabdomen suggestive of mesenteric cyst. After optimization, she underwent surgery, which revealed a non-communicating cyst having intimate contact with jejunum that caused localized midjejunal volvulus along with Type III-A jejunal atresia (Figure 2). The cyst was resected and jejunojejunal end-to-end anastomosis was performed. The patient was allowed orally on the 5th post-operative day and discharged on full feeds on the 7th post-operative day. Histopathology confirmed it a duplication cyst of jejunum along with ectopic gastric mucosal lining.

Case 3

A 3-day-old male neonate (full term, SVD, 3 kg) presented with neonatal intestinal obstruction since birth. On examination, there was a huge cystic mass palpable and occupying the entire right side of abdomen (Figure 3). X-ray abdomen showed few dilated gas shadows in the left upper quadrant (Figure 3). Ultrasound abdomen revealed 5 cm × 5 cm cyst suggestive of a mesenteric cyst. After optimization, the surgery was performed that divulged a huge cyst in the mesentery of jejunum, intimately attached to the jejunal wall (Figure 3). The cyst had resulted localized small bowel volvulus that led to stenosis of jejunum, just proximal to the cyst attachment to the jejunum; proximal jejunal loops were hugely distended. Through the

stenotic part of bowel, only negligible amount of bowel gas could be negotiated with pressure (Figure 3). The cyst was initially decompressed followed by excision along with part of the intimately attached jejunum; bowel continuity was restored with end-to-end jejunojejunal anastomosis. Post-operative recovery remained uneventful and the patient was discharged in good condition on the 7th post-operative day. The histopathology showed it a jejunal duplication cyst.

DISCUSSION

Duplication cysts of alimentary tract are named as per their attachment to the part of normal gastrointestinal tract (GIT). The common duplications are ileal in origin. The criteria for a cyst to be classical duplication cyst include intimate contact with any part of GIT, smooth muscles in the walls of the cyst, and GIT mucosa lining the cyst [3]. All these criteria were met in all of our index cases. Our case one had ileal duplication cyst, whereas cases 2 and 3 had jejunal duplication cysts. Ectopic gastric mucosa in the cyst lining was seen in two cases on histopathology.

The presentation of duplications cyst, in majority, is within the first 2 years of life. Presentation in neonatal life is rare and usually indicates some sort of complications related to the duplication cyst [1,2]. Common complications are hemorrhage within the cyst, infection, spontaneous perforation of the cyst, intestinal obstruction, and twist of the cyst causing localized

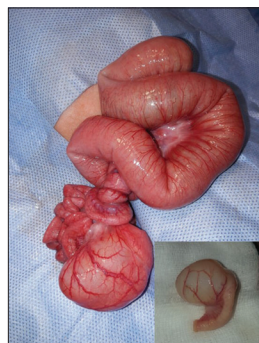


Figure 1: A cyst with twists of small bowel and its mesentery. Inset shows resected cyst attached to distal ileum

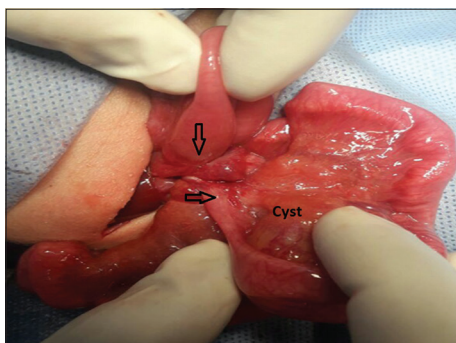


Figure 2: Cyst in the mesentery of jejunum. Arrows showed proximal and distal bowels at the site of atresia

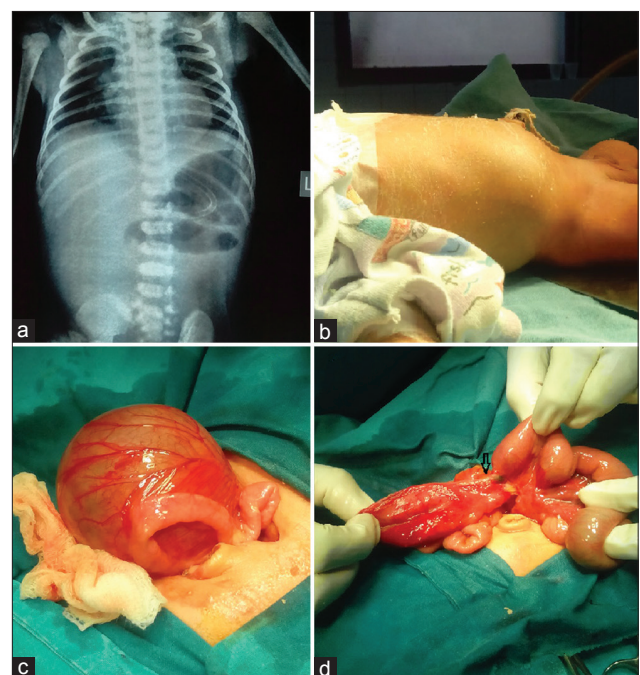


Figure 3: (a) X-ray abdomen. (b) A visible mass in the right abdomen. (c) Duplication cyst intimately attached with jejunum. (d) Decompressed cyst. Arrow shows stenotic area and proximal jejunal dilated loops

volvulus [4]. In utero complications of duplication cyst often lead to the development of intestinal atresias. Few reports of intestinal atresia due to antenatal volvulus of duplication cyst have been reported in literature [1-3]. In our first two cases, intestinal atresias just proximal to the site of duplication cyst; and in our third case, intestinal stenosis just proximal to the duplication cyst, were found. The atresia/stenosis was obviously related to the in utero vascular insult related to intestinal volvulus caused by the duplication cyst. Intestinal stenosis as a complication of in utero volvulus secondary to twisting duplication cyst is extremely rare event. It can be speculated that an early in utero volvulus might lead to the development of intestinal atresias and a late event might only cause intestinal stenosis.

Although the sonologist's diagnosis was that of mesenteric cyst in two of the three cases, this entity is rarely encountered at this age [5]. Other differential diagnoses in a neonate with intestinal obstruction related to a cyst in abdomen, since birth, could be duplication cyst, giant meconium cyst, or rarely a segmental dilatation of bowel [6,7].

The volvulus may involve small part of bowel or it may involve extensive length of bowel. Mainstay of the treatment is urgent surgical exploration as a delay in diagnosis and intervention may lead to sinister outcomes.

CONCLUSION

Duplication cyst with antenatal volvulus may cause a myriad of complications. In utero volvulus leads to mesenteric vascular events and the development of intestinal atresias and stenosis. Postnatal volvulus is a lethal complication and should be picked up and treated promptly.

Author's Contribution

All authors contributed equally in concept, design, literature review, drafting the manuscript, and approval of the final manuscript.

Consent Statement

Authors declared that they have taken informed written consent, for publication of this report along with clinical photographs/material, from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient however it cannot be guaranteed.

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