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CASE REPORTS

SPONTANEUOS PERFORATION AND INTUSSUSCEPTION OF ILEUM IN FETUS SECONDARY TO RUPTURE OF OMPHALOCELE MINOR: REPORT OF TWO CASES

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ABSTRACT

We are reporting two cases of intrauterine perforation of ileum with intussusception of ram's horn type in one case and unihorn in other case following rupture of omphalocele membrane. Both cases were managed successfully by surgical intervention. Aim of this study is to add in literature that this type of complication can occur with rupture of omphalocele minor during intrauterine period.

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INTRODUCTION

Omphalocele is the congenital abdominal wall defect with herniation of intra-abdominal organs covered by peritoneum, Wharton's jelly and amnion. Estimated incidence of omphalocele is 1 in 4000 to 10000 live births (Peranteau *et al*., 2015; Strauss and Kuller, 2004). If the size of abdominal wall defect is < 5 cm, it is classified as omphalocele minor and > 5cm defect is called omphalocele major. Various other associated congenital malformations are reported like Meckel's diverticulum, undescended testis, Beckwith-Wiedmann syndrome, ventricular septal defects and chromosomal anomalies (Liang *et al*., 2013).

Case reports

Case 1

A newborn male child was referred to the emergency ward of our hospital with a horned mass coming out through the abdominal wall. The full term baby was born by uneventful vaginal deliveryss to a multigravida mother. The baby weighed 2.5 kg and had cried immediately after the birth. The antenatal USG was suggestive of gastrochisis.

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On gross examination, gut loop covered by thin membrane was seen protruding through defect in umbilicus. It was omphalocele minor through which small gut was prolapsed leading to ram's horn (bilateral) intussusceptions (Fig 1a). A perforation of size 2×2 cm was seen on antimesentric side of prolapsed gut (Fig 1b). X-ray abdomen showed multiple air fluid levels. USG abdomen showed no other associated anomalies. After adequate resuscitation, baby was taken for emergency laparotomy done through sub umbilical incision. The sac of omphalocoele was excised, gut was delivered, intussusception was reduced and the gut perforation was closed in layers. Post operative course of the patient was uneventful.

Case 2

A full term female baby weighing 2.8 kg was born by uneventful vaginal delivery at government hospital. Baby cried immediately and was pink in color. Baby had regurgitation of feed multiple times. Antenatal USG was normal study. But baby was referred to our centre in view of mass protruding through anterior abdominal wall (Fig 2). On examination, there was gut protruding out through small umbilical defect. Prolapsed part was associated with intussusception and perforation of gut on antimesentric side. X-ray and USG abdomen done, which were normal. After initial resuscitation, baby was subjected to emergency laprotomy.

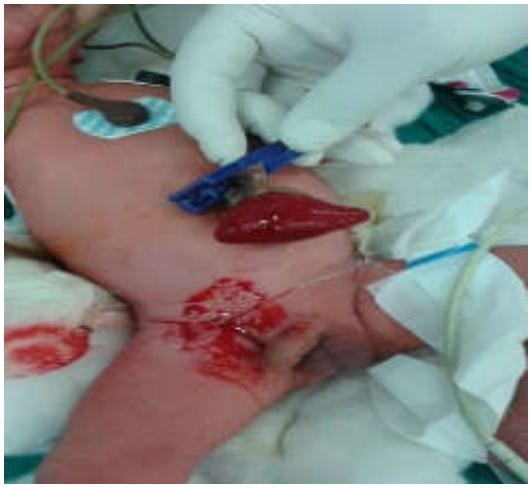

Figure 1a: Bilateral (rams horn) prolapsed of gut through umbilical defect associated with intussusceptions

