CASE REPORT

Fetus with Lateral Gastrochisis Born to a Celiac Mother

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ABSTRACT

Gastrochisis is a rare anterior abdominal wall defect which develops secondary to an early embryonic vascular defect. We report here a rare site of gastrochisis involving the posterior aspect of lateral abdominal wall on the right side, born to a celiac mother. There is no reported literature on any association between gastrochisis and celiac disease. We propose here possible role of palmitic acid pathway in this index case.

Key words: Gastrochisis; Lateral gastrochisis; Celiac disease

INTRODUCTION

Gastrochisis has been traditionally thought to be caused due to early gestation vascular accident. The defect spares the midline and does not involve the umbilicus; it is usually paramedian and is more common on the right side. We report here a rare site of gastrochisis involving the posterior aspect of lateral abdominal wall on the right side in a fetus, born to a celiac mother.

CASE REPORT

Baby X was born to a 28-year-old primiparous mother at 27 (+5) weeks of gestation by vaginal delivery. Mother was a biopsy-proven case of celiac disease on dietary restrictions and was a booked case at another hospital. Level II ultrasound done at 22-week gestation showed severe oligohydramnios with absent diastolic flow; no gross congenital malformation was detected. Unfortunately, no subsequent visits were made to the treating hospital and mother went into spontaneous labor about 6 weeks later to give birth to a macerated stillborn baby. The birth weight was 600 g. Physical examination revealed herniation of the loops of small intestine from a defect in the posterior aspect of the right lateral wall of abdomen (Figure 1). No membrane or sac was present over the herniating intestinal loops. The umbilicus was normal in position. Anal opening and external genitalia were normal with no other gross congenital anomaly.

DISCUSSION

Gastrochisis is an important anterior abdominal wall birth defect that has shown an increased prevalence in recent decades. Factors possibly increasing the risk for gastrochisis include maternal age <20 years, cigarette smoking, alcohol and exposure to aspirin, cocaine, amphetamines, or the agricultural chemical atrazine [1-3]. We are not aware of any reported association of gastrochisis with celiac disease.

The gastrochisis defect does not have any covering; however, exposure to amniotic fluid elicits an inflammatory response which may give the appearance of a membrane. The normal position of the umbilicus, separate from the defect, reliably differentiates it from an omphalocele. Various theories regarding the development of gastrochisis include abnormal involution of the right umbilical vein, failure of mesoderm to form body wall, rupture of amnion around umbilical ring, and disruption of the right vitelline artery.

Lubinsky, in 2012, hypothesized estrogen-induced thrombosis for pathogenesis for gastrochisis. He rationalized that high maternal estrogen during early pregnancy causes thrombosis of fetal vessels; pal-
IUGR in these fetuses. IUGR may be overestimated if formulae relying on abdominal circumference are used. In fetuses with abdominal wall defects, special formulae to determine fetal weight, which are based on biparietal diameter, occipitofrontal diameter, and femur length should be used [9].

The fertility period in celiac disease is shortened and celiac women on a normal diet suffer from spontaneous abortions and other complications of pregnancy more often than those maintaining a gluten-free diet [10]. Our case was born stillbirth, may be affected by non-compliance of gluten-free diet or late diagnosis of celiac disease.

To conclude, we present this case with very unusual location of defect in the posterior aspect of lateral abdominal wall. This site of defect cannot be explained with vascular perfusion deficit. We hypothesize the external disruption as cause for this unusual site. Furthermore, its presence in celiac mother may open window to future research. We hypothesize the likelihood role of palmitic acid in this index case.

REFERENCES