

# Systemic-to-Pulmonary Collateral Impact in Premature Infant: Clinical Case Report

*Impacto de Colateral Sistêmico-Pulmonar em Recém-Nascido Prematuro: Relato de Caso Clínico*

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## Introduction

Congenital heart disease with left-to-right (L-R) shunt is an additional challenge to premature newborn (PTNB) management. Prematurity is responsible for an increased risk of acute and/or chronic lung disease due to pulmonary immaturity. Cardiac disorders of pulmonary hyperflow may worsen this condition.

Patent ductus arteriosus (PDA) is a clinical condition that determines frequent pulmonary hyperflow, but it is not the only L-R shunt condition in this population. Besides PDA, aortopulmonary window (rare pathology) and systemic-to-pulmonary collaterals (SPCs) are differential diagnoses. The onset of SPCs can cause congestive heart failure (CHF) in PTNB and mimic PDA with hemodynamic repercussions.<sup>1</sup> On the other hand, SPCs can occur transiently without increased morbidity.<sup>2</sup>

SPCs are bronchopulmonary communications that increase or proliferate from some stimulus, such as hypoxia or hypercapnia, commonly found in this population.<sup>3</sup>

Echocardiography is a noninvasive exam of great importance in this context, since it is able to diagnose and evaluate the degree of hemodynamic repercussion caused by SPCs.

Given the scarcity of studies in the literature on SPC in PTNBs, a situation that may worsen morbidity in this population, we will present a case of this condition and discuss peculiarities of the theme.

## Case Report

A 25-year-old mother, G3P3A0, uneventful prenatal on prenatal corticosteroids. Female newborn, second twin, extremely premature (GA = 28 5/7 weeks), suitable for gestational age (W = 932 g), donor fetus in fetofetal transfusion, cesarean section due to first-twin fetal distress, APGAR 3/5/7. Did not cry at birth. She was intubated in the delivery room, presented clinical stability and was referred to a neonatal intensive care unit.

## Keywords

Echocardiography; Infant, premature; Lung Diseases.

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Manuscript received 19/8/2019; revised 27/9/2019; accepted 18/11/2019

**DOI:** 10.5935/2318-8219.20200013

On the second day of life, echocardiography for prematurity protocol was performed. The test showed patent foramen ovale (PFO) and 1.2 mm CA without hemodynamic repercussion, still present with 1 mm on a control exam at 7 days of life, no longer detected at 14 days of life.

Received a surfactant dose due to grade 3 respiratory distress syndrome and mechanical ventilation ventilatory support for 17 days with maximum FiO<sub>2</sub> of up to 70%, followed by NIPPV (Nasal Intermittent Positive Pressure Ventilation) for 15 days, CPAP (Continuous Positive Airway Pressure) for 7 days and circulating O<sub>2</sub> for 5 days. Received antibiotics for 10 days for early neonatal infection. Remained stable with weight gain.

At 31 days of age, echocardiography was repeated due to difficulty weaning from oxygen therapy. It showed PFO, absence of flow in the ductus arteriosus topography, and presence of SPCs emerging from the descending aorta toward distal portions of the left pulmonary artery with continuous low-velocity flow from the aorta to the pulmonary tree (Figures 1 and 2). Left atrial/aortic ratio of 1.6 and left ventricular diastolic and systolic diameters were at the upper limit of normality. Volume restriction and diuretics were indicated, with good clinical outcome and successful weaning of oxygen therapy.

## Discussion

In the early stages of fetal development, pulmonary blood is supplied by arteries from the dorsal aorta. By the 40<sup>th</sup> day of gestation, blood supply via cardiac anterograde flow through the pulmonary artery is associated with the first supply. Around the 50<sup>th</sup> day of gestation, the latter disappears.<sup>1</sup>

SPCs occur frequently in congenital heart disease with reduced pulmonary blood flow and pulmonary diseases such as Fallot's Tetralogy (most prevalent cyanogenic congenital heart disease), bronchiectasis and pulmonary bronchodysplasia.<sup>4</sup> Such conditions have chronic hypoxia in common. Thus, it is postulated that hypoxemia is a trigger for the development of SPCs in premature newborns, i.e., bronchopulmonary arterial communications that increase and/or proliferate (angiogenesis), generating an alternative pulmonary blood supply. In this situation, the lungs receive blood from pulmonary circulation as well as from systemic circulation, causing pulmonary hyperflow and its consequences.<sup>5</sup>

Shaugnessy et al., in a retrospective echocardiographic study, detected an incidence of 4% (20/500) of SPC in premature newborns. Median age at SPC detection was 21 days. Fourteen patients had previous exams that did not detect SPC, suggesting the evolutionary character of the condition. The average gestational age of the newborns was 28 +/- 3 weeks and the

