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

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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 peristaltic 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 malformations¹. 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 throughout her 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 x Longitudinal 4.0 (2.3 ± 0.5) 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^{3,4} 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

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.

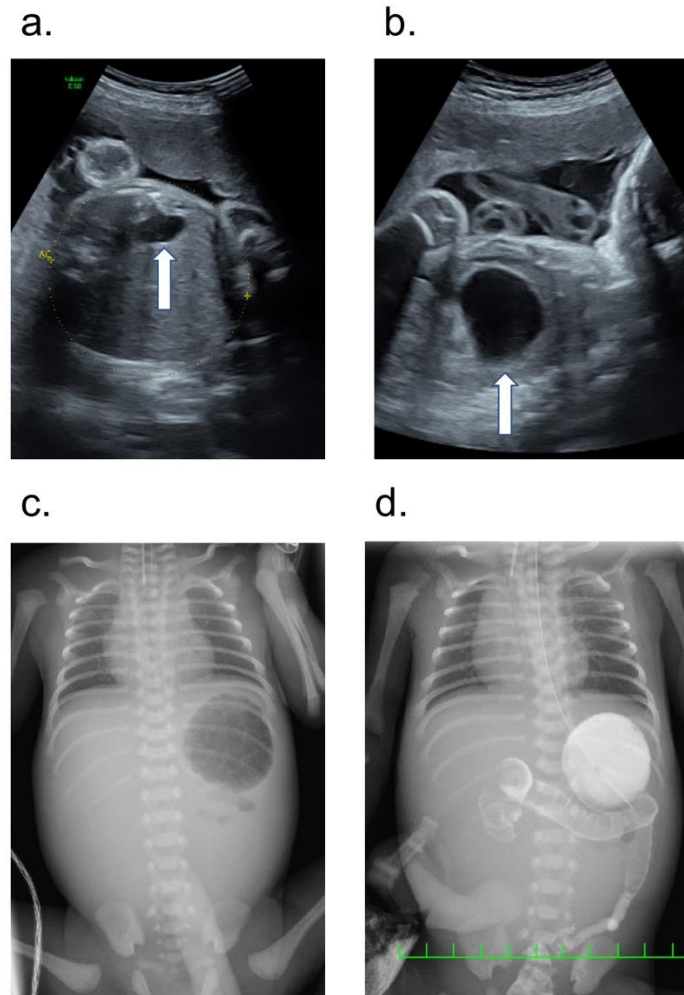


Figure 1. (a) Fetal ultrasonography at 32 weeks, 1 day of gestation without gastric dilatation. (b) Fetal ultrasonography at 33 weeks, 0 days of gestation and (c) an abdominal X-ray at birth and (d) post-contrast study show massive gastric dilatation. The white arrow indicates gastric bubbles.

DISCUSSION

Midgut volvulus generally occurs based on intestinal malrotation or atresia. Hence, fetal primary intestinal volvulus is a rare and life-threatening condition¹. 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 sign³ or coffee bean sign⁵. 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 sign¹.

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 origin⁶. Fetal gastric dilatation visualized by ultrasound has been observed with gastroschisis and other gastrointestinal obstructions, including hypertrophic pyloric stenosis and congenital pyloric atresia⁷. We speculate that the mechanism of acute gastric dilatation in our case was peristaltic malfunction 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.

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REFERENCES

1. **Chung, J.H., Lim, G.Y., and We, J.S.** 2013. Fetal primary small bowel volvulus in a child without intestinal malrotation. *J Pediatr Surg.* **48**: e1-5.
2. **Goldstein, I., Reece, E.A., Yarkoni, S., Wan, M., Green, J.L., and Hobbins, J.C.** 1987. Growth of the fetal stomach in normal pregnancies. *Obstet Gynecol.* **70**: 641-4.
3. **Yoo, S.J., Park, K.W., Cho, S.Y., Sim, J.S., and Hhan, K.S.** 1999. Definitive diagnosis of intestinal volvulus in utero. *Ultrasound Obstet Gynecol.* **13**: 200-3.
4. **Zhang, W., Sun, H., and Luo, F.** 2017. The efficiency of sonography in diagnosing volvulus in neonates with suspected intestinal malrotation. *Medicine (Baltimore).* **96**: e8287.
5. **Park, J.S., Cha, S.J., Kim, B.G., Kim, Y.S., Choi, Y.S., Chang, I.T., et al.** 2008. Intrauterine midgut volvulus without malrotation: diagnosis from the 'coffee bean sign'. *World J Gastroenterol.* **14**: 1456-8.
6. **Wekselman, R.J., Skoryna, S.C., and Webster, D.R.** 1963. Acute Gastric Dilatation. *Gastroenterologia.* **100**: 7-18.
7. **Alfaraj, M.A., Ryan, G., Langer, J.C., Windrim, R., Seaward, P.G., and Kingdom, J.** 2011. Does gastric dilation predict adverse perinatal or surgical outcome in fetuses with gastroschisis? *Ultrasound Obstet Gynecol.* **37**: 202-6.