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Case Report

A Rare Case of Fetal Meconium Peritonitis Developing Coagulopathy *In utero*

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Abstract

Hydrops fetalis in association with meconium peritonitis is a rare condition, and the mechanism underlying hydropic changes has not been fully recognized. We present a case of fetal meconium peritonitis with hydrops and coagulopathy. Clinically, the cause of fetal disseminated intravascular coagulation is considered to be a consequence of a systematic inflammatory response based on progressive, but mild fetal anemia without other apparent triggers, thrombocytopenia, elevated white blood cell count and serum C reactive-protein, hypoalbuminemia, and increased vascular permeability. The infant was born at 32 weeks of gestation and survived after postnatal multidisciplinary treatment. Our experience suggests that recognition of this rare condition will enable early diagnosis and better clinical management for fetuses with meconium peritonitis.

Keywords: Coagulopathy, fetal anemia, fetal ascites, hydrops fetalis, meconium peritonitis

Introduction

Fetal meconium peritonitis is a sterile chemical inflammatory process due to the spillage of meconium into the peritoneal cavity secondary to perforation of the fetal bowel. Fetuses diagnosed *in utero* have a good prognosis compared to fetuses diagnosed postnatally.[1,2] Meanwhile, some affected infants result in death, and systemic inflammatory response syndrome (SIRS) is thought to be a possible cause in the field of neonatology.[3]

We present an extremely rare case of meconium peritonitis that developed coagulopathy and hydrops *in utero*. These complications are considered to be a consequence of the SIRS.

Case Report

A 31-year-old primipara was referred to our hospital at 30^{+5} weeks of gestation for fetal ascites. On referral, a detailed sonography showed fetal ascites with intraperitoneal echogenic bowel [Figure 1a]. A fetal anatomic survey excluded structural anomalies. The peak systolic velocity (PSV) of the middle cerebral artery (MCA) was 59.0 cm/s (1.46 multiples of the median [MoM]). The blood type was O Rh-positive without irregular antibodies. Maternal immunoglobulin M analysis excluded TORCH infections.

The follow-up examination demonstrated fetal skin edema, a minimal pleural effusion, and bowel calcification at 31^{+2} weeks of gestation [Figure 1b]. At 31^{+5} weeks of gestation, percutaneous umbilical blood sampling (PUBS) was performed to evaluate the fetal anemia because the MCA-PSV had increased to 1.60 MoM. The results revealed mild anemia with a haemoglobin (Hb) concentration of 9.7 g/dL. The white blood cell (WBC) count was 12710/μL and the platelet count was 234,000/μL. We continued to manage the fetus expectantly; however, at 32^{+5} weeks of gestation, the MCA-PSV markedly increased to 1.68 MoM, and a fetal cardiotocogram revealed a sinusoidal pattern [Figure 2]. We suspected acute worsening fetal anemia or fetal deterioration, and thus reevaluation by PUBS was performed. The fetal Hb level had decreased to 7.2 g/dL. Of note, repeat laboratory testing revealed the following: WBC count, 16240/μL; platelet count, 49,000/μL; and C-reactive protein (CRP), 9.19 mg/dL.

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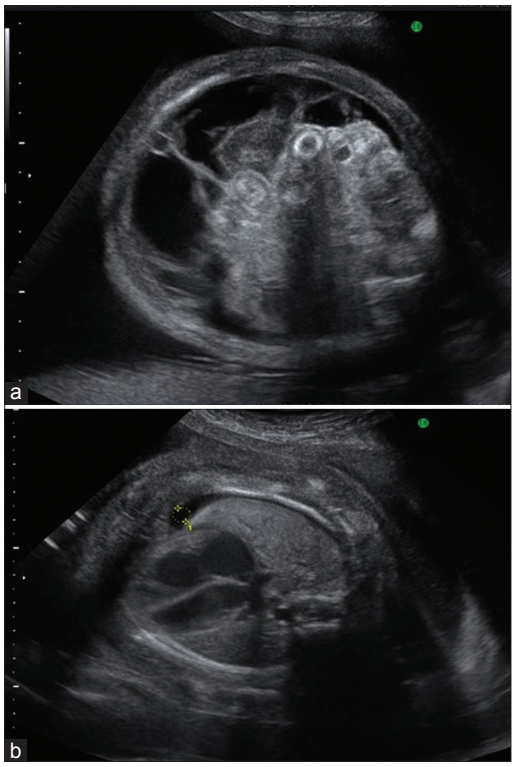

Figure 1: Ultrasound transverse section of the fetal abdomen (a) and thorax (b), showing fetal ascites with intraperitoneal echogenic bowel, a pleural effusion, and skin edema

An umbilical venous gas analysis revealed acidemia with a pH of 7.227.

