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CASE REPORT

Transient Dynamic Subaortic Stenosis in Premature Neonates After Patent Ductus Arteriosus Ligation

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Abstract We describe 2 premature infants with PDA that did not respond to medical therapy and required surgical ligation. Both infants developed transient dynamic subaortic obstruction that resolved without specific therapy. This may have occurred due to sudden changes in the left ventricular volume.

Keywords Subaortic stenosis · Patent ductus arteriosus · Prematurity

Patent ductus arteriosus (PDA) results from failure of normal closure mechanisms after birth. Prematurity or immaturity of an infant at the time of delivery contributes to patency of the ductus. Conditions that contribute to low oxygen tension in the blood such as immature lungs, coexisting congenital heart defects, and high altitude are also associated with persistent patency of the ductus. PDA in the preterm infant is usually treated medically with indomethacin, and if pharmacology fails, it is closed surgically by ligation.

The early and late sequelae of ductus arteriosus ligation include pneumothorax, chylothorax, left phrenic nerve palsy, mistaken ligation of the left pulmonary artery, and recurrent laryngeal nerve palsy.

We report here two children with transient dynamic subaortic obstruction following ductus arteriosus ligation surgery. To our knowledge, this phenomenon has never been described following PDA ligation.

Case 1

A female infant was born at 26 weeks' gestation by spontaneous vaginal delivery to a 13-year-old, G1P0 African-American female. The mother had no significant family history of congenital heart disease. She received a limited amount of prenatal care and did not receive any prenatal steroids or indomethacin for preterm labor. The newborn's birth weight was 825 g. The Apgars were assigned as 4, 6, and 7 at 1, 5, and 10 min, respectively. Her early (preop) NICU course was complicated by respiratory distress syndrome (for which she received two doses of surfactant) and a Grade 1 intraventricular hemorrhage. She had persistent patency of the ductus arteriosus despite two courses of indomethacin therapy. Due to the continued need for mechanical ventilatory support and requirement of a high inspired oxygen fraction, the ductus arteriosus was surgically ligated on the seventh day of life. Intraoperatively, there was minimal blood loss. Follow-up examination after surgery revealed stable vital signs as well as excellent pulses and good peripheral perfusion. On cardiac examination a 2/6 systolic murmur was audible at the left lower sternal border. Follow-up echocardiogram

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showed normal cardiac anatomy, with normal, unobstructed flow in the pulmonary arteries and no residual ductal shunt. The left ventricular systolic function was hyperdynamic, with obliteration of the midventricular cavity in systole. Color Doppler showed increased flow velocity in the subaortic region and the spectral Doppler pattern indicated dynamic midsystolic obstruction with a peak velocity of 2.5 m/s. Supportive care with observation and with no specific medical therapy was provided. Over the next few days the murmur was no longer audible and a repeat echocardiogram 2 weeks later demonstrated no residual ductal shunt and no subaortic obstruction.

Case 2

A female infant, one of a set twins was born at 25 weeks' gestation via cesarean section secondary to breech presentation to a 24-year-old G1P0 Caucasian female whose prenatal course was complicated by preterm labor. Her maternal history was not significant and the mother had not received any steroids or indomethacin prior to delivery. The infant's birth weight was 732 g. Her Apgars were assigned as 5 and 5 at 1 and 5 min, respectively. Her early NICU course was complicated by respiratory distress syndrome (for which she received two doses of surfactant), adrenal insufficiency (requiring steroid supplement), and persistent PDA. The patient received indomethacin at birth for a moderate PDA, which was closed after the first course, but reopened after 2 weeks of life. Two subsequent courses of indomethacin failed to close her ductus and led to a need for surgical closure. Her echocardiogram demonstrated normal left ventricular function, with no signs of hypertrophy. The ductus was ligated on day of life 24. The patient received stress doses of steroids prior to surgery. She tolerated the procedure well. Following the PDA ligation, a persistent murmur was heard and an echocardiogram was performed. It demonstrated no residual ductal shunt, with normal left and right pulmonary arterial flow. There was a hyperdynamic left ventricular systolic function with mild dynamic subaortic cavity obliteration. Color Doppler showed increased flow velocity in the subaortic region (Fig. 1) and the spectral Doppler pattern indicated dynamic midsystolic obstruction with a peak velocity of 3.2 m/s (Fig. 2). Over several days the murmur resolved spontaneously. A repeat echocardiogram 10 days later demonstrated normal left ventricular function, with no evidence of any midcavity narrowing. Color and spectral Doppler flows in the subaortic region were normal (Fig. 3).

Discussion

We report herein two premature infants with transient subaortic dynamic stenosis post PDA ligation. This finding was detected by the appreciation of a murmur and confirmed echocardiography. To our knowledge this is the first report of this postoperative phenomenon. In both cases the obstruction resolved without any intervention.

