Case Report

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Tieguanyin Tea Like-Colored Amniotic Fluid Can be the Earliest Diagnostic Sign of Food Protein-Induced Enterocolitis Syndrome – 2 Case Reports

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ABSTRACT

Food Protein-Induced Enterocolitis Syndrome (FPIES) is a non-immunoglobulin E-mediated disorder. We report two rare cases of suspected FPIES sensitized and developed in utero. The one infant presented with hematochezia and abdominal distention. She was noted to have a dilated gastric bubble on fetal ultrasound. The other infant presented with tarry stools, old hematemesis, and abdominal distention. An allergen-specific lymphocyte stimulation test was performed, both infants were positive for cow's milk protein. Notably, both infants showed translucent brownish amniotic fluid appearing similar in color to Tieguanyin tea, which suggested that the infants had vomited the contents of the gastric hemorrhage in utero. When characteristically appearing such amniotic fluid with no finding of meconium staining is present, FPIES should be suspected.

Keywords: Food Protein-Induced Enterocolitis Syndrome; Allergen-Specific Lymphocyte Stimulation Test; Abdominal Distension; Fetal Sensitization; Brownish Amniotic Fluid

Introduction

Food Protein-Induced Enterocolitis Syndrome (FPIES) is a non-immunoglobulin E (IgE)-mediated disorder usually caused by antigen ingestion after birth, with symptoms appearing during the neonatal period and infancy [1]. Cell-mediated hypersensitivity is assumed to be the major mechanism of FPIES [2], although the detailed mechanism has not been clarified. To our knowledge, there is only one report of symptoms appearing in the fetal period [3]. We report two rare cases of newborns with suspected FPIES developed following gastric hemorrhage in utero. They were presented with Tieguanyin tea-colored amniotic fluid and were suspected as having FPIES based on a positive reaction to an Allergen-Specific Lymphocyte Stimulation Test (ALST). The parents of these infants were informed of the study design, and their written informed consents were obtained for submission of this case report.

Case Report

Case 1

The patient was a female neonate born at 35 weeks of gestation by normal vaginal delivery and weighed 2242 g. Fetal ultrasound at 34 weeks of gestation showed abnormal dilatation of a gastric bubble and increased echo brightness of the gastric contents (Figure 1a). The amniotic fluid presented as a translucent dark reddish-brown color similar to that of Tieguanyin tea and was not stained by meconium.

The infant was admitted because of frequent hematochezia before her first formula, and the gastric fluids were almost the same consistency as the amniotic fluid (Figure 1b). Placental pathology showed stage II chorioamnionitis of Blanc's classification, but no evidence of premature placental abruption or diffuse chorioamniotic hemosiderosis. Laboratory results showed a total white blood cell count of $30.9\times10^3/\mu L$ with 14.0% eosinophils. C-reactive protein was 0.04 mg/dL, and liver function and other blood chemistries were normal. PIVKA-II was normal at 71 mAU/mL and vitamin K deficiency was ruled out. She started on extensively hydrolyzed formula at 2 days of age and progressed without symptoms. The patient's feeding quantity was carefully increased, weight gain was good, and the percentage of eosinophils in her blood gradually decreased, eventually to 2.0%. Although her mother consumed adequate amounts of dairy products during her pregnancy, starting on day 27, a small amount of breast milk without dairy products was added. An ALST performed at 28 days of age showed the stimulation indices of β casein and κ casein to be elevated, and we preliminary diagnosed her as having FPIES. She was referred to an allergist and an oral food challenge will be done in the future.

