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Case Report

Early Onset Necrotizing Enterocolitis in Full Term Newborn Associated with Placental Chorangiosis: A Case Report

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Abstract

Necrotizing Enterocolitis (NEC) is a serious inflammatory disease that affects intestinal wall in premature infants. Full-term infants account for approximately 10% of cases. NEC can cause significant morbidity and mortality in neonates.

Case presentation: A full - term baby girl was born at 38 weeks and 2 days of gestation via vaginal delivery to gravida 4 para 3 woman. Pregnancy was complicated with diabetes, obesity and maternal anemia. Placental pathology showed small placenta and increased vascularity in chorionic villi that consistent with chorangiosis. The infant developed significant feeding intolerance after feeding of breast milk and early onset NEC at 24 hours of age. Abdominal X ray showed abdominal distension and pneumatosis intestinalis. We are the first to report the association of placental chorangiosis and early onset NEC in full-term. Placental chorangiosis causes perinatal hypoperfusion that decreases fetal mesenteric blood flow and results in early onset NEC.

Keywords: NEC; Placenta; Chorangiosis; Morbidity

Introduction

Placental chorangiosis is a rare condition of placental hypervascularity. Placental chorangiosis is a vascular change involving the terminal chorionic villi in the placenta. It is characterized by more than 10 capillaries in more than 10 terminal chorionic villi in several areas of the placenta [1]. Placental chorangiosis is rarely occurs in normal pregnancy. It has been associated with multiple comorbidities associated pregnancy such as smoking, multiple pregnancies, maternal anemia, preeclampsia, and diabetes mellitus. Placental chorangiosis is associated with higher incidences of perinatal and neonatal morbidity and mortality [2]. Necrotizing enterocolitis is a primarily disease of gastrointestinal tract of premature infants that results in inflammation and bacterial invasion of bowel wall [2]. Necrotizing enterocolitis is inversely correlates with gestational age at birth. There is also a correlation between gestational age at birth and length of interval between birth and onset of NEC. The lower the gestational age at birth, the longer the interval till the onset of NEC. NEC is a common cause of mortality and morbidity in premature infants [3]. The incidence of NEC in full term infants is uncommon. There have been only few studies of full-tern infants with NEC. Of note, multiple risk factors have been associated with increase the risk of development of NEC in full-term infants such as congenital heart diseases, perinatal hypoxia, and indomethacin medication [4].

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Case Presentation

A full-term baby girl was born at 38 weeks and 2 days of gestation *via* vaginal delivery. The baby was born to a 40-year-old, gravida 4, para 3, African American woman. Pregnancy was complicated with obesity with Body Mass Index (BMI) of 34, un-controlled gestational diabetes recurrent Escherichia coli urinary tract infection and Group B *streptococci* positive vaginal-rectal swab. The amniotic membranes ruptured 8 hours before delivery for clear fluid. Mom was treated with three doses of penicillin prior to delivery. In addition, pregnancy was complicated with of iron deficiency anemia, her hemoglobin at 28 weeks of gestation was 7.3 mg/dl and her ferritin level was 3.4 ng/ml for which she received intravenous iron therapy in addition to oral ferrous sulphate. Routine prenatal ultrasound was performed at 23 weeks of gestation and demonstrated thickness of fetal cardiac ventricular walls.

The pathological exam of the placenta revealed a small placenta weighted 440 gm with mature chorionic villi, increased vascularity suggestive of chorangiosis, focal increase in syncytial knots suggestive of advanced maturation, no evidence of funisitis, vasculitis or chorioamnionitis (Figure 1).

