Quantitative Perfusion Imaging Assessing Acquired Discrete Peripheral Pulmonary Artery Stenosis

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We present the case an 8-yr-old boy evaluated for anastomotic stenosis of the right pulmonary artery after surgical repair of hemitruncus at 6 wk of age. Pulmonary angiography revealed only mild narrowing and a 10-mm pressure gradient across the anastomosis, but quantitative perfusion imaging demonstrated that the right lung only received 16% of pulmonary blood flow. Subsequently, balloon angioplasty of the anastomotic site was performed, resulting in complete resolution of the stenosis and gradient. Early postangioplasty perfusion imaging demonstrated increased perfusion of the right lung to 35% of total pulmonary blood flow. It is theorized that initially a chronically hyperperfused lung may develop more capacious vessels and recruit new capillaries during the years of hyperperfusion such that a "perfect" angioplasty may result in less than symmetric perfusion. The inexpensive, noninvasive quantitative perfusion study is more sensitive and accurate in evaluating acquired (postsurgical) pulmonary artery stenoses.

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Peripheral pulmonary artery stenosis often occurs over time at the site of anastomosis when the peripheral vessel is surgically rejoined to the main pulmonary artery (MPA). Children who are born with hemitruncus require reanastomosis of either the right or the left pulmonary artery to the main, as do many patients who have had pulmonary artery banding for complex intracardiac lesions. The scarring at the anastomotic site is a natural response to the surgical procedure and not an intrinsic part of the congenital anomaly (*1–4*). The stenosis is therefore said to be acquired and is characteristically discrete. Balloon angioplasty of acquired (postsurgical) discrete peripheral pulmonary stenosis is performed when the cine angiogram shows a discrete narrowing in the presence of a significant pressure gradient (*5–7*). Although the success of angioplasty is evaluated by angiographic and pressure gradient criteria, actual perfusion (left lung as compared to right lung) can only be measured by a nuclear perfusion study (*8–11*).

For two decades, macroaggregates of human serum albumin have been used to evaluate regional pulmonary blood flow quantitatively (*12–13*) with a 0.97 correlation coefficient between MAA distribution and red blood cell distribution (*14*). The accuracy of differential perfusion imaging has been well demonstrated in the management of bronchogenic carcinoma with reliable prediction of postoperative forced expiratory volume per second (FEV1) following lung resection (*15–19*). Sodium pertechnetate MAA perfusion scans have been utilized in postoperative follow-up of tetralogy of Fallot and related anomalies adequately identifying residual areas of distal stenosis (*20–21*). Little data exist to define an ideal result on perfusion study after balloon angioplasty of an isolated peripheral lesion in an otherwise healthy pulmonary vascular bed (resistance <3 Woods units). We present a patient who had his discrete right pulmonary artery stenosis assessed by nuclear imaging before and after angioplasty and who had an ideal result by both pressure and angiographic criteria.

CASE REPORT

S.A. presented at 6 wk of age with classic signs of congestive heart failure. He was referred to a medical center where the diagnosis of right hemitruncus (right pulmonary artery taking its origin from the ascending aorta) was established. Under cardiopulmonary bypass, a revision of the hemitruncus was performed with the right pulmonary artery reanastomosed to the main pulmonary artery at the usual site of origin. The patient did well and was followed at infrequent intervals until 8 yr of age when a routine color Doppler study performed in the absence of any physical findings showed continuous turbulence in the right pulmonary artery. Since a chest film suggested decreased perfusion of the right pulmonary vascular bed, a cardiac catheterization and main pulmonary cine angiogram were performed to evaluate the hemitruncus repair. Because the AP angiogram showed only mild to moderate narrowing at the origin of the right pulmonary artery and because the systolic gradient across the origin of the right pulmonary artery was only 10 mmHg, without elevated distal pressures, the right lung was considered to be adequately per-

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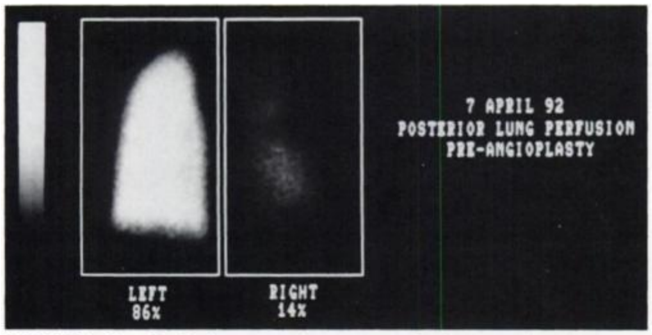

FIGURE 1. Perfusion study pre-angioplasty demonstrates marked reduction in pulmonary blood flow to right lung.

