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Ileal Volvulus without Malrotation in Newborn-Rare Presentation

Case Report

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Abstract

Intestinal volvulus is a rare but life-threatening surgical emergency. Volvulus without malrotation is extremely rare, only few cases are reported till now in literature [1]. Prompt diagnosis and urgent surgical treatment is required if bowel necrosis is to be prevented, which is associated with increased mortality. We present a case of new born preterm baby (32 weeks) who presented with distension of abdomen immediately after birth which was gradually increasing. An USG abdomen revealed a "whirlpool sign" in the umbilical region, suggesting small bowel volvulus with intestinal obstruction. Laparotomy revealed a twisted, necrosed loop of small bowel in the umbilical region with abnormal adhesions between the distal ileum and caecum with mesenteric narrowing. Subsequent resection of the involved segment was done. Baby was discharged in stable condition on 7th post-operative day.

Keywords: Volvulus; Malrotation; Bowel Necrosis; Gangrenous.

Background

Majority of neonatal midgut volvulus cases are due to malrotation. Some other causes can be due to postoperative adhesions, duplication cyst, meconium "ileus", and internal hernia. [2] Small bowel volvulus refers to the abnormal twisting of a loop of small bowel about the axis of its mesentery. This results in partial or complete mechanical intestinal obstruction and eventually to bowel ischemia, necrosis, perforation and peritonitis. [3] Segmental volvulus without malrotation is extremely rare and has been reported only in few studies. Common clinical presentation is bowel distension with features of intestinal obstruction. Volvulus, in general, leads to necrosis of the involved bowel segment. Prompt diagnosis and treatment are required to prevent unnecessary resection and associated increased morbidity and mortality.[4]

Case presentation

We report a case of preterm baby (32weeks), weighing 1400 g, with APGAR score 7 and 8 at 1 and 5 min, respectively who presented with distended abdomen and vague lump in right lumbar region after birth. Investigational work-up was started including chest X-ray which was normal, arterial blood gases showed mild metabolic acidosis. The baby was referred to our radiological department for plain abdominal X-ray in view of distended abdomen. The X-ray showed a distended and gasless abdomen, except for a small amount of air in the stomach (Figure 1a). As the abdominal distension is increasing and due to excessive cry of baby, radiograph was repeated after 4 hrs which showed gas less abdomen in right lumbar, umbilical region and pelvis with few airs filled bowel loops to left hypochondrium (Figure 1b).

In view of distension of abdomen and vague lump, USG abdomen was done which showed aperistaltic dilated fluid filled bowel loops with maximum diameter of 2cm in umbilical region with twisting giving an appearance of 'whirl pool'(Figure 2). Upper GI study using water soluble contrast was done which ruled out a malrotation showing a normal gastric, duodenal, and normal location of the

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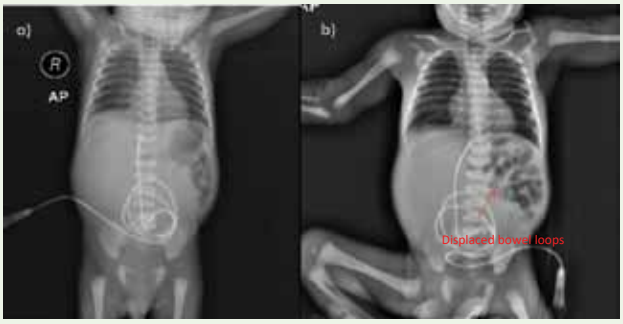

Figure 1: a:Supine radiograph taken after 1 hour of birth showing air filled bowel loops in left hypochondrium. b: radiograph taken after 4 hours of birth showing air filled bowel loops in left hypochondrium with absent gas shadows in rest of abdomen.

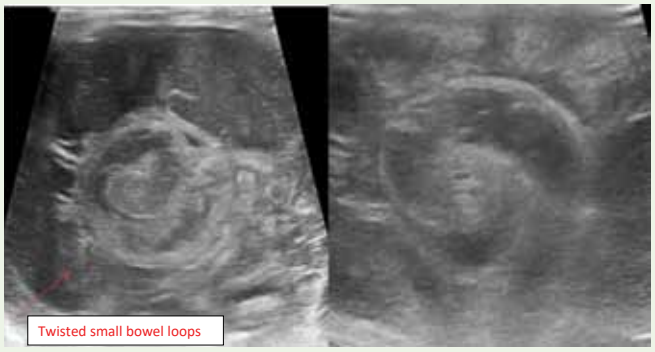

Figure 2: Dilated bowel loops with twisting noted in periumbilical region giving an appearance of whirl pool sign. Maximum diameter of bowel loop is 2cm.

