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LETTER TO THE EDITOR

A Rare Occurrence of Infantile Hypertrophic Pyloric Stenosis following a Primary Surgical Closure of Gastroschisis in a Neonate

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DEAR SIR

A female infant, twin B, was born to a 30-year-old, gravida 6 para 5 mother at a gestational age of 35 weeks with a birth weight of 2198 grams. Pregnancy was complicated by a late prenatal care, dichorionic-diamniotic twin gestation, discordant twin size and prenatally diagnosed gastroschisis in infant twin B. These infants were born by caesarean section secondary to poor biophysical profile. Gastroschisis with eviscerated intestinal loops was grossly visible on physical examination. A complete reduction of the eviscerated bowels with a primary repair of the gastroschisis was performed on the first day. Post-operative period was uneventful with no acute complications. Conservative enteral feeding regimen with donor breast milk was initiated on postnatal day 16. After having tolerated the initial advancement of enteral feeds, the infant developed non-bilious, non-projectile emesis on postnatal day 20. The radiographic evaluation of the abdomen showed a non-specific dilatation of small bowel loops without any obvious radiographic signs of intestinal obstruction. Initially, intestinal dysmotility and gastroesophageal reflux were considered as possible causes of these symptoms. Subsequently, the caring team reduced the amount of enteral feeds and the rate of gastric infusion. Despite the adjustments in the feeding regimens, emesis persisted. A water soluble contrast enema was performed which showed no evidence of distal small bowel and large bowel obstruction. Next, feeding regimen with an amino acid based formula was initiated secondary to a family history of severe milk protein intolerance in the sibling. However, it did not improve infant's emesis. Oral erythromycin was also initiated for possible dysmotility, but stopped after three days of treatment as there was no improvement in the feeding intolerance. As emesis persisted, a sonographic evaluation of the pyloric region of the abdomen was performed on postnatal day 34. The wall thickness of pylorus measured approximately 0.36 cm, and pyloric channel length measured 1.79 cm. The stomach appeared distended with no gastric contents passing through the pylorus. All these sonographic findings were consistent with infantile hypertrophic pyloric stenosis (IHPS). Open pyloromyotomy was performed. The infant had uneventful recovery and discharged on full feeds.

Since both gastroschisis and IHPS are not common gastrointestinal pathologies during the infancy, the co-occurrence of both conditions is exceptionally rare. To the best of our

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knowledge, only three cases [1-3] have been reported so far in the literature. All three cases followed a similar pattern of development of IHPS and presented with persistent emesis and feeding intolerance at approximately three weeks of life following the primary closure of gastroschisis soon after birth. In all cases, emesis resolved with open pyloromyotomy. In the index case, although the age of onset of symptoms and their characteristics were typical of IHPS, the diagnosis of IHPS was made after 14 days of onset of these symptoms. This delay in identifying the cause of feeding intolerance and emesis was not initially expected as IHPS is a rare occurrence in infants with gastroschisis and as feeding intolerance is commonly seen in infants with gastroschisis. In addition, due to the unremarkable abdominal physical examination findings and absence of typical electrolyte abnormalities described in association with IHPS, initial suspicion for IHPS as a cause of feeding intolerance in the index case was low. However, after ruling out other common causes of feeding intolerances among infants with surgically repaired gastroschisis, the presence of IHPS became highly suspicious. Subsequently, the diagnosis and the treatment of IHPS resulted in the prevention of multiple unnecessary diagnostic evaluations, potentially harmful empirical therapies and prolonged hospitalization.

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