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Case Report

Prenatal Diagnosis and Management of Long-Standing Intestinal Volvulus

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Abstract

Fetal intestinal volvulus is a life-threatening condition. We report a case in which the diagnosis was difficult to be established due to absence of specific signs for intestinal volvulus on prenatal ultrasound (US). Fetal magnetic resonance imaging (MRI) showing whirlpool sign and coffee bean sign with intermediate intensities on T2, strongly suggested the diagnosis of intestinal volvulus containing meconium or haemorrhage. The premature section caesarean (SC) was performed to safe the baby. However, operation could not be done right after SC because of abdominal plastered due to long standing obstruction. Unfortunately, the baby could not be saved due to repeated perforation and post-operative sepsis. Thus the timing of obstruction antenatally is one of important prognostic factor in fetal volvulus.

Keywords: Intestinal volvulus, prenatal US, fetal MRI, long standing obstruction.

Introduction

Fetal intestinal volvulus is a rare condition. Establishing the diagnosis is challenging. We report a case of intestinal volvulus which was referred to the hospital at 34 weeks of gestation. Intrauterine Sonography showed an echogenic mass suspected as a liver mass. This initial result of US was inconclusive and therefore needed further study. MRI has wide field of view (fov) and good contrast resolution. The fetal MRI showed clearly the origin of the mass and also the whirpool sign which was the indication of volvulus with closed loop obstruction. The pregnancy was then terminated immediately. While proceeding to SC, cardiotocography (CTG) showed contraction and non-reassuring fetal status.

Case report

A 26 year-old woman, gravida1, at 34 weeks of gestation age, was referred to the hospital with the suspicion of fetal intraabdominal mass. Intrauterine US showed hyperechoic lesion adjacent to liver, which filled the whole abdominal cavity, without peristaltic movement, thus mimicking liver mass (Figure 1a) and without color flow on Doppler US (Figure 1b). Scalp oedema was observed as well (Figure 1c). Fetal MRI showed multiple loop bowel dilatation (Figure 2a) with whirpool sign and coffee bean sign (Figure 2a,b), strongly suggested that the lesion was originated from intestine with intermediate signal intensity at T2W and lack of meconium intensity in the large bowel or rectum on T1W.

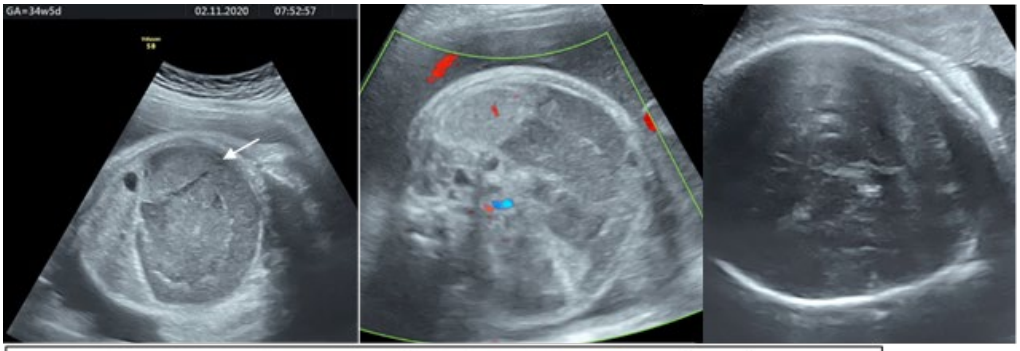

Fig 1a. Echogenic mass adjacent to liver filled whole abdominal cavity (arrow).
Fig 1b. Doppler US showed no vascularisation at the mass.
Fig 1c. Scalp edema (arrow)

