Cardiol Young 2002; 12: 581–583

ISSN 1047-9511

Brief Report

Pulmonary hypertension due to spontaneous premature ductal constriction in fetal life: association with right bundle branch block

Alessandra Benettoni,[1] Maria Angela Rustico[2]

[1]*Pediatric Cardiology Unit;* [2]*Department of Obstetrics and Gynaecology, IRCCS, Istituto per l'Infanzia, "Burlo Garofolo", Trieste, Italy*

Abstract We describe a case of fetal pulmonary hypertension and tricuspid regurgitation due to non pharmacologically induced ductal constriction observed at 36 weeks' gestational age. The hypertension resolved spontaneously soon after birth, with no functional consequences. Right bundle branch block is the only permanent anomaly, still being seen on the electrocardiogram at the age of 34 months.

Keywords: Ductus arteriosus; fetal echocardiography; abnormal conduction

Structural anomalies, or pharmacologically induced constriction, of the arterial duct during the third trimester of pregnancy are known causes of fetal and neonatal pulmonary arterial hypertension. Right ventricular dilation and dysfunction, and tricuspid valvar regurgitation, are frequently reported in these cases.[1,2] Typically, there is spontaneous improvement of the right ventricular function, with complete normalization of the flow across the tricuspid valve, after birth, and no permanent anomalies are detectable after the neonatal period. We describe here a 34 months old infant, who suffered from fetal pulmonary hypertension due to non-pharmacologically induced premature constriction of the arterial duct, and in whom right bundle branch block has persisted on the electrocardiogram.

Case report

A healthy 28-year-old woman was referred for detailed fetal echocardiography at 36 weeks' gestation, after a dilated right ventricle was observed a few days before in another hospital. The previous scans were reported as normal. There was a familial history of Ebstein's malformation of the tricuspid valve in one cousin, and pulmonary valvar stenosis in another. The patient had not taken non-steroidal antiinflammatory drugs during pregnancy. Fetal echocardiography showed a dilated and hypertrophic right ventricle, with poor contractility and septal curvature towards the left ventricle, suggesting right ventricular pressure overload (Fig. 1a). The right atrium was only slightly dilated. The tricuspid valvar annulus was dilated, with normal leaflets. The arterial duct was tortuous and narrowed at its aortic connection (Fig. 1b). Colour flow Doppler showed tricuspid valvar regurgitation, with the aliasing phenomenon suggesting flow at high velocity (Fig. 1c). Due to technical reasons, we could not measure the peak velocities of the tricuspid regurgitation jet nor the ductal flow. The fetus was monitored with serial scans until term. No signs of heart failure were observed. The woman gave birth spontaneously at 41 weeks' gestation to a male weighing 4100 g. The apgar scores of the neonate were 8 and 10, respectively, at 1 and 5 min after birth. Wide splitting of the second heart sound, without any murmur, was noted on physical examination of the infant on the first day of life. The electrocardiogram showed complete right bundle branch block, with coarse notched R and S waves (Fig. 2a). The echocardiographic examination on

Correspondence to: Alessandra Benettoni, Pediatric Cardiology Unit, IRCCS Istituto per l'Infanzia Burlo Garofolo, via dell'Istria 65/1, 34137 Trieste, Italy. Tel: +39 040 3785463; Fax: +39 040 3785448; E-mail: benettoni@ burlo.trieste.it

