

Intestinal volvulus without malrotation in a dichorionic diamniotic twin: case report and review of literature

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Summary

Volvulus is a condition in which the small bowel and proximal colon twist around the superior mesenteric artery, and a surgical emergency associated with high intrauterine morbidity and mortality. Intestinal volvulus can occur at any age, but most cases of volvulus occur in infants with anomalies of intestinal rotation and fixation. Intestinal volvulus without malrotation is a rare condition, as only normal intestinal rotation and fixation are present. The authors report a case of intestinal volvulus without malrotation in one fetus of twins.

Key words: Fetus; Intestinal volvulus without malrotation; Dichorionic diamniotic twin.

Introduction

Intestinal volvulus can occur at any age, but is most commonly seen in the neonatal period. Volvulus is a condition in which the small bowel and proximal colon twist around the superior mesenteric artery. Volvulus is a surgical emergency associated with high intrauterine morbidity and mortality. Most cases of volvulus occur in infants with anomalies of intestinal rotation and fixation. Intestinal volvulus without malrotation is a rare condition, as only normal intestinal rotation and fixation are present. For antenatal diagnosis, we use sonography and MRI to identify bowel abnormalities, but it is difficult to make a definitive diagnosis. The authors report a case of intestinal volvulus without malrotation in one fetus of twins.

Case Report

A 38-year-old primigravida had levothyroxine sodium for hypothyroidism. She received a heterologous artificial insemination and became pregnant a dichorionic, diamniotic twin (DD twin). She was referred to the present institution for maternal-fetal care. At 30 weeks of gestation, she presented with reduced fetal movements. Transabdominal sonography showed an increased fetal abdominal circumference and a markedly dilated stomach (Figure 1-A) and bowel loops and bowel wall luminance increment. MRI showed a stricture of fetal ileum and dilated stomach and jejunum (Figure 1-B). The fetal condition was diagnosed as meconium peritonitis or intestinal volvulus. The authors were able to confirm the fetal status by cardiotocography (CTG). The volume of the amniotic fluid of the fetus increased from 32 weeks of gestation, and amniotic fluid removal was performed twice. At 33 weeks and one day, the infant was delivered via an emergency cesarean section because of uncontrollable uterine contraction. The infant with meconium peritonitis or intestinal volvulus was female, weighed 2,558 grams, and the Apgar score was 3/4/5 at

1/5/10 minutes, respectively. Immediately after birth, her abdominal skin was reddish and had distention (Figure 2-A), and no bowel sound was heard. Initial laboratory values revealed a hemoglobin level of 12.4 g/dL, WBC level of 17,600, CRP level of 2.56 mg/dL, and platelet level of 124,000. Blood gas analysis re-

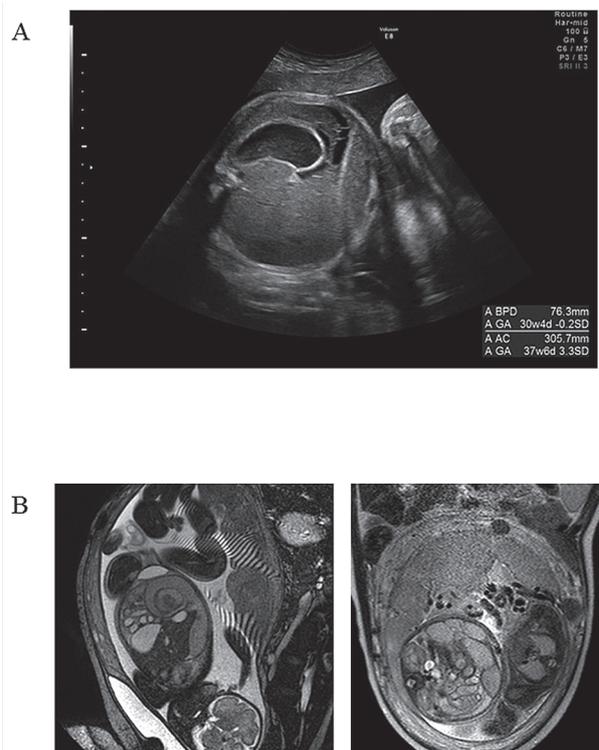


Figure 1. — (A) Fetal abdominal sonographic imaging performed at 30 weeks. Dilated stomach and bowel loops can be seen. (B) MRI examination of the fetus at 30 weeks of gestational age.