## Case Report

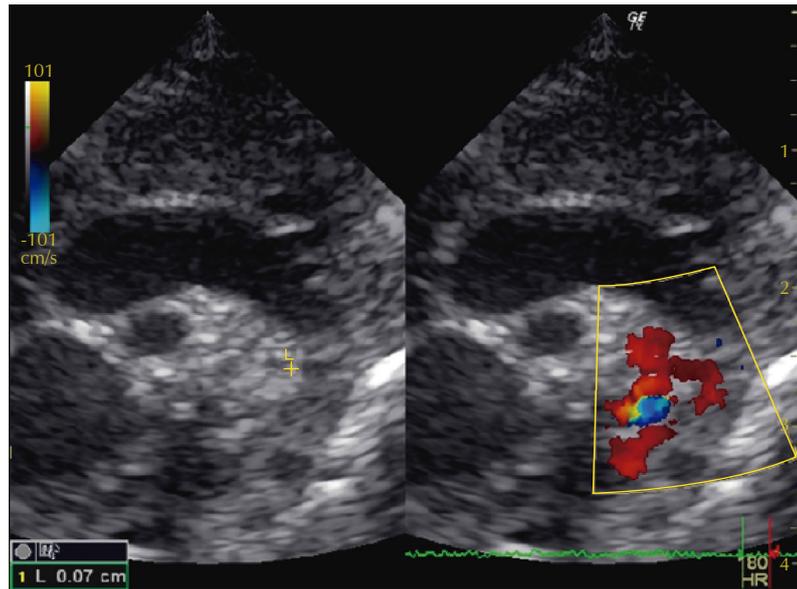


Figure 1 – Systemic-to-pulmonary collateral emerging from the descending aorta toward the pulmonary tree.

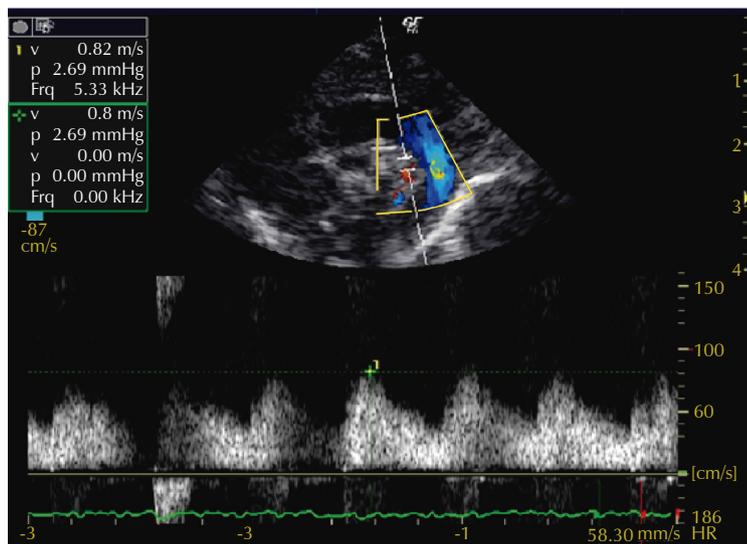


Figure 2 – Continuous flow of systemic-to-pulmonary collateral.

weight was 1,155 +/- 370 grams. Sixteen patients had hyaline membrane (80%) and 17 required orotracheal intubation (85%).<sup>2</sup> Such profile is similar to the one in this case.

Ancherman et al., in a prospective echocardiographic study, found an incidence of 66% (88 patients) of SPC in 136 newborns with very low birth weight (<1,500 grams). Patients with SPC stayed longer under positive pressure ventilation and

in hospital. Of the 88 patients with SPC, 10 (11%) had CHF, mimicking hemodynamic repercussion due to PDA. Nine patients had improvement of symptoms with anti-congestive medications, which were discontinued after 3 months. One patient had CHF and needed catheterization for SPC embolization, progressing with improvement. At follow-up up to 1 year of age, most patients had regression of the SPCs.<sup>1</sup>

The discrepancy of incidence between the two studies may be justified by the methodological difference, as the first one is retrospective and the second one is prospective.

The clinical picture may range from a transient benign condition (in most cases) to cardiac decompensation with interstitial edema and pulmonary alveolar edema, prolonging ventilatory support and oxygen supplementation time.<sup>5</sup> In 1995, Skinner described a PTNB with bronchodysplasia that presented clinical examination and echocardiogram suggestive of L-R shunt heart disease (PDA and SPCs). PDA ligation was performed, but there was no improvement. The SPCs maintained the CHF condition. The child died without clinical conditions for collateral closure.<sup>6</sup>

Color Doppler echocardiography is a noninvasive and affordable bedside imaging test that can diagnose SPC and its potential hemodynamic repercussion. Skinner et al. first published the use of color Doppler echocardiography for this diagnosis in 1995.<sup>6</sup> The suprasternal window is the best for SPC evaluation. Color Doppler can detect continuous flow mainly from the aortic arch, its branches and the proximal descending aorta toward distal portions of the pulmonary artery. This differentiates it from the SPC, whose terminal portion usually occurs at the distal part of the pulmonary trunk and is hourglass-shaped (SPCs are usually tortuous). The origin of the collateral is easily identified, but its distal portion in the pulmonary tree is not always visible. Signs of shunt repercussion such as enlargement of left

chambers and reverse flow in the descending aorta should be investigated mainly in newborns whose respiratory evolution is not satisfactory.<sup>1,2,5</sup>

Goel et al. in 2018 suggested that the gold standard imaging tests for the evaluation of SPC in premature newborns with cardiac decompensation and refractory to anti-congestive medications were computed tomography, nuclear magnetic resonance or cardiac catheterization. Such tests would provide more accurate information on the number (several small vessels or few large vessels), pathway and diameter of the SPCs than the echocardiogram for interventional treatment (surgical ligation or coil occlusion).<sup>5</sup>

## Conclusion

The presence of SPCs should be investigated as a cause of L-R shunt in PTNBs that do not have good respiratory pattern evolution. Color Doppler echocardiography is an affordable and sufficient imaging test to provide this diagnosis and to guide preliminary clinical management in this population, and the investigation may be complemented with other imaging methods when the outcome is delayed and unsatisfactory, requiring intervention.

## Conflict of interest

The authors declare that there is no conflict of interest regarding this manuscript.

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