Accepted for publication 14 August 2002

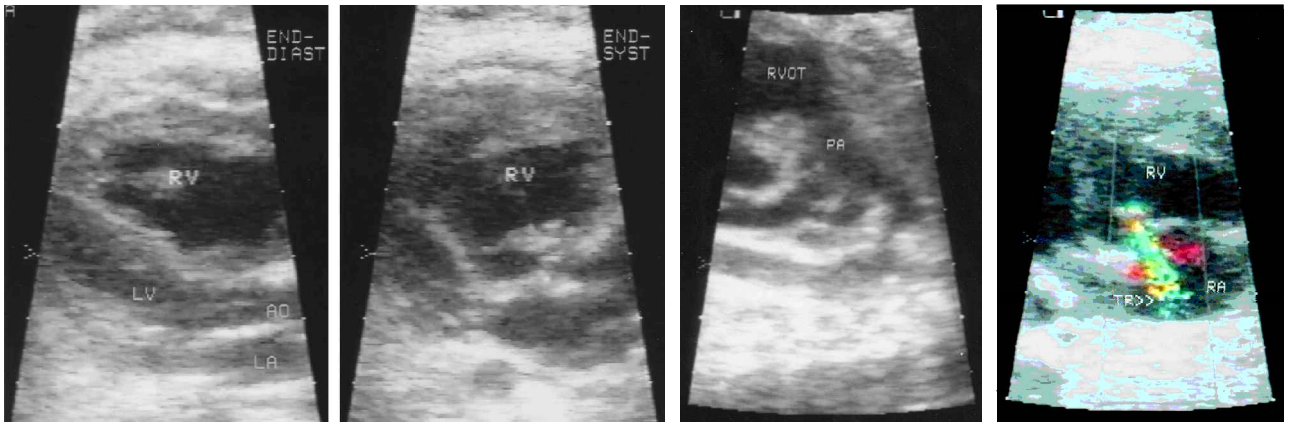

Figure 1.
Fetal echocardiography at 36 weeks' gestational age. (a)shows the long axis of the ventricles in systole and diastole. Note the right ventricular dilation and the inverted curve of the ventricular septum. (b) shows the short axis at the level of the ductal arch, revealing the tortuous and narrowed arterial duct. (c) is the four chamber view, with colour Doppler at the level of the tricuspid valve regurgitant flow with the aliasing phenomenon. RVOT: right ventricular outflow tract; PT: pulmonary trunk; RV: right ventricle; LV: left ventricle; AO: aorta; LA: left atrium.

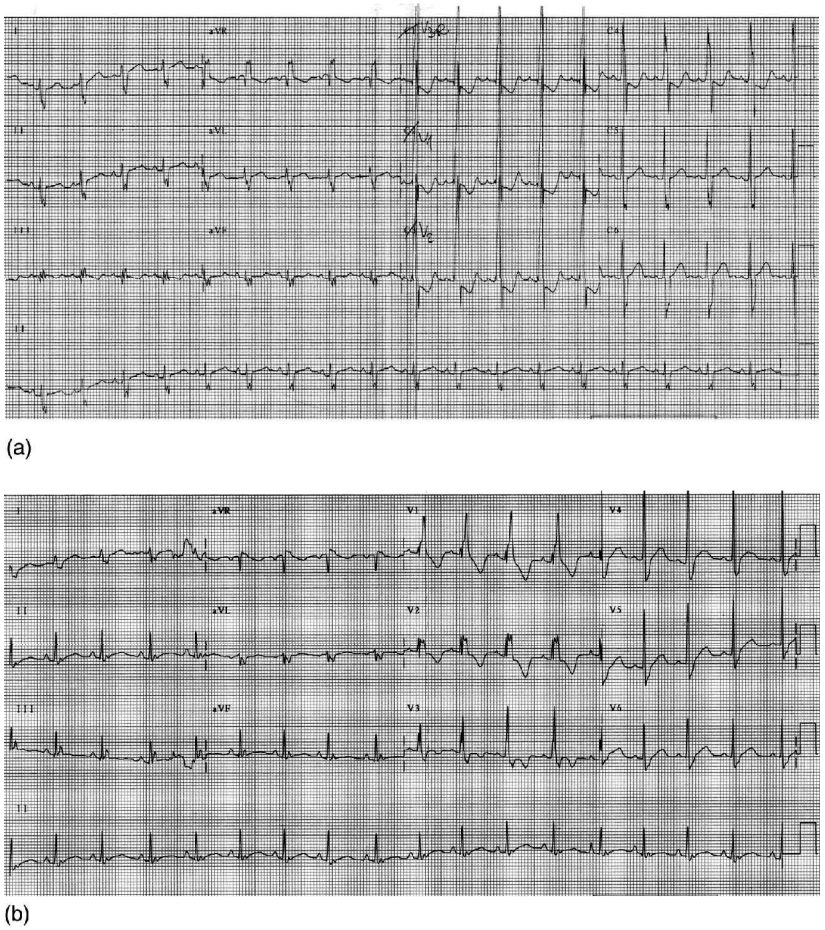

Figure 2.
Electrocardiograms of the infant. (a) is the electrocardiogram at the age of 1 day. Note the notched QRS complex, with the pattern of right bundle branch block. (b) shows the electrocardiogram at the age of 34 months, with persistence of the pattern of right bundle branch block.