A cesarean section was performed, and a male infant weighing 2432 g with Apgar scores of 6 and 6 was delivered. After aggressive resuscitation, mechanical ventilation was initiated. The initial laboratory data showed anemia (Hb, 7.6 g/dL), thrombocytopenia (platelet count, 80,000/μL), and coagulopathy (prothrombin time percentage, 72%; International Normalized Ratio, 1.19; activated partial thromboplastin time, 64.3 s; fibrinogen level, <25 mg/dL; and fibrin degradation products, 109.7 μg/mL). The serum albumin level was low (1.9 g/dL). A chest X-ray revealed mild pulmonary edema [Figure 3]. Thereafter, the thrombocytopenia and coagulopathy were treated with the administration of red blood cells, fresh frozen plasma, and a platelet transfusion. Hypoalbuminemia was treated with the administration of hypertonic albumin. After stabilization, the first surgical intervention was performed, which revealed ascites, a conglomeration of bowel covered with fibrin, and the suspected site of the perforated bowel. Due to intra-abdominal inflammation and the difficulty in delineating viable bowel, intra-abdominal drainage was performed to mitigate systemic inflammation. At 36 days of life, a temporary ileostomy was performed during the second surgical procedure. In the last 6 months of follow-up, this infant demonstrated normal growth without any gastroenterologic complications.

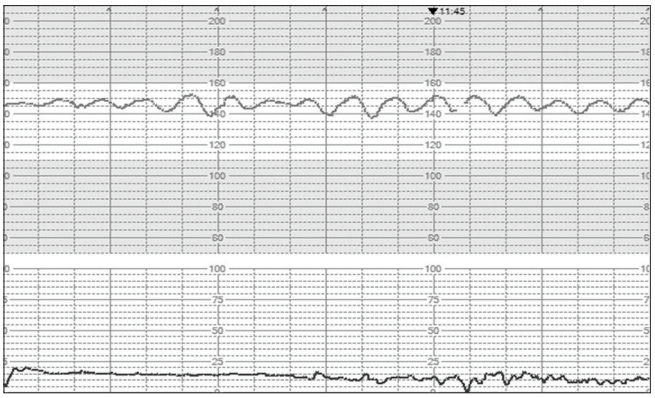

Figure 2: Cardiotocogram showing sinusoidal pattern at 32 weeks and 5 days of gestation

Discussion

Although meconium peritonitis was considered to be a severe disease with a high mortality rate in the past,[4] recent overall survival rate approaches 90% due to the improvements in prenatal diagnosis and postnatal management.[1,2,5,6] Nonetheless, some infants die during the neonatal period. Reported causes of neonatal death include prematurity, necrotizing enterocolitis, and postoperative sepsis.[2,5] Furthermore, the SIRS has been reported as a possible complication,[3] but there are no previous reports of this complication during the fetal period.

Clinically, we speculate that the causes of fetal coagulopathy and acidemia in our case were consequences of the SIRS. In support of this opinion, we found progressive fetal anemia without other apparent triggers, thrombocytopenia subsequent to the occurrence of fetal ascites, elevated WBC count and serum CRP, hypoalbuminemia, and increased vascular permeability. Previously, Kanamori *et al*.[3] showed that the concentrations of several cytokines and a chemokine from cystic or ascitic fluid of some neonates with meconium peritonitis immediately after birth were very high, suggesting that these patients had already undergone an anti-inflammatory response at the time of birth. Furthermore, a role of tumor necrosis factor-alpha (TNF-α) in the inflammatory reaction in meconium peritonitis has also been suggested by Lally *et al*.[7] Based on the results provided by Lally *et al*., Kanamori *et al*.[3] have presented the following speculations. In short, TNF-α secreted by peritoneal macrophages induces an inflammatory reaction in the peritoneal cavity, and such an inflammatory reaction can lead to pseudocyst formation. Interleukin (IL)-6 and IL-8 are produced by activated macrophages and neutrophils recruited by inflammation evoked by TNF-α; however, pseudocyst formation can serve as a further brake and contribute to prevent the overflow of cytokines into the circulation. Viewed in this way, a lack of pseudocyst formation might be hazard-related for the SIRS in the current case.

Hydrops in association with meconium peritonitis is a rare condition, and the mechanism underlying hydropic changes is not understood. In 2000, Kamata *et al*. investigated 20 cases of fetal meconium peritonitis and speculated that the cause

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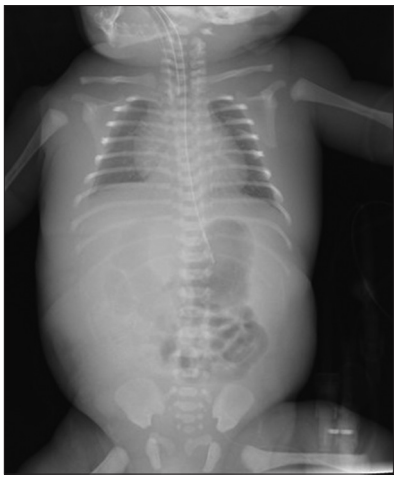

Figure 3: Chest X-ray showing mild pulmonary edema with small bilateral pleural effusions

of hydropic changes might result from the effect of a giant pseudocyst and massive meconium ascites on fetal circulatory dynamics.[1] However, most fetuses with meconium peritonitis and marked abdominal distension do not develop hydrops. Indeed, in the current case, the ascites gradually decreased, and no huge pseudocysts were detected during the fetal period. Although our knowledge about the hydropic change in fetuses with meconium peritonitis is still limited, the inflammatory response syndrome may be a frequent occurrence.

Conclusion

The SIRS can occur in patients with meconium peritonitis, even during fetal life, and trigger coagulopathy and fetal acidemia *in utero*. The fetus has relatively immature peritoneal defense mechanisms, and anticipatory recognition of this rare condition will enable early diagnosis and better clinical management for fetuses with meconium peritonitis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initial will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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None.

Conflicts of interest

There are no conflicts of interest.

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