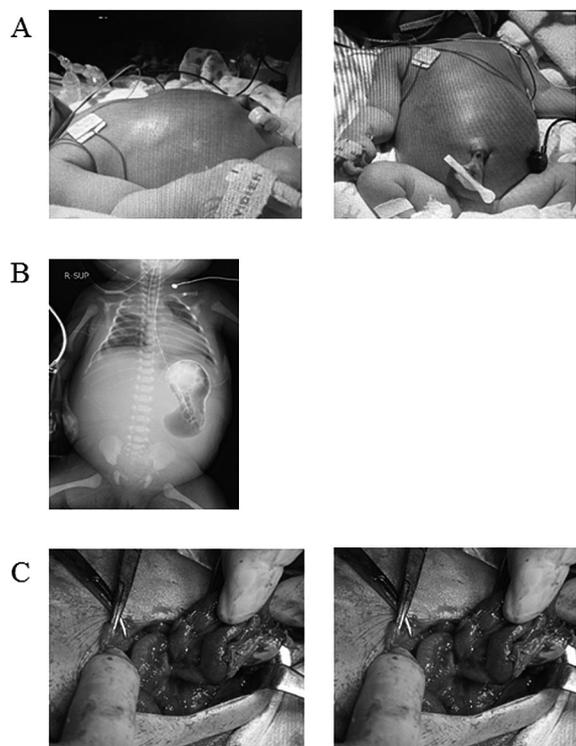


Figure 2. (A) Images immediately after birth show abdominal skin reddening and distention. (B) Abdominal X-ray image of the infant 72 hours after birth. (C) Images show volvulus with dilation and necrosis during operation.

vealed: pH 7.141, $p\text{CO}_2$ 73.7 mmHg, BE -6.4 mmol/L, and HCO_3^- 24.1 mmol/L. Her blood type was AB and Rh positive; her mother's blood type was A and Rh positive. Initial plane abdominal radiography showed no visible dilation of the stomach; however, 72 hours after birth, it showed dilation and contrast medium pooling in the stomach (Figure 2-B). Abdominal ultrasonography performed immediately after birth showed a small amount of ascites, dilated bowel, bowel peristalsis, and no whirlpool sign. She required respiratory management immediately after birth, and gastric intubation was also conducted to reduce the intestinal pressure. Her pulmonary hypertension began to improve at three days of age; thus, laparotomy was performed. Intraoperative findings included volvulus with dilation and necrosis extending from 40 cm distal to the ligaments of Treitz, which was 40 cm in length. (Figure 2-C) The volvulus had no malrotation and mesenteric defects. Resection of the necrotic bowel segment and creation of a stoma in the prenaternal anus were performed. She was fed via a tube at POD 5, and the stoma was closed at POD 90. She was discharged from the hospital at POD 171. One year later, the patient showed normal growth and development.

Discussion

Intestinal volvulus is classified into two groups: 1) idiopathic group and 2) secondary group. The secondary group has malrotations, whereas the idiopathic group has no malrotation and congenital malformations. This case belonged

to the idiopathic group. In the prenatal period, it is difficult to accurately diagnose a type or seriousness of intestinal volvulus. The primary diagnosis of fetal volvulus includes ascites, dilated stomach and bowel, and dilated bowel loops that form a convoluted mass with a typical whirlpool or snail configuration by sonography and MRI; the definite diagnosis is made through direct visualization of the twisted bowel loop or the whirlpool-like configuration of the superior mesenteric vein and mesentery around the mesenteric artery when the operation is performed after birth.

Fetal intestinal volvulus is quite rare. To the present authors' knowledge, only 31 cases have been reported for the past 20 years [1-15]. Of these, only ten cases were classified into the idiopathic group [1-10], and in twins, only one case had fetal volvulus [10]. Significant relations of fetal volvulus to preterm gestation and extrauterine volvulus were observed; fetal stress may activate the release of both adrenal and hypothalamic stress hormones, which may lead to premature uterine activity and preterm delivery [2, 11-13]. In this case, delivery by cesarean section because of undercontrolled uterine contractions, so fetal stress may lead to premature uterine activity. On the other hand, this entity can be classified into two groups on the basis of clinical findings: 1) mild volvulus group and 2) severe volvulus group [14]. In the mild volvulus group, the state of volvulus persists for a relatively long duration; therefore, it often appears as an abdominal mass polyhydramnios by fetal ultrasonography. The fetal condition seems to be well, and the clinical course at the postnatal period could be better in this group. However, in the severe volvulus group, reduced fetal movement and diminished variability on CTG are important findings for the timely performance of cesarean sections [3, 7-9, 14]. Furthermore, neonatal condition seems to be poor, and intensive care is necessary in this group. In the present case, the authors strictly and frequently monitored the fetal condition by ultrasound sonography and CTG; thus, they could decide the best time for delivery before the fetal condition could change from a mild to a severe type. The present case involved twins; thus, the authors had to consider the postnatal outcomes. They were able to follow them up carefully, continue the tocolysis treatment, and perform a cesarean section at the best time for both fetuses. If no maternal and fetal problems occur, we should wait for the fetal lungs to mature. There is only one report of a surviving fetus with intrauterine volvulus in a twin, but no report of a surviving fetus with unruptured bowel before laparotomy in a twin. If fetal volvulus occurs in multiple pregnancy, obstetricians must decide to initiate delivery when both affected and healthy fetuses are at their best condition. The present authors encountered a case of intestinal volvulus without malrotation in one fetus of twins. If a volvulus occurs in a fetal twin, we must proceed to delivery when both the impaired and healthy fetuses are in optimal condition.

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