the first day of life showed a dilated and slightly hypertrophic right ventricle, with paradoxical bulging of the septum towards the left ventricle in systole. No ductal shunt or tricuspid regurgitation were detectable after birth. Serial scans performed within the first month of life and afterwards showed complete recovery of the right ventricular anomalies. The infant remained totally asymptomatic. The electrocardiogram at last follow-up, at 34 months of age, continued to show right bundle branch block (Fig. 2b).

https://doi.org/10.1017/S104795110200104X Published online by Cambridge University Press

Discussion

Premature constriction of the arterial duct may be the consequence of structural anomalies,[1] or may occur in patients who have been treated with indomethacin for preterm labour.[2] In our case, the morphology of the duct, which was narrowed and tortuous, suggested it had constricted prematurely, with consequent fetal pulmonary hypertension and right ventricular dysfunction. Acute ductal occlusion produced experimentally in animals is followed by tricuspid regurgitation, a significant increase in pulmonary arterial pressure, and a decrease in the right ventricular ejection fraction.[3,4] Right ventricular dysfunction and tricuspid regurgitation due to ductal constriction in fetuses exposed to indomethacin are considered to be reversible after birth, because the load on the right ventricle decreases as a result of the physiological transition from the fetal to the neonatal circulation.[5–7] In the majority of patients, no permanent anomalies are reported after birth, although severe tricuspid valvar regurgitation has been described in a child exposed to diclofenac during fetal life.[8] In this case, the authors suggested that subendocardial ischaemia of the papillary muscles could have occurred as a consequence of the pulmonary hypertension.

In our case, the premature fetal ductal constriction occurred spontaneously in a fetus not exposed to non steroidal antiinflammatory drugs. It caused pulmonary hypertension, right ventricular dysfunction, and secondary tricuspid regurgitation that rapidly resolved after birth. The finding of right ventricular dysfunction did not change the conservative management of the pregnancy, allowing spontaneous delivery at term. When normalization of the pulmonary arterial pressure occurred after birth, neither the right ventricular dysfunction, nor the tricuspid insufficiency could be detected. Right bundle branch block, however, was observed in the electrocardiogram that still persisted at the age of three years, without any functional consequence.

The interest of this case, in our opinion, is the documentation of prenatal right ventricular overload due to spontaneous premature constriction of the duct with no consequences after birth, save from right bundle branch block. This could be the consequence of the fetal changes, but could equally be a coincidental association.

Acknowledgement

We thank Dr. James C Huhta and Dr. Gilda Caruso for their contributions.

References

1. Mielke G, Peukert U, Krapp M, Schneider-Pungs J, Gembruch U. Fetal and transient neonatal right heart dilatation with severe tricuspid valve insufficiency in association with abnormally S-shaped kinking of the ductus arteriosus. Ultrasound Obstet Gynecol 1995; 5: 338–341.
2. Tulzer G, Gudmundsson S, Sharkey AM, Wood DC, Cohen AW, Huhta JC. Doppler echocardiography of fetal ductus arteriosus constriction versus increased right ventricular output. JACC 1991; 18: 1532–1536.
3. Eronen M, Pesonen E, Kurki T, Ylikorkala O, Hallman M. The effects of indomethacin and a Beta-sympathomimetic agent on the fetal ductus arteriosus during treatment of premature labor: a randomized double-blind study. Am J Obstet Gynecol 1991; 164: 141–146.
4. Tulzer G, Gudmundsson S, Tews G, Wood DC, Huhta JC. Incidence of indomethacin-induced human fetal ductal constriction. J Matern fetal Invest 1992; 1: 267–269.
5. Tulzer G, Gudmundsson S, Rotondo KM, Wood DC, Yoon GY, Huhta JC. Acute fetal ductal occlusion in lambs. Am J Obstet Gynecol 1991; 165: 775–778.
6. Levin DL, Mills LJ, Weinberg AG. Hemodynamic, pulmonary vascular, and myocardial abnormalities secondary to pharmacologic constriction of the fetal ductus arteriosus. Circulation 1979; 60: 360–364.
7. Harada K, Rice MJ, Shiota T, McDonald RW, Reller MD, Sahn DJ. Two-dimensional echocardiographic evaluation of ventricular systolic function in human fetuses with ductal constriction. Ultrasound Obstet Gynecol. 1997; 10: 247–253.
8. Zenker M, Klinge J, Kruger C, Singer H, Scharf J. Severe pulmonary hypertension in a neonate caused by premature closure of the ductus arteriosus following maternal treatment with diclofenac: a case report. J Perinat Med 1998; 26: 231–234.