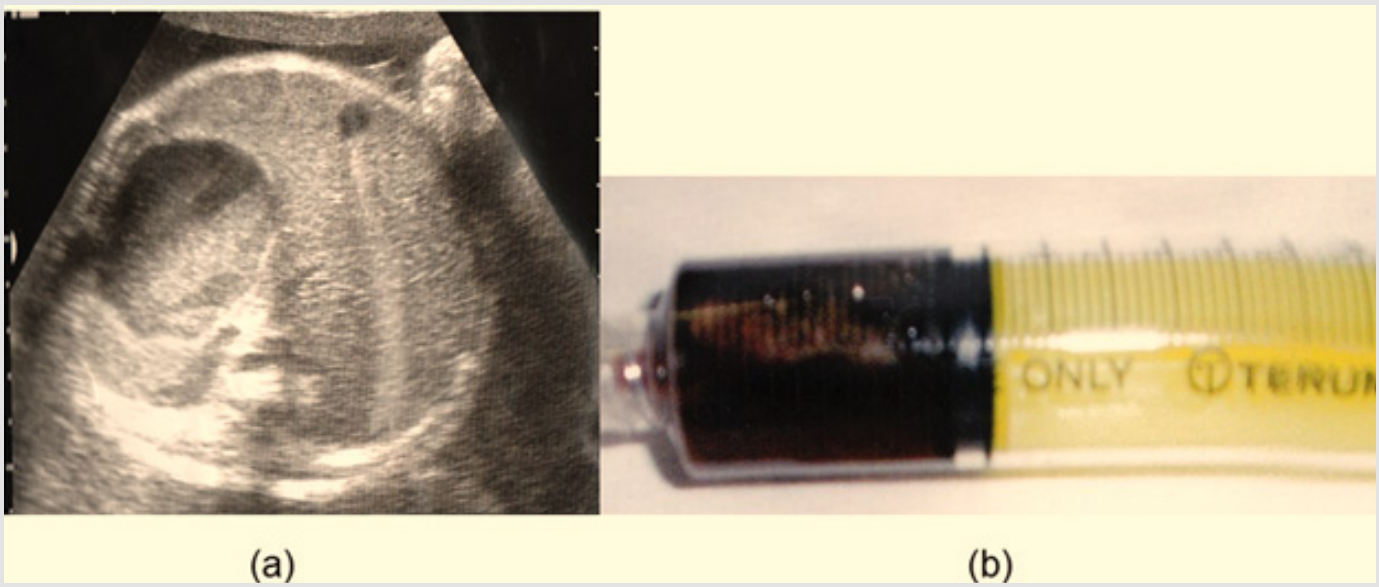

Figure 1: Findings of Case 1:

(a) Fetal ultrasound at 34 weeks of gestation showed abnormal dilatation of a gastric bubble and increased echo brightness of the gastric contents.

(b) Photograph of the gastric fluid showing the same properties as those of the amniotic fluid.

Case 2

The patient was a female neonate born at 37 weeks of gestation by cesarean section from a multipara woman and weighed 2670 g. The pregnancy progressed steadily, and fetal ultrasound did not indicate any abnormalities including oligoamniotic fluid. The amniotic fluid presented with a translucent color similar to that of Tieguanyin tea, and the skin color stain was almost the same as that of the amniotic fluid (Figure 2). She had marked abdominal distention from birth and was admitted because of grossly tarry stools and vomiting with old blood after her first formula. The contents aspirated through the inserted gastric tube were uniform brownish color, and abdominal x-ray showed intestinal dilatation. Placental pathology showed stage I chorioamnionitis of Blanc's classification only. Laboratory results showed a total white blood cell count of $23.0\times10^3/\mu L$ with 6.0% eosinophils, C-reactive protein was 0.60 mg/dL, and other laboratory test results including PIVKA-II were normal. Then, suspecting FPIES to cow's milk protein, we were sure her mother was not consuming large amounts of dairy product, started enteral feeding with a small amount of breast milk from day 1 of age. Despite ingesting increased quantities of non-antigen-released breast milk or extensively hydrolyzed formula, she had no vomiting or tarry or bloody stools, but her abdominal distention persisted. Allergen-specific IgE antibodies in cow's milk-based formula, casein, β-lactoglobulin, and α-lactalbumin, were all negative. An ALST performed at 6 days of age showed the stimulation indices of lactoferrin and α-lactalbumin to be elevated, and we preliminary diagnosed her with FPIES and referred her to an allergist.