Dynamic subaortic stenosis is a common finding in infants of diabetic mothers with hypertrophic cardiomyopathy. However, the exact etiology of the subaortic obstruction reported herein is unclear. The initial and follow-up echocardiograms demonstrated no evidence of any hypertrophic changes of the left ventricle observed in infants with features of hypertrophic cardiomyopathy. This

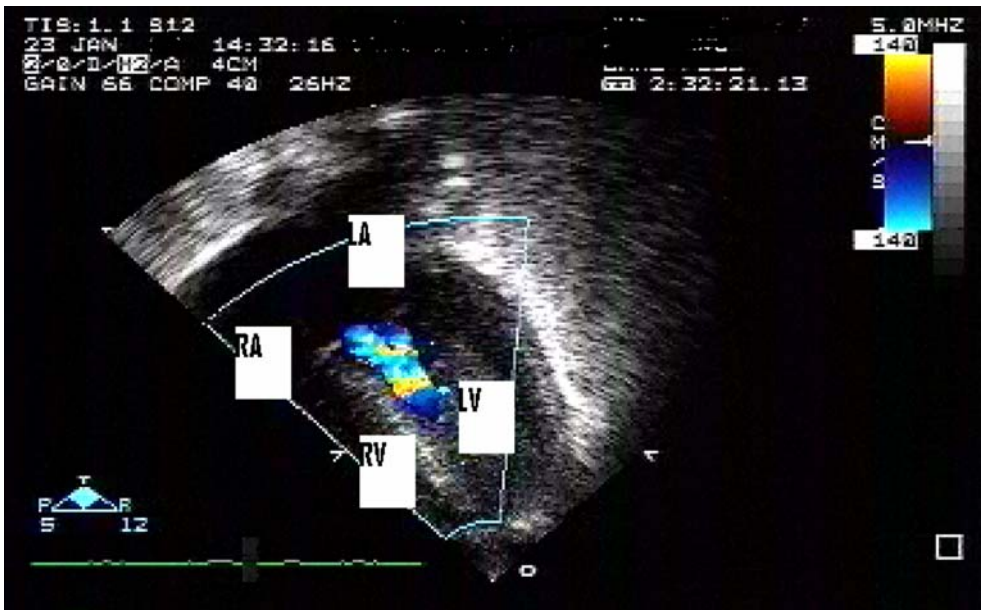

Fig. 1 Apical view of patient 2. Color Doppler demonstrates turbulent flow in the subaortic region. LA, left atrium; RA, right atrium; LV, left ventricle; RV, right ventricle

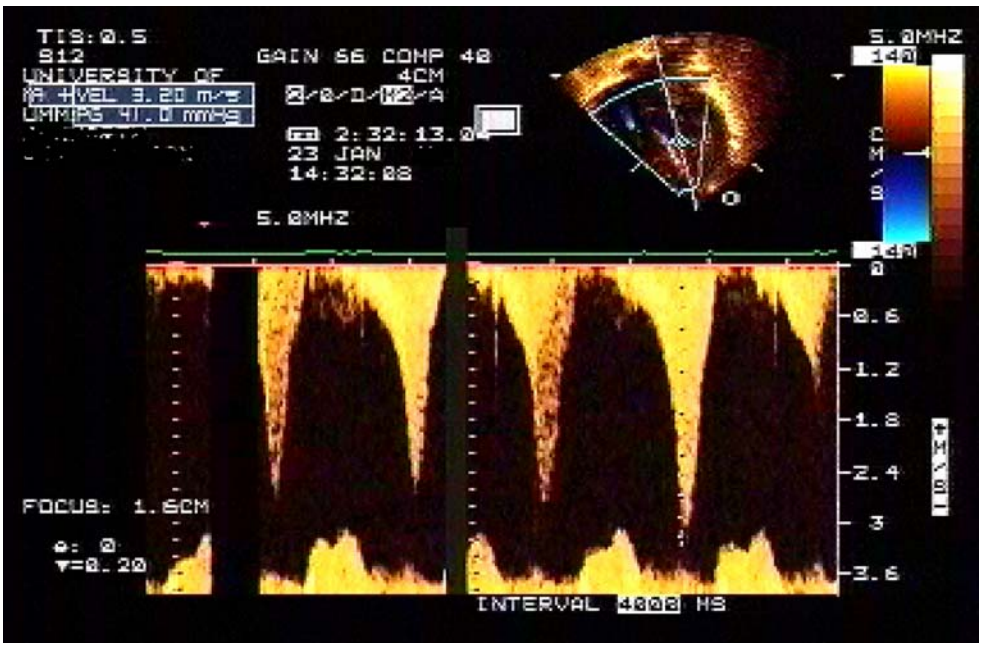

Fig. 2 Spectral continuous-wave Doppler through the left ventricular outflow tract, demonstrating dynamic obstruction with a peak velocity of 3.2 m/s

suggests that changes in the geometry, volume, and function of the left ventricle would explain these findings. The left-to-right shunt through the PDA increases the pulmonary venous return and, when large enough, leads to left atrial and ventricular dilation with signs of congestive heart failure. The left ventricular systolic function in neonates with PDA is normal. Moreover, there is an increase in the cardiac output proportionate to the size of the shunt [3, 4]. Using echocardiographic techniques, the change in left ventricular volume and its effect on systolic function were studied before and after the closure of the ductus arteriosus. Although left ventricular output was significantly higher during the patency of the ductus arteriosus, there was no significant difference in indexes of contractility before and after spontaneous ductal closure in full-term neonates [4].

