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Poor oral intake in a late preterm twin – usual symptom with an unusual diagnosis

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ABSTRACT

Background: At three weeks of age, a previous 34 weeks' gestation male infant (twin A) was transferred to our regional perinatal center (RPC) with complaints of poor oral feeding and intermittent tachypnea. Twin B was discharged at 37 weeks with an uneventful course.
Case: Twin A briefly required respiratory support but continued to have difficulty transitioning from gavage to oral feeding. Initially, his inability to feed orally was thought to be secondary to nasal congestion and prematurity, but with worsening respiratory distress he was transferred for further evaluation and management.
Diagnosis & Conclusion: On admission to RPC, the examination prompted a cardiac assessment which revealed a large aortic-pulmonary window type II. After surgery, the infant quickly improved and went home on-demand oral feeds. Cardiac lesions are more common in monochorionic twins but should be suspected in dichorionic twins especially if one twin has a normal course.

Introduction

Late preterm infants (34 0/7 – 36 6/7-weeks' gestation) are at higher risk of respiratory and feeding issues compared to term infants.[1] Poor oral intake is not uncommon in late preterm neonates admitted to neonatal intensive care unit.[1] Although the non-nutritive suck matures into a nutritive suck at 34 weeks, these infants may still have poor suck-swallow-breathing coordination resulting in intermittent respiratory distress and feeding difficulty.[2] Feeding competency is a major criterion for discharging these infants home.[1] Multiple gestation late preterm infants may have a different course compared to a singleton. Feed-related issues in these infants often come to light when they are above 37 weeks, and difficulty exists transitioning from gavage to oral feeding. Failure to transition to oral feeds in one twin while the other is normal, especially when they are term corrected, should prompt a clinician to consider other causes of feeding intolerance rather than prematurity. Besides feeding evaluation, infection, respiratory pathology, cardiac and central nervous system related issues should be considered and ruled out.

Case

A 3-week old male infant twin A born at 34-weeks gestational age was transferred to our regional perinatal center (RPC) due to poor oral intake and intermittent tachypnea with oral feeding attempts.

He was born to a 31-year-old primigravida with spontaneous dichorionic diamniotic twin gestation. A prenatal sonogram showed varix of the umbilical cord and a subsequent fetal echocardiogram was reported normal. The twins were delivered via C-section secondary to pre-eclampsia and admitted to a level II neonatal intensive care unit (NICU) for further management.

Twin A was an asymmetric small for gestational age (SGA) infant with a birth weight of 2.1 kg (6.2%). At birth, the exam was significant for nasal deviation, suspected craniosynostosis and required minimal respiratory support. He was weaned off nasal cannula to room air on day 2. Gavage feeds were introduced on day 2, and parenteral nutrition was weaned off in a week. Attempts at oral feeding were associated with intermittent tachypnea. He was continued on gavage feeds until 37 weeks with adequate weight gain. A complete blood count with differential, chest x-ray and a head ultrasound done during the first three weeks were within normal limits.

Twin B who was also an asymmetric SGA had an unremarkable course, tolerated oral on demand feeds and was discharged home at 37 weeks' gestational age. Multiple gestation complicated by maternal pre-eclampsia were considered to be the cause of the twins being SGA.

Abbreviations: RPC, regional perinatal center; APW, aortopulmonary window.

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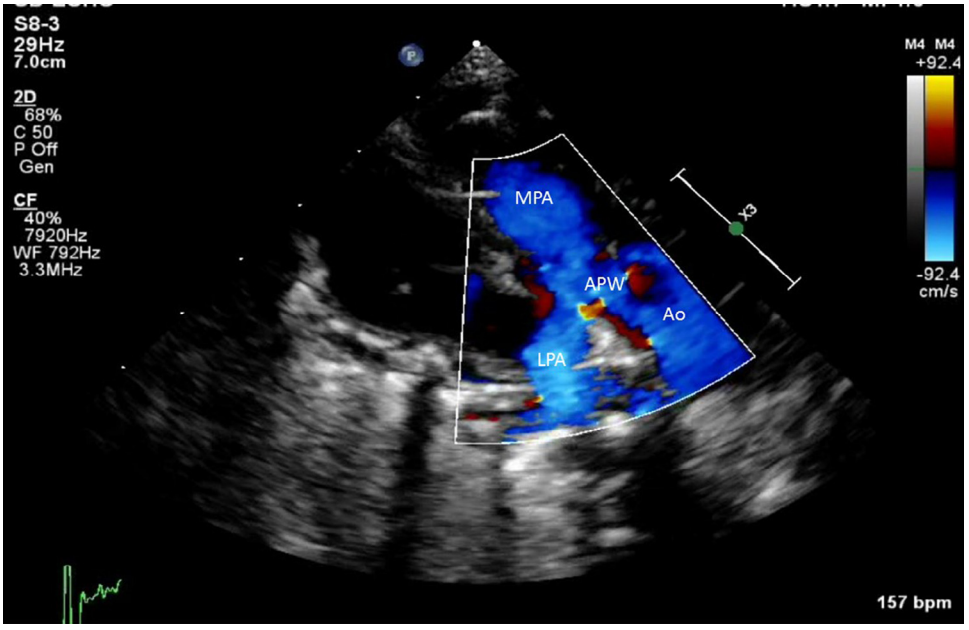