FIGURE 3. Pulmonary angiogram postangioplasty. Perfect result evident when compared with Figure 2.

fused and no balloon angioplasty was performed. The patient was followed clinically without any change in physical examination or chest x-ray.

Eighteen months after cardiac catheterization, the patient was seen routinely for echocardiographic study. The patient's increase in size and weight made color Doppler interrogation of the right pulmonary artery difficult, so a perfusion study was performed using 3.5 mCi ^{99m}Tc-macroaggregated albumin to further document the integrity of the hemitruncus repair. Remarkably, the study showed only 14% of pulmonary blood flow directed to the right lung (Fig. 1). It was assumed that a recent change had occurred in the patient's anatomy so he was taken back to the cardiac catheterization lab for restudy. At catheterization, the pressures across the anastomosis were identical to those at the previous study and the MPA angiogram showed that no change in the right pulmonary artery anatomy had occurred over the intervening period (Fig. 2). Based on the perfusion study, an angioplasty of the proximal right pulmonary artery was performed using a 20-mm balloon diameter with a 4-cm length with three inflations to six atmospheres. A near complete resolution of the stenosis was achieved with no systolic pressure gradient remaining across the previous site of narrowing (Fig. 3). A perfusion scan performed 8 days after the angioplasty showed 35% perfusion to the right lung with 65% perfusion to the left lung (Fig. 4). A study 1 yr later showed essentially equal flow to each lung.

DISCUSSION

Discrete postsurgical peripheral pulmonary stenosis can be an insidious and progressive lesion which may escape detection on physical examination (*1*). Our patient had no physical findings (e.g., a continuous murmur) to suggest severe stenosis. His chest x-ray showed an equivocal decrease in vascularity on the affected side as opposed to the unaffected side. Color Doppler was useful only in suggesting the presence of stenosis because views of the right pulmonary artery showed systolic and diastolic turbulence on color study. Pulsed wave and continuous wave Doppler were not particularly useful in our patient as the jet of turbulence could not be perfectly accessed due to overriding lung. Additionally, a slight or small gradient on Doppler study would constitute no source of reassurance since the pulmonary circuit is a low pressure system. Even our patient's first angiogram and pressure data (which were identical to the second) suggested little cause for concern. (The angiogram can generate false reassurance because it evaluates stenosis only from the frontal plane). The perfusion scan proved to be the most sensitive method for measuring selective pulmonary blood flow. In the absence of any dramatic clinical, echo or angiographic findings, only 14% of cardiac output was directed to the affected side by perfusion study.

In contrast to patients with diseased pulmonary vessels secondary to rubella, William's syndrome, end stage tetralogy of Fallot and related lesions, patients who have had pulmonary vessels reconstructed after hemitruncus and pulmonary artery band repairs have normal pulmonary vascular beds (*22*). These infant patients present for initial surgery with the picture of congestive heart failure caused by overperfusion of the lungs; therefore, the possibility of important pulmonary vascular disease (increased resistance) is precluded. Although such patients have increased

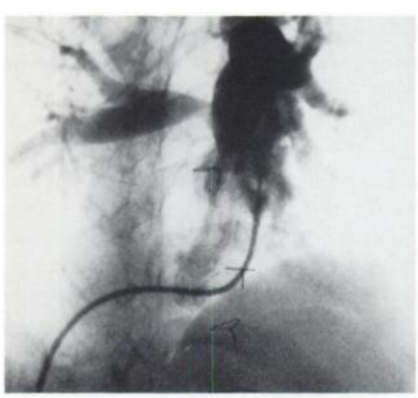

FIGURE 2. Pulmonary angiogram pre-angioplasty. Note narrowing at anastomotic site near origin of right pulmonary artery.