The baby did not require active resuscitation at birth. Apgar scores were 9 and 9 at one and five minutes, respectively. Birth Weight (BW) was 2700 gm. Despite, the infant was vitally stable after delivery and her clinical examination was normal, she was admitted to the NICU for postnatally echocardiographic assessment. Echocardiogram examination upon admission showed patent foramen oval, small patent ductus arteriosus, flattened interventricular septal motion, mild right ventricular hypertrophy, and mild dilatation of the right

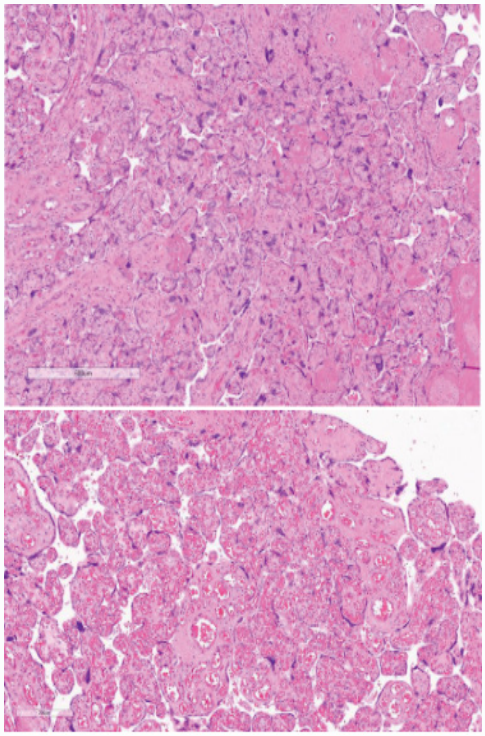

Figure 1: Pathological findings of the placenta that is consistent with placental chorangiosis. Third trimester placenta with mature chorionic villi showing increased vascularity consistent with chorangiosis. Chorionic villi showed more than 10 capillaries in at least 10 terminal villi in high-power microscopic fields. The chorionic villi show increase in syncytial knots by approximately 30% that is suggestive of advanced maturation.

ventricle with normal systolic function of the right ventricle, and normal left ventricular size and systolic function. Breast milk oral feeding on demand was started based on the feeding protocol. At age of 12 hours our patient had feeding intolerance. She had multiple significant vomiting associated with breast feeding, the vomitus was also curdled milk, her vital signs and physical examination were within normal limits for age at that time. Basic metabolic panel and complete blood count were normal at age of 24 hours. At age of 47 hours the infant had significant bloody stool. Abdominal examination showed a mildly distended and tender abdomen. An abdominal X-ray was performed and showed pneumatosis intestinalis (Figure 2). The feeding was completely stopped, baby was kept NPO on IV fluids, and antibiotics including piperacillin-tazobactam plus gentamicin for 14 days were started after sending blood cultures. Surgical services were consulted. Serial abdomen radiographs were obtained and by day 3 there was no evidence of pneumatosis intestinalis. The infant was in NPO for 10 days, and antibiotics were given for 14 days. Gradual advancement of breast milk feeding was started after completed 10 days of NPO, and the infant reached full feeds by Day of Life (DOL) 25.

Outcomes and Follow Up

The patient was discharged home at DOL 31 on full feeding and she was seen in the outpatient clinic where she continues to have adequate weight gain with good elimination.

Discussion

This case study is the first to report the association of placental chorangiosis and development of early onset NEC in healthy, full-

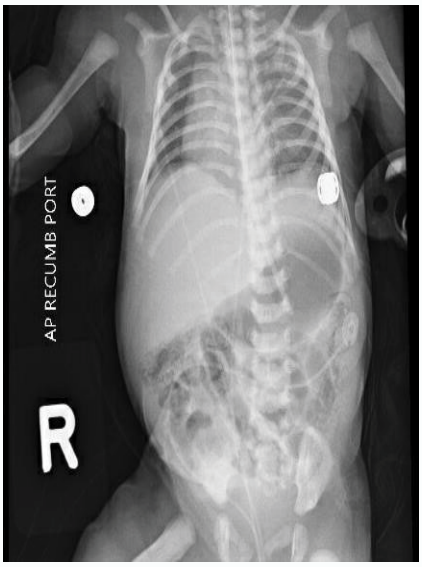

Figure 2: Abdominal radiographic findings of pneumatosis intestinalis.