Fig. 1. Cardiac echo – Long Axis Parasternal view. MPA – main pulmonary artery, Ao – ascending aorta, APW – Aortopulmonary window, LPA – left pulmonary artery.

The worsening of symptoms in Twin A prompted a transfer to the RPC for further evaluation. Infant's weight on admission to RPC on postnatal day 21 was 2.8 kg. The exam was remarkable for tachypnea which worsened on crying and feeding. He had a strong cry with no stridor or added breath sounds. A systolic ejection murmur was auscultated over the left parasternal border with prominent second heart sound. Peripheral pulses were palpable with good perfusion. His neurological exam was within normal limits for gestational age with normal head ultrasound. A chest x-ray exam was done which showed cardiomegaly and increased pulmonary vascularity. An echocardiogram (Fig. 1) and CT chest with contrast were performed (Fig. 2).

The cardiac exam along with echocardiogram and CT chest with IV contrast confirmed the diagnosis of aortic-pulmonary window type II.

Discussion

Aortopulmonary window (APW) is a defect involving the great vessels where there is a communication between the ascending aorta and the main pulmonary trunk (Fig. 2).[3] First described in 1800's, APW is a rare condition that comprises about 0.1–0.3% of congenital heart diseases.[4,5] The defect arises from the failure of the conotruncal ridges to fuse.[6] The aortopulmonary septum is formed by two truncal cushions very early in gestation, which quickly enlarges and fuse to separate aortic trunk and pulmonary channels. Neural crest cells play an essential role in influencing this division, and removal of neural crest cells results in various malformations including truncus arteriosus, transposition, and aortic arch interruption but, interestingly does not lead to APW. The defect may usually begin just above the sinuses of valsalva and extend distally into the arch. Unlike truncus arteriosus, the semilunar valves (aortic and pulmonary) are preserved in APW.[7]

APW is characterized by a large left-to-right shunt that progressively worsens with postnatal fall in pulmonary vascular resistance (PVR). Jacobs et al. classified APW into four types which are adopted by the society of thoracic surgeons' congenital heart surgery.[3]

- Type I is a proximal APW located just above the sinus of Valsalva, a few millimeters above the semilunar valve
- Type II is a distal APW located in the uppermost portion of the ascending aorta
- Type III is a total defect involving the majority of the ascending aorta
- Type IV is the intermediate defect[8] - similar to the total defect but with adequate superior and inferior rims

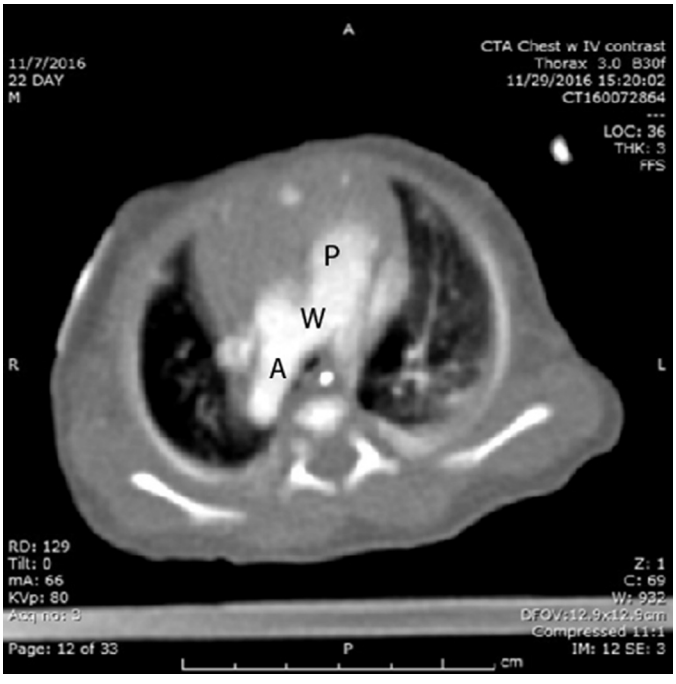

Fig. 2. CT chest angiogram with IV contrast, a coronal section at the level of pulmonary arteries. P – main pulmonary artery, W – Aortopulmonary window, A – ascending aorta.