Either spontaneous or pharmacological closure of a patent ductus with indomethacin occurs over several hours to days. Thus, there is a gradual reduction in the shunt through the ductus, with corresponding changes in the size of the left heart. Previous studies have demonstrated that left ventricular contractility as measured by the mean normalized systolic ejection rate did not change in full-term infants before and after PDA closure [4]. Similarly, in preterm infants the shortening fraction did not change after

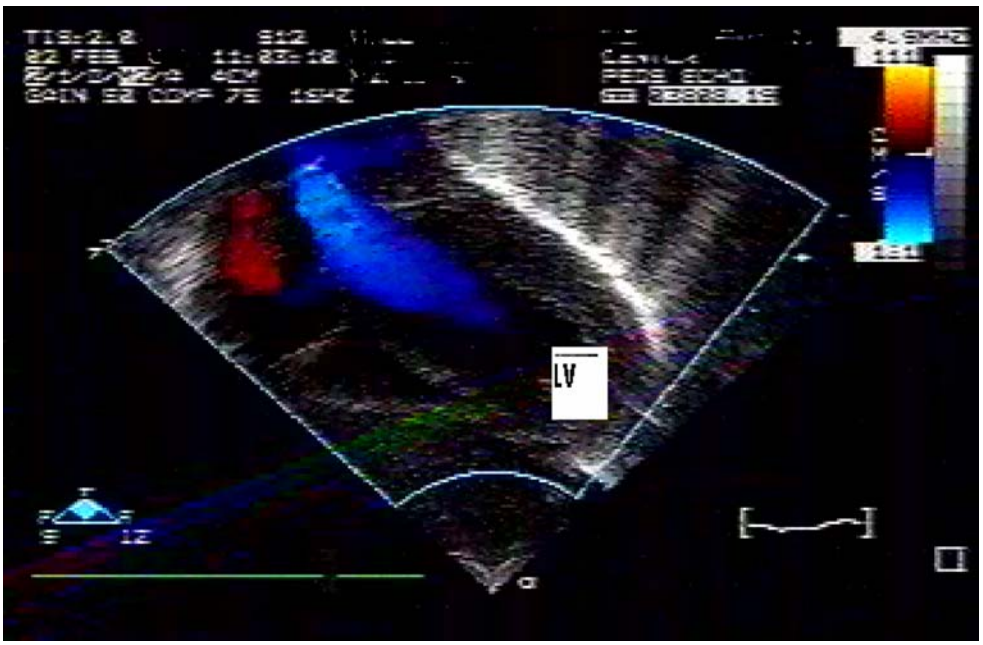

Fig. 3 Subxyphoid image of the left ventricular outflow tract, 10 days after patent ductus arteriosus ligation, demonstrating a normal systolic color Doppler flow pattern, with no evidence of any residual subaortic obstruction

the first day of life regardless of the patency of the ductus [6]. We hypothesize that the dynamic subaortic obstruction was due to a transient mismatch between the left ventricular muscle mass and the volume of the cavity. A similar phenomenon has been observed in an adult with renal failure after hemodialysis and a significant volume extraction [5]. Moreover, unlike the spontaneous and pharmacologic closure of the PDA, when intrinsic catecholamine secretion should not increase, our patients had the added stress of the surgery. This added stress may have generated a significant inotropic drive to the myocardium, resulting in the clinical picture that we observed. Our findings are similar to those observed in adult patients exposed to high doses of catecholamines who demonstrated transient left ventricular outflow tract obstruction, some requiring β-blockade in addition to withdrawal of inotropic support [1, 2]. Thus, intrinsic catecholamine release, coupled with the sudden decrease in the pulmonary venous return and loss of intravascular volume due to edema of the tissues in the operative field, could have combined to produce these dynamic changes. Although we did not attempt any specific therapy to reduce myocardial contractility and heart rate (such as a β-blocker), the use of these modalities as well as increasing the intravascular volume may be useful if a severe case should occur [2]. It is important to realize that this was a transient phenomenon and long-term therapy may not be needed.

In conclusion, this unusual finding of dynamic subaortic obstruction appears to be transient and not associated with adverse cardiac outcome. To our knowledge, this transient finding has not previously been reported in the literature.

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