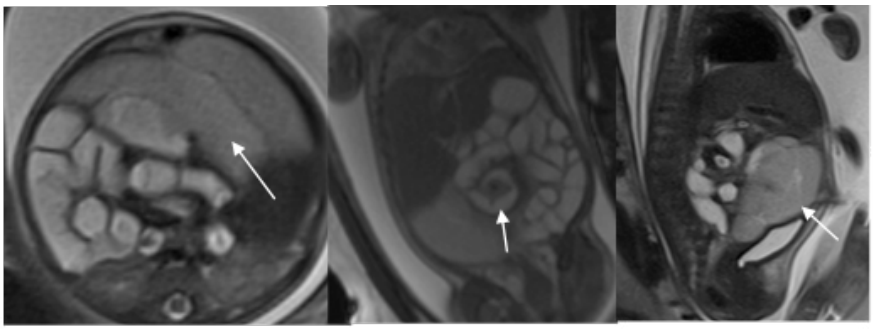

Fig 2a. Fetal MRI axial T2 , showed multiple loops of huge dilated bowel with intermediate signal intensity (white arrow), compared with mild dilated high intensity of surrounding bowel (black arrow). Fig 2b. Coronal T2TRUFISP showed swirling pattern of dilated bowel loop (whirpool sign). Fig 2c. Sagital T2HASTE showed coffee bean sign.

Follow up doppler US was performed 1 day after MRI examination, with peak systolic velocity of middle cerebral artery 1.67 MoM, suggestive of fetal anemia. Since life-threatening condition and viable fetus was expected, it was decided to have an immediate SC. At the time of observation in the room, before SC procedure, it turned out that the CTG result showed contraction and fetal distress (Figure 3a,b) which led to the decision to termination earlier than the initial plan.

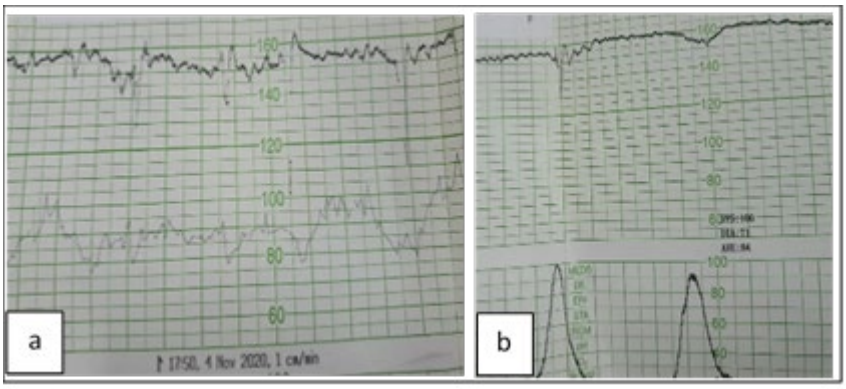

Figure 3a & 3b: normal premature CTG & CTG showed contraction and non reassuring fetal status.

A female neonate was delivered, with birth weight 2214 g, Apgar scores were 2 and 7 at 1 and 5 minutes. The baby was intubated soon after birth and mechanically ventilated for respiratory distress. The abdomen was distended and hyperemia, with dilated vessel on hyperemia area (Figure 4a). Orogastric decompression was performed. Initial hemoglobin value was 12.2 g/dl, other laboratory test results were unremarkable and normal karyotyping. Plain radiograph taken immediately after delivery showed abdominal distention with small amount of gas in the stomach. And follow up photo at 12 hours showed gasless in the right side of the abdomen and dilated bowel loops in the left side (Figure 4b). Surgical management was planned after the hemodynamic condition was stable. On the 3rd day, only peritoneal drainage was performed in order to reduce the infection. Surgical exploration was done on the 13th day, revealing a volvulus of the distal part ileum with 360 degrees twisting, necrosis, perforation and dilated small bowel at the proximal part of volvulus (Figure 5a,b). The strangulated bowel was filled with old blood and meconium. There was no evidence of intestinal malrotation, the duodenojejunal junction at ligament of Treitz. Intestinal obstruction was caused by anomalous congenital bands. After resection of necrotic tissue, ileostomy Santulli was performed due to the different calibre of the small bowel and large bowel diameter. After one month the baby showed a distended abdomen again. Contrast meal was performed and revealed leakage contrast media ± 13.5 cm from Treitz ligament. In the 3rd operation there was indeed a perforation with a severe adhesion and minimal adhesiolysis was performed due to the risk of subsequent perforation at another small bowel segment.

