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Letter to the Editor

Anomalous extension of ductal tissue within the pulmonary artery

Sir,

We read with interest the article from Gennery et al.[1] describing 3 asymptomatic infants with a cardiac murmur in whom the echocardiogram revealed "duct-related intimal tissue plaques" within a normal left pulmonary artery. No associated cardiac lesions were detected in any of them. In the report, the authors concluded that such a similar condition is unlikely to cause significant haemodynamic disturbance.

We recently performed surgery on a neonate diagnosed echocardiographically as having severe obstruction to pulmonary outflow, due to an isolated mass located in the left pulmonary artery. He was asymptomatic, but with a grade 3/6 ejection systolic murmur heard at the pulmonary area. The site of the white circular mass (Fig.1a), its echocardiographic appearance, the lack of regression in serial echocardiograms, and a resonance imaging scan, convinced us that the mass might be "organized" ductal tissue. The fear of pulmonary embolism, or organized thrombus, along with right ventricular hypertrophy with tricuspid incompetence estimating a Doppler peak instantaneous pressure gradient of 80mmHg, represented for us good indications to proceed to surgery. The mass was resected using cardiopulmonary by-pass (Fig. 1b). After the mass had successfully been removed, the lumen of the left pulmonary artery was judged as appropriate, and there was no need to reconstruct the vessel. The pathology showed that it was organized calcified thrombus, most likely ductal tissue.

Gennery et al.[1] report different anatomical variants, the purported common etiology being the migration of ductal tissue within pulmonary tree. According to this concept, they suggest that the possibilities range from limited ductal tissue, within a pulmonary artery of good size, as reported in their 3 cases, to severe hypoplasia of the pulmonary arteries in cases with acquired pulmonary discontinuation[2] due to massive ductal obliteration.

The case we treated is peculiar in that it represents migration of ductal tissue within a normally sized pulmonary artery. According to this, we think that it is correct to characterize our case as the extreme form of the same pathological entity as described by Gennery et al.[1] On the other hand, coarctation of the pulmonary artery,[3–5] even if also

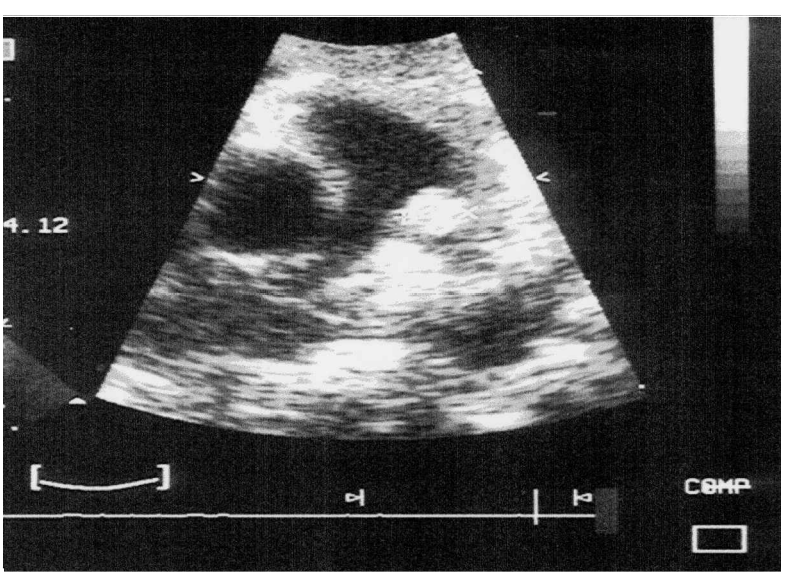

Figure 1a.
Parasternal short-axis view, demonstrating the mass at the origin of the left pulmonary artery

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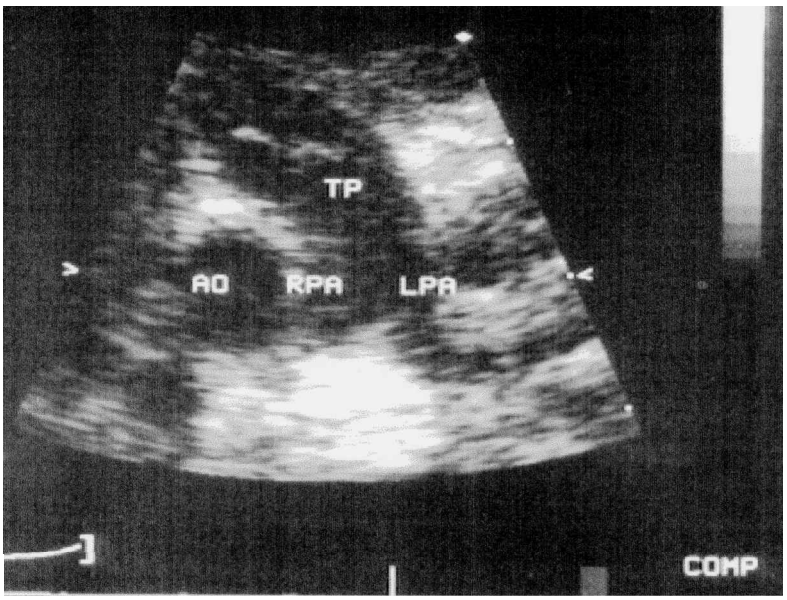

Figure 1b.
Parasternal short-axis view of the same patient after surgical removal of the mass

the consequence of ectopic ductal tissue, is usually associated with cardiac malformations characterised by a ventricular septal defect and obstruction of the pulmonary outflow tract. This common association is suggestive of a pathophysiological event occurring very early during cardiac fetal development,[6] which is then magnified by reduced or even absent forward flow through the pulmonary trunk, predisposing the development of abnormal pulmonary arborization. In these cases, the "benign" form may be represented by mild stenosis of the left pulmonary artery, while the extreme form is usually represented by discontinuation of the hypoplastic left pulmonary artery from the pulmonary bifurcation.[7]

We would suggest, therefore, that the pathological entity represented by invasion of ductal tissue within a pulmonary artery is a spectrum of abnormality, determined not only by the amount of ductal tissue obliterating the pulmonary vessel, but also by the timing of its abnormal migration during fetal life.

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This letter was shown to Dr Gennery and his colleagues, who concur with the concepts advanced, commenting that they have nothing further to add.

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