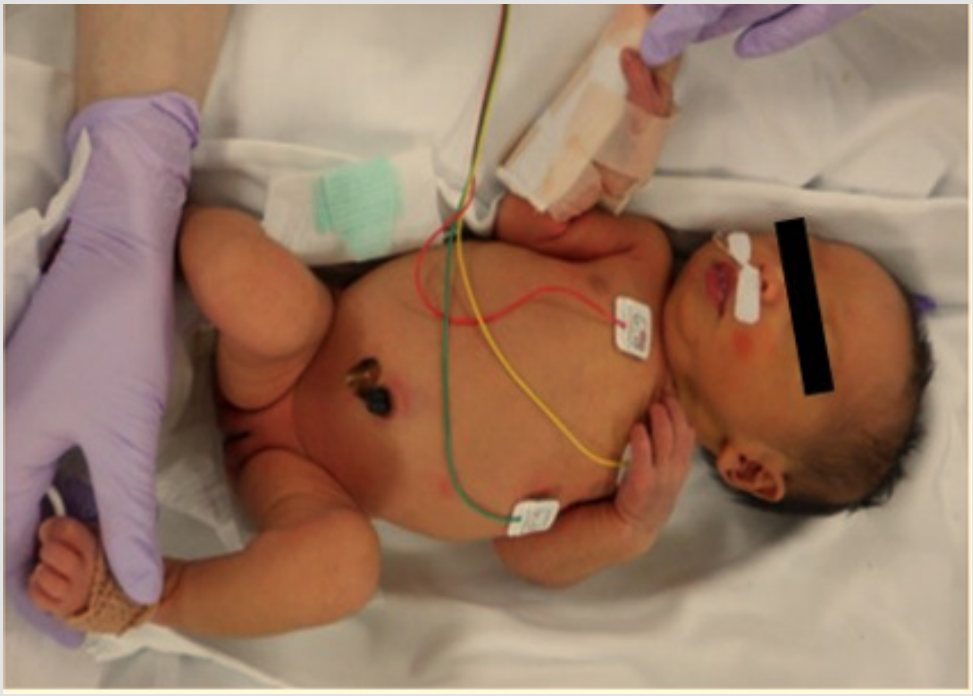

Figure 2: Findings of Case 2: The skin color of the infant was almost the same color as the amniotic fluid. This photograph was taken at day 3 of age. Thus, the skin color had faded, and the infant appeared jaundiced, but total bilirubin values were not elevated.

Discussion

FPIES is a non-IgE-mediated gastrointestinal food allergy usually caused by antigen ingestion with symptoms such as vomiting, diarrhea, and hematochezia appearing during the neonatal period and infancy1. Neither of the two cases had vomiting and did not exactly meet the criteria, but hematochezia is ranked as one of the major FPIES primary symptoms. Cell-mediated hypersensitivity is assumed to be the major mechanism of FPIES [2,4]. However, the detailed mechanism remains to be clarified. Although leukocytosis with eosinophils and thrombocytosis are often present, laboratory findings cannot confirm the diagnosis of FPIES alone[1]. Oral Food Challenge is necessary (OFC) to diagnose FPIES, but it may cause a severe reaction; thus, diagnosis is most often based on clinical suspicion. Considering the risk of anaphylaxis, OFC was not performed in the neonatal period in our cases. Since ALST has been shown to be useful in the diagnosis of FPIES for cow's milk protein, we made a provisional diagnosis with the ALST results [5].

The detection of allergen in the amniotic fluid and fetal blood has provided evidence of transplacental transfer [6,7], and there have been a few reported cases of FPIES sensitized in utero [3,8,9]. Ichimura et al. reported a case that developed in utero with intestinal dilatation on fetal ultrasound but did not describe the amniotic fluid [3]. Another differential diagnosis for neonatal hematochezia includes neonatal transient eosinophilic colitis, and there have been case reports suspected in utero onset [10]. However, both present cases showed translucent brownish amniotic fluid with a color similar to Tieguanyin tea that was obviously different from amniotic fluid stained by meconium. The point is that hematochezia was not thought to have been in utero. The amniotic fluid and gastric contents were consistent with each other on visual inspection, suggesting that the infants had vomited the contents of the gastric hemorrhage in utero. Especially, glitter in the stomach on fetal ultrasound in Case 1 was considered a sign of blood contamination, not just gastric juice. Three limitations of this report must be considered. First, cow's milk protein could not be detected in amniotic fluid. And second, ALST was only proved by cow's milk protein. Finally, the results of OFC are in the future.

Conclusion

In conclusion, FPIES must be suspected in the presence of characteristic translucent, brownish-colored amniotic fluid and abdominal distention from birth. Early diagnosis may allow for intervention before serious complications appear.

Funding

None.

Consent

The authors confirm that consent for submission and publication of this case report including images and associated text has been obtained from the patient's guardians. In addition, approval was obtained from the hospital's Ethics Committee.

Conflict of Interest

None declared.

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Authorship

K.I. and T.N. wrote the manuscript. H.T., M.K, D.H, T.N, and E.F. collected and analyzed the date. All the authors read and approved the final manuscript.

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