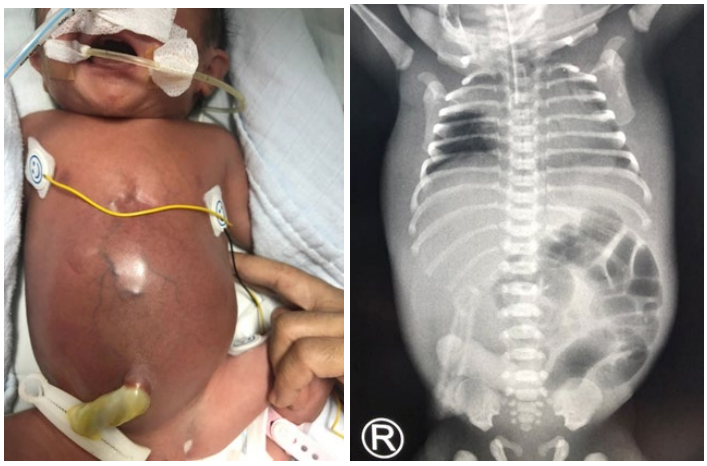

Figure 4a & 4b: Abdominal distended with discoloration of skin and dilatation vascular (Cullen's sign. **&** Plain photo at 12 hours after birth, showed gasless in the right-side abdomen with dilatation of the bowel loops at contralateral side. No meconeum calcification. The tip of orogastric tube is seen within the stomach.

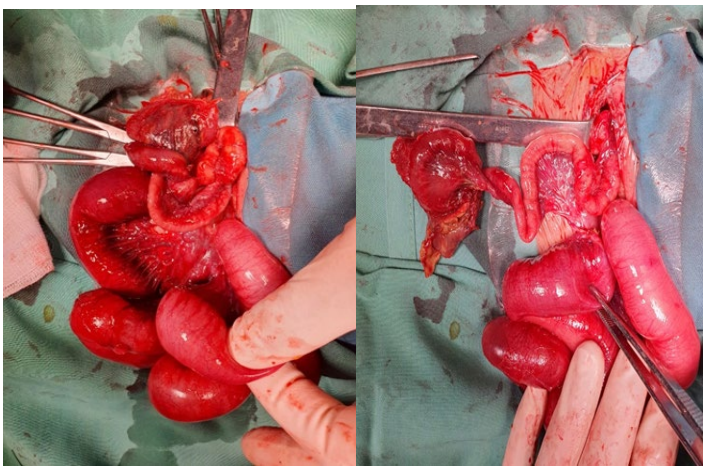

Figure 5a & 5b: Showing volvulus at distal part ileum with dilatation of small bowel proximal volvulus with the length approximately 70-80 cm. **&** After releasing volvulus, the perforated of ileum parts was visible (arrow). This particular part gives the coffee bean appearance on MRI.

Discussion

Intrauterine midgut volvulus is a very rare and life-threatening condition [1-4]. The bowel loops twist around mesenteric artery or its branches resulting in intestinal obstruction, vascular insufficiency and bowel necrosis (1)(2)(5). Delayed diagnosis and intervention can contribute to morbidity and mortality [4]. Majority of intrauterine midgut volvulus cases are due to malrotation, other causes are adhesions, duplication cyst, meconium ileus and internal hernia [3,5]. Occasionally, intrauterine midgut volvulus has been associated with delayed return of fetal midgut into the abdomen [6].