term neonates. Necrotizing enterocolitis is common gastrointestinal emergency in preterm infants while development of NEC in healthy full-term infants is unlikely. Multiple risk factors have been associated with NEC in full-term neonates such as perinatal hypoxia and mesenteric hypoperfusion. The early onset of NEC both with and without any major risk factors in full term infants might suggest a common mechanism related to a perinatal ischemic insult. The exact pathogenesis of placental chorangiosis is still unclear. Chronic maternal tissue hypoxia and hypoperfusion is associated with development of placental chorangiosis [5]. Notably, tissue hypoxemia and hypoperfusion stimulate expression of vascular growth factors and activate proliferation of connective tissues that consequently, initiate neovascularization in the placental tissues [6]. Suzuki et al. [7] reported an association between maternal blood oxygen saturation in intervillous gaps and the development of chorangiosis. Low efficiency of oxygen transfer from maternal to fetal circulation causes vascular remodeling in adaptation to low oxygen supply, resulting in neovascularization and development of placental chorangiosis [7]. Our case is the first to assess the association between placental chorangiosis and development of early onset NEC in full-term neonates. This association could be explained by placental hypoperfusion and chronic hypoxic placental injury, result in reduced intake of oxygen from the intervillous space. Placental chorangiosis compromises the fetal intestinal blood supply. Mesenteric hypoperfusion is the common denominator in the pathogenesis of NEC. Recently, a prospective study reported the mesenteric hypoperfusion and increased mesenteric blood resistance is associated with increased the incidence of NEC and feeding intolerance in neonates [8]. The classic presentation of NEC is feeding intolerance including vomiting, abdominal distention, bloody stools, and rapid progression to overwhelming sepsis [9]. However, many patients could have an indulgent course of abdominal distention and bloody stools which resolve quickly with bowel rest, broad spectrum antibiotics, and volume resuscitation alone [9]. Several patients may progress rapidly from milder to severe forms of NEC resulting in multi-system organ failure and death within hours of initial presentation [10,11]. 'NEC totalis', which often reflects the presence of complete intestinal necrosis, is associated with nearly 100% mortality [12]. Intraoperatively, patchy areas of intestinal

necrosis typically involving the jejunum and ileum would be seen, accompanied by intra-abdominal fluid and the presence of air within the wall of the bowel (pneumatosis intestinalis) [13]. Resection of the dead intestinal segments results in clinical improvement and is successful in 60% to 70% of cases [14].

Clinical examination and laboratory tests are not specific for NEC diagnosis [15]. Despite the lower sensitivity of radiological findings, diagnosis of NEC can be confirmed when pathognomonic signs such as portal venous gas or pneumatosis intestinalis are present [16]. The most sensitive finding is bowel wall dilation. Despite abdominal dilation and dilated bowel loops are in greater than 90% of cases, is not specific to NEC [17,18]. Abdominal ultrasound is an effective assessment tool to predict NEC, as it helps to assess intestinal peristalsis, vascular perfusion, bowel wall thickening and abdominal fluid [18]. Our case reported that the pregnancy was complicated with uncontrolled diabetes, obesity and maternal hypotension. The association of chorangiosis with pregnancy associated comorbidities and placental pathological conditions such as preeclampsia, diabetes mellitus, drug ingestion, urinary tract infections, placental abruption, villitis, and umbilical cord anomalies were also previously reported [19]. Multiple studies reported the association between charangoists and intrauterine growth restriction [20]. Chorangiosis increases the risk of neonatal morbidity and mortality. The majority of infants born to women diagnosed with placental chorangiosis have low Apgar at 1 and 5 minutes, low birth weight, metabolic acidosis, fetal anomaly, and increased risk of mortality [21]. This case report highlighted the association between chorangiosis and early onset NEC in full-term infants. Full-term infants delivered to women diagnosed with placental chorangiosis should be closely evaluated for any signs of feeding intolerance after initiation of feeding to predict early onset NEC and prevent complications.

Conclusion

Placental chorangiosis is uncommon placental pathology. Placental chorangiosis is associated with development of early onset of necrotizing enterocolitis in full-term infants. Prospective randomized studies are in need to explore this association.

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