Fetal Primary Small Bowel Volvulus Associated with Acute Gastric Dilatation Detected by Ultrasonography

SACHIYO FUKUSHIMA 1, KAZUMICHI FUJIoka 1,*, MARIKO ASHINA 1, SHOHEI OHYAMA 3, TOSHIHIKO IKUTA 1, KOSUKE NISHIDA 1, HARUNORI MIYAUCHI 2, YUICHI OKATA 2, YUKO BITOH 2, KENJI TANIMURA 3, MASASHI DEGUCHI 3, HIDETO YAMADA 3, and KAZUMOTO IIJIMA 1

1Department of Pediatrics, Kobe University Graduate School of Medicine, Kobe, Japan;
2Department of Pediatric Surgery, Kobe University Graduate School of Medicine, Kobe, Japan;
3Department of Obstetrics and Gynecology, Kobe University Graduate School of Medicine, Kobe, Japan

*Corresponding author

Received 27 August 2018 / Accepted 3 October 2018

Key words: intestinal volvulus, gastric dilatation, prenatal ultrasonography

Fetal intestinal volvulus is a rare condition, and fetal diagnosis of this disease is still challenging, especially in primary cases not accompanied by other comorbidities, such as intestinal malformations. Herein, we report a case of fetal primary small bowel volvulus associated with acute gastric dilatation detected by ultrasonography. We speculate that the mechanism of acute gastric dilatation in our case was peristatic malfunction of the whole intestine caused by a strangulated ileus resulting from fetal intestinal volvulus. In conclusion, acute gastric dilatation detected by fetal ultrasound can indicate the fetal intestinal volvulus.

INTRODUCTION

Fetal intestinal volvulus is a rare condition, and fetal diagnosis of this disease is still challenging, especially in primary cases not accompanied by other comorbidities, such as intestinal malformations 1. We report a case of fetal primary small bowel volvulus associated with acute gastric dilatation detected by ultrasonography.

CLINICAL CASE

A 41-year-old, inherently healthy, primiparous woman was referred to our center because of monozygotic twin pregnancy. She was admitted at 22 weeks’ gestation because of threatened preterm labor. The mother did not have oligo-polyhydramnios during pregnancy. At 32 weeks and 1 day of gestation, fetal ultrasonography revealed no gastric dilatation (Fig. 1a). At 32 weeks and 6 days of gestation, a daily non-stress test showed minimal baseline variability of fetal heart rate and a decrease in the biophysical profiling score of 6 (normal range ≥ 7). At 33 weeks and 0 days, reduced fetal movement, worsening of the biophysical profiling score to 2, and acute gastric dilatation (Fig. 1b). Transverse 3.0 (normal range of the fetus at 33 weeks; 1.6 ± 0.4 cm) cm x Longitudinal 4.0 (2.3 ± 0.5 cm) cm without ascites were detected in one of the twins by fetal ultrasound. Fetal bowel dilatation or bowel wall thickening were not observed. Pregnancy was terminated via emergent cesarean section because of non-reassuring fetal status. The amniotic fluid was clear and transparent without meconium staining.

A male neonate (birth weight: 1584 g) was born as a second twin with Apgar scores of 3 and 6 at 1 and 5 minutes, respectively. He was tracheally intubated soon after birth and mechanically ventilated for respiratory distress. He showed tachycardia (180 bpm) and massive abdominal distention with bluish skin discoloration, but the blood pressure was normal (62/32 mmHg). An initial blood test showed increased C-reactive protein (1.79 mg/dL), lactate dehydrogenase (715 IU/L), and lactate (7.3 mmol/L) levels, but normal levels of creatine kinase (325 IU/L), aspartate aminotransferase (56 IU/L), alanine aminotransferase (8 IU/L), and no anemia (hemoglobin, 16.7 g/dL). A routine X-ray showed a dilated gastric bubble accompanied by minimal intestinal gas (Fig. 1c). Initial abdominal ultrasound showed decreased peristalsis, but no whirlpool sign 34 or ascites. Following physical and radiological examinations, we performed a gastrointestinal contrast test with the suspicion of ileus. The contrast agents did not pass the pylorus in an upper gastrointestinal contrast study and no microcolon was detected in a lower gastrointestinal contrast study (Fig. 1d). Therefore, we diagnosed fetal onset ileus, which required prompt surgery, and performed emergent laparotomy at 9 hours after birth. Laparotomy
S. FUKUSHIMA et al.

showed a volvulus of the distal part of ileum with twisting of 540 degrees accompanied by complete necrosis from 45 cm distal to the ligament of Treitz (total length = 40 cm). There was no evidence of intestinal malrotation or atresia. After detorsion of the volvulus, the necrotic lesion was resected and ileostomy was performed. On the 3rd day after surgery, gastric dilatation disappeared and intestinal gas reached the small intestine, and enteral feeding commenced on the 7th day and reached full feeding on the 14th day. The subsequent course was uneventful and he was discharged on day 86 after ileostomy closure on day 60.

**DISCUSSION**

Midgut volvulus generally occurs based on intestinal malrotation or atresia. Hence, fetal primary intestinal volvulus is a rare and life-threatening condition. Early detection of this devastating morbidity is important, but it is still challenging because of a lack of specific fetal symptoms or ultrasonographic signs.

There are various ultrasonographic signs of fetal intestinal volvulus, including whirlpool sign or coffee bean sign. A literature review did not reveal any previous cases being diagnosed as fetal volvulus via ultrasonographic findings of acute gastric dilatation, except one case prenatally presented with a dilated stomach and bowel loops accompanied by whirlpool sign.

Generally, acute gastric dilatation represents two pathological conditions as follows: (1) mechanical obstruction at the pylorus or duodenum with simultaneous inhibition of the vomiting reflex, or (2) a defective contractility mechanism based on neural or muscular origin. Fetal gastric dilatation visualized by ultrasound has been observed with gastroschisis and other gastrointestinal obstructions, including hypertrophic pyloric stenosis and congenital pyloric atresia. We speculate that the mechanism of acute gastric dilatation in our case was peristaltic dysfunction of the whole intestine caused by a strangulated ileus resulting from fetal intestinal volvulus. This possibility is compatible with the postoperative course where gastric dilatation spontaneously
regressed after surgical repair of volvulus. Regarding primary small bowel volvulus, early intervention in the presence of definitive sonographic findings and fetal distress, and the omission of time-consuming diagnostic modalities under such conditions are generally recommended. We believe careful fetal ultrasound in combination with daily non-stress test might contribute to early intervention in our case, since the infant did not show anemia at birth, which occurs in the consequence of mucosal hemorrhage following long term intestinal ischemia. In conclusion, acute gastric dilatation detected by fetal ultrasound can indicate the fetal intestinal volvulus.

ACKNOWLEDGEMENTS

We thank Ellen Knapp, PhD, from Edanz Group (www.edanzediting.com/ac) for editing a draft of this manuscript.

REFERENCES