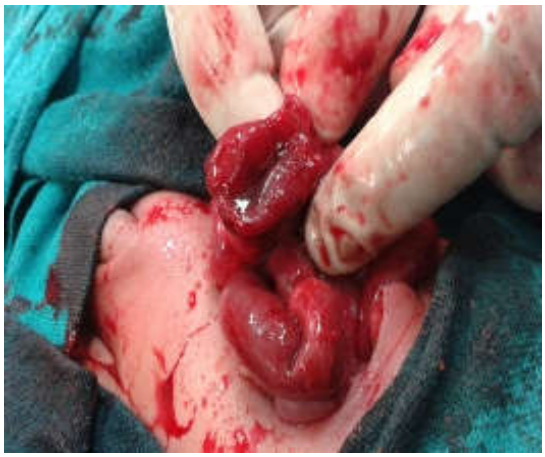

Figure 1b: Large perforation of ileum at antimesentric side

In laprotomy intussusceptions was reduced and gut perforation was closed in layers. No complication was detected in post operative. During hospital stay we had done ECHO, repeat ultrasound abdomen, which ruled out other associated congenital anomalies in both patients. The patients were discharged after 10 days and are doing well on follow up.

DISCUSSION

Ompalocele is congenital anomaly of anterior abdominal wall occurring due to failure of closure of lateral folds. Abdominal and/or pelvic viscera can herniate in to the sac through umbilical defect. There is no skin, abdominal muscle and fascial covering on sac. These neonates have increased risk of other gross congenital anomalies in 27% to 70% cases and chromosomal abnormalities in 10% to 30%. Cardiac and gastrointestinal anomalies are more common. Incidence of chromosomal abnormality is more if the size of defect is less than 4 cm i.e. in omphalocele minor (Lurie *et al*., 2013). These anomalies play important role in outcome of patients. In our case there was no chromosomal and other serious associated anomalies detected. The cause of perforation in index case may

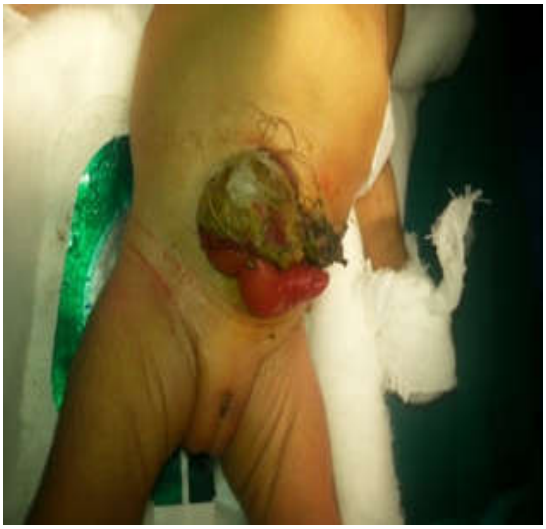

Figure 2: Unilateral prolapsed gut associated with intussusception

be because of the firm attachment of membrane with the underlying gut. Because of the rupture of the membrane that might have resulted in the perforation leading to intussusception. It's our observation that intrauterine rupture of the small gut is associated with intussusception in omphalocele minor. In literature this type of intussuception is reported only in patent VID and that also in postnatal life (Lone *et al*., 2015; Borkar, 2013; Panait *et al*., 2013; Rohtagi and Gorthi, 1984; Blair and Beasley,1989). Index case was different from earlier cases as (1) all events took place in intrauterine period (2) no evidence of patent VID was found preoperatively (3) site of involvement is away from typical location of VID i.e. two feet from ileocecal junction and (4) it was a wide perforation with intussusception, not the mucosal prolapsed. Omphalocele can be diagnosed during antenatal USG investigation. In our case, it was diagnosed as gastrochisis on antenatal USG, so these mothers were referred to our tertiary care centre for further management.

USG guided antenatal supervision of pregnancy is of great importance in early detection of omphalocele and timely intervention to improve outcome (Liang *et al*., 2013). Surgical interventions are the mainstay for treatment which can be primary or delayed. Type of surgery depends on size of defect, organs involved in herniation and extent of involvement of other organs like pulmonary hypoplasia. In our case the intussuception was reduced and perforation of gut was closed and primary repair was done.

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