Diagnosis of prenatal volvulus was really challenging. This patient was referred to the hospital at 34 weeks of gestational age because of the suspicion of an intraabdominal mass. Ultrasound showed hyperechoic mass filling abdominal cavity adjacent to liver, without peristaltic mimicking liver mass. It was very difficult to establish the diagnosis of volvulus with US in this case, because it was detected at the late stage, when the bowel has enlarged significantly, with the obscuring intestinal wall, hyperechoic and no peristaltic movement. Furthermore, there was no previous US data for comparison. From the study of 8 midgut volvulus cases by Sciarrone, et al, only 3 cases showed specific features such as whirpool or coffee bean sign. Other cases were diagnosed only based from indirect sign such as dilated bowel loops without peristalsis, pseudocyst, peritoneal calcification, ascites and polyhidramnions [7]. In this patient, the other indirect sign besides dilated bowel was not detected.

MRI examination was performed to confirm the diagnosis. Since MRI showed that the lesion was originated from the bowel loop with whirlpool sign and coffee bean sign, no abnormalities in other organs, then the diagnosis of midgut volvulus was established. A whirlpool sign indicates closed loop obstruction due to internal hernia [8]. Laparotomy exploration revealed midgut volvulus with anomalous bands which twist the bowel loops, without malrotation and internal hernia. Doppler US may allow the diagnosis of midgut volvulus by visualizing the twisted mesenteric vessels that accompany the involved bowel and mesentery, if the involved bowel is not dilated [9]. However, in this case the involved bowel showed huge dilatation with compressed mesenteric vessel. Increased echogenic on US dan intermediate hyperintense on T2 at the affected bowel loop should be considered as hemorrhagic change [10], as in this case was proven by surgery.

The red flag of fetal volvulus for urgent delivery are fetal anemia signs such as increasing peak velocity of MCA, ascites, scalp edema and fetal distress. Fetal anemia is a dangerous condition that can cause IUFD, so it is necessary to consider the right timing to terminate pregnancy [2]. This fetus is already at 34 weeks of age, which is thought to be viable if it is delivered immediately. Therefore, premature SC was planned. During observation for SC, CTG showed contraction and fetal distress. At the time of birth, the fetus was not in good condition. The operation was carried out in stages considering that the baby had unfavorable condition. On day 3, the baby had only drainage of peritoneal contents due to plastered abdomen. Perforation and severe adhesion were found at that time. On day 13, laparotomy exploration and Santulli ileostomy was done because of the difference in the diameter of the small intestine and colon so that it could not be directly connected. The colon calibre was small due to disuse atrophy, while the proximal calibre of the small intestine was dilated. On day 30, the baby showed abdominal distention and perforation at the incision site. Operation was planned to repair perforation at the incision site, unfortunately the intestine was very fragile, that only minimal adhesiolysis was performed. A few days after last operation, the baby's condition was deteriorated and finally she died due to sepsis.

Prognosis of fetal midgut volvulus is not just depends on the length and level of obstruction, the birth weight, the presence of meconium peritonitis, associated anomalies, and gestational age, but also the timing of obstruction antenatally. In this case, body weight was 2200 g, without meconium peritonitis nor associated anomalies. The gestational age was 34 weeks and SC was performed immediately, however the prognosis was still unfavourable due to plastered abdomen which indicated long standing obstruction. This long-standing obstruction caused difficulties in management due to severe adhesion, necrosis and fragile tissue, which led to recurrent perforation. Unfortunately, there was no previous US data to identify the exact time of obstruction.

Conclusion

The diagnosis of volvulus is not easy. Prenatal USG does not always reveal specific findings especially in long standing obstruction with increased echogenicity of dilated bowel loop, loss peristaltic and loss bowel wall appearance mimicking intraabdominal mass. Fetal MRI has excellent natural contrast and wide field of view which can enhance the diagnosis. The timing obstruction of antenatal is one of prognostic factor in intestinal volvulus case.

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