Misleading hyperoxia test

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Summary A 2 day old infant with infracardiac total anomalous pulmonary venous connection achieved an arterial oxygen tension > 250 mmHg, breathing 100% oxygen. Diagnosis of cardiac disease was delayed several weeks. The anatomical basis for this phenomenon was streaming of systemic venous return across an inferior vena cava type atrial septal defect.

Cardiorespiratory distress in the newborn infant may result from a variety of disorders. The hyperoxia or nitrogen washout test is widely used and claimed to be a sensitive and accurate discriminator between cyanotic and acyanotic cardiac lesions and pulmonary disease. An oxygen tension greater than 250 mmHg in an arterial sample from the right arm after 100% oxygen has been administered for 5–10 minutes is said to exclude cyanotic heart disease; while a value of more than 150 mmHg makes its occurrence extremely unlikely. In the patient presented here, however, a combination of factors obscured the typical presentation of a common mixing type, cyanotic heart lesion.

Case report

A 1800 g girl was transferred to the Royal Liverpool Children’s Hospital at 3 weeks of age with increasing tachypnoea. Delivery to a 21 year old gravida 2 Asian mother at 34 weeks’ gestation had been complicated by birth asphyxia, necessitating intubation and ventilation for roughly 10 minutes. At 6 hours the baby had suffered cyanotic spells and was nursed in 60% oxygen. This increased the transcutaneous oxygen tension from 25 to 60 mmHg, but at 48 hours brief periods of apnoea were associated with bradycardia. Chest x ray film and electrocardiogram (ECG) showed no abnormality, and, on a single hyperoxia test, arterial oxygen tension rose to 259 mmHg in 100% oxygen.

During the next fortnight the infant became progressively lethargic and developed hepatomegaly, mild jaundice, and blood in her stools. Although her transcutaneous oxygen tension was maintained between 55 and 70 mmHg, with inspired oxygen concentrations of 45% to 80%, the arterial carbon dioxide tension steadily increased. Chest x ray films eventually showed bilateral diffuse opacities, and the hyperoxia test was repeated at 17 days. On this occasion, arterial oxygen tension increased only to 75 mmHg, and the patient was referred for cardiac investigation.

On admission, the respiratory rate was 70 breaths per minute with intercostal recession and mild central cyanosis. The second heart sound was single and accentuated, but no murmurs could be heard.

Fig. 1 Venous phase of the pulmonary angiogram, showing drainage to a large intrahepatic channel, which is obstructed near the diaphragm (arrow). Early opacification of the left ventricle is also seen.
Chest x-ray film showed severe, generalised pulmonary congestion with a normal cardiac silhouette, but the only ECG abnormality was an upright T wave in V1, suggesting right ventricular hypertrophy. Two dimensional echocardiography visualised a retrocardiac pulmonary venous confluence, without connection to the heart. Cardiac catheterisation confirmed the diagnosis of infracardiac total anomalous pulmonary venous connection with a patent ductus arteriosus and pulmonary hypertension (Fig. 1). Representative saturations were 48% in superior vena cava, 83% in right atrium, 90% in inferior vena cava, and 82% in right ventricle, with 94% in both left atrium and left ventricle.

Emergency total correction was performed under profound hypothermia and circulatory arrest. All the pulmonary veins joined a confluence behind the heart and drained through the right side of the diaphragm by way of a dilated channel. A large atrial septal defect with no inferior rim occupied the fossa ovalis position. This was closed with a pericardial patch, the descending vein was divided, and the pulmonary venous confluence was anastomosed to the left atrium. The infant required prolonged postoperative respiratory support but otherwise made a complete and uneventful recovery.