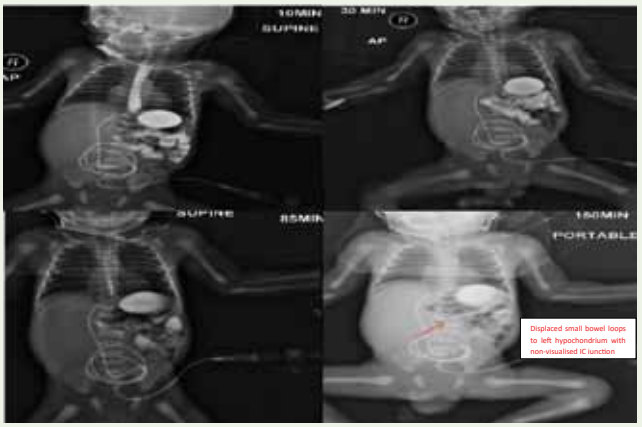

Figure 3: Ugi Study Serial radiographs are taken at 10min, 30min,1hr30min and 2hrs 30min after giving oral contrast. Oesophagus, stomach and duodenum are well opacified. The contrast is seen crossing DJ flexure with opacification of proximal small bowel loops in left lumbar region. Rest of the bowel loops are not opacified in delayed radiographs.

Figure 4: Intraoperative findings: Necrosed distal ileum noted close to IC junction with twisting – s/o ILEAL VOLVULUS. Proximal small bowel loops are normal.

Treitz ligament (Figure 3). Based on imaging, provisional diagnosis of intestinal (ileal) volvulus without malrotation was made. The baby was operated on immediately after 10 hrs of birth in view of intestinal obstruction. Intraoperative findings showed Ileal volvulus with necrosed segment up to 30 cm extending up to IC Junction coursed by adhesive band causing the twisting of loops. Proximal healthy small bowel up to 50 cm was observed (Figure 4). Resection of necrotic ileum with santulli enterostomy was done. The postoperative period was uneventful and baby was discharged on 7th post-operative day.

Discussion

Congenital volvulus is a life-threatening condition which requires early and accurate diagnosis in order to identify the appropriate treatment and reduce the risk of neonatal mortality. Midgut volvulus usually occurs during the first year of life, with 60% of cases occurring within the first month.[14] Neonatal volvulus is usually secondary to malrotation and involves twisting of bowel loop with its feeding vessels that leads to ischemia, necrosis, and perforation with fatal complication, if not recognized and treated in time. Intestinal volvulus without malrotation occurs in 19%-20% of the small bowel volvulus in children.[15,16] The etiology of neonatal volvulus without malrotation remains unknown. However, other rare predisposing causes are Meckel's diverticulum, duplication cyst, and meconium plug. Delayed diagnosis can lead to significant morbidity and mortality.[5] In our case, band like mesentry was identified which could be cause for ileal volvulus similar to case series reported by Kitano et al., where there was long, narrow, band-like mesentery seen in 3 cases, but the authors could not clarify whether this was the result or the cause of the volvulus.[8]

The typical clinical picture of primary segmental volvulus is acute intestinal strangulation manifesting early in life, and such conditions have even been reported in fetuses. Compared to volvulus with malrotation, the ischemic changes in the twisted bowel without malrotation are thought to progress rapidly because the colon does not act as a cushion.[9]

Diagnosis of volvulus in any suspected neonatal intestinal obstruction by plain film and barium study is extremely difficult especially when malrotation is ruled out.[2]

The sensitivity, specificity and positive predictive value of the clockwise whirlpool sign for midgut volvulus are 92%, 100% and 100%, respectively. It should be noted, however, that sonography is operator dependent and this may result in a missed diagnosis resulting in fatal outcomes. Timely diagnosis of small bowel volvulus is necessary as bowel necrosis is associated with increased mortality.

Prompt diagnosis and surgical management is necessary to prevent the eventual ischaemia and gangrene due to small bowel

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volvulus. The mortality associated with small bowel volvulus has been quoted as high as 42–67% [5]

Conclusion

Based on our case, we suggest that in a case of suspected intestinal obstruction, first we have to rule out intestinal malrotation with an upper gastrointestinal barium study. Once malrotation is ruled out, immediately we need to perform an ultrasound examination of the abdomen to rule out an ileal volvulus as late diagnosis in volvulus could be fatal. Thus, ultrasound examination that shows a target lesion "bird beak" sign of the involved bowel allows to quickly identify this morbid entity without loss of precious time.

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