Discussion

As a result of high resistance in the hepatic microcirculation, infracardiac total anomalous pulmonary venous connection usually presents during the first days of life with signs of obstructed pulmonary venous return. The fetal tendency for blood from the inferior vena cava to stream across a patent foramen ovale often raises systemic oxygen saturation above that in the pulmonary artery; but, none the less, arterial saturation is still generally lower in this than other types of total anomalous pulmonary venous connection, and sufficient mixing occurs at atrial level to reduce arterial oxygen tension well below 250 mmHg in the hyperoxia test.4

In the patient considered here a large, inferior vena caval type, atrial septal defect probably accentuated streaming, such that oxygenated blood from the liver returned almost exclusively to the left atrium (Fig. 2). Certainly, her left atrium and left ventricle were unusually well developed for an infant with total anomalous pulmonary venous connection, suggesting they had carried a reasonable portion of the cardiac output during fetal life. Because of prematurity a widely patent ductus venosus probably delayed development of pulmonary venous obstruction, while patency of the ductus arteriosus allowed further decompression of the pulmonary circulation into the aorta.5 Thus the electrocardiogram showed little evidence of right ventricular dominance. By the time of the second hyperoxia test, restriction of flow through these two channels resulted in pulmonary hypertension and venous congestion, with secondary rise of the right atrial pressure and consequent mixing of venous return.

Experience with open heart surgery in patients of this size is limited, but, on this occasion, no special difficulties were encountered with either the conduct of cardiopulmonary bypass or technical repair of the cardiac defect. Although the small, preterm infant may be at greater risk of postoperative pulmonary complications, it is still possible to obtain a good surgical result. The diagnosis of total anomalous pulmonary venous connection in a neonate with increasing cardiopulmonary distress cannot be excluded completely by a single, high arterial oxygen tension on the hyperoxia test. Awareness of the potential for unusual presentation and the application of two dimensional echocardiography should facilitate early diagnosis and treatment of cardiac disease in this group of patients.

**Fig. 2** Diagram of infracardiac pulmonary venous drainage with a patent ductus venosus and streaming of inferior vena caval blood to the left atrium.

PV=pulmonary veins; HV=hepatic venous channels; DV=ductus venosus; IVC=inferior vena cava; RA=right atrium; LA=left atrium.
References

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Commentary

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The hyperoxia or nitrogen washout test is used to differentiate pulmonary disease from cardiac disease in the cyanotic neonate. A timely reminder that a major rise in systemic arterial oxygen tension (>).150 mmHg) does not always exclude the presence of a basically cyanotic lesion is given above, and the lesion described is not the sole lesion responsible for this response in the first few days of life.

A more common failure of differentiation occurs in the presence of pulmonary disease associated with a cardiac lesion. In the very young neonate increasing the ambient oxygen concentration does not invariably overcome pulmonary venous desaturation, so that clinically inapparent lung disease may limit the rise in systemic arterial saturation, and an apparently minor chest radiological abnormality can be associated with so profound a change in pulmonary circulatory physiology that there is no rise in systemic arterial saturation under the conditions of the test.

The result of the test may only be appropriately interpreted if the arterial samples are taken from the right arm (assuming situs solitus), the carbon dioxide tension is normal, and the infant exhibits no clinical features of respiratory distress. The chest radiograph should be carefully scrutinised for any pulmonary abnormalities. Remember a change in ductal calibre may lead to a different result on repeat testing. Cross sectional echocardiography is an essential examination in the differentiation of pulmonary from cardiac disease and in delineating the nature of any cardiac lesion in the neonatal period. In experienced hands it is the more reliable examination.

Vanishing earrings

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Case histories

Case 1. A child of 8 years who was known to have asthma and mild eczema had her earlobes pierced with a paediatrician’s blessing six weeks before presenting to the hospital. Sterling silver earrings were used. Three weeks after the procedure she demanded and was permitted a pair of gold plated ornamental studs. Within a week the right earlobe became itchy and crusty. The condition resolved after removal of the studs. A few days before she presented, however, she had replaced the studs. The right earlobe became mildly itchy again, and one morning the lobe was found to be extremely swollen and painful. The earring could not be located and was thought to have fallen out.

SUMMARY Four children who presented with impacted earrings are described. We suggest that the insertion of earrings in children under 10 years has hazards and recommend the use of sterling silver or 9 ct gold if the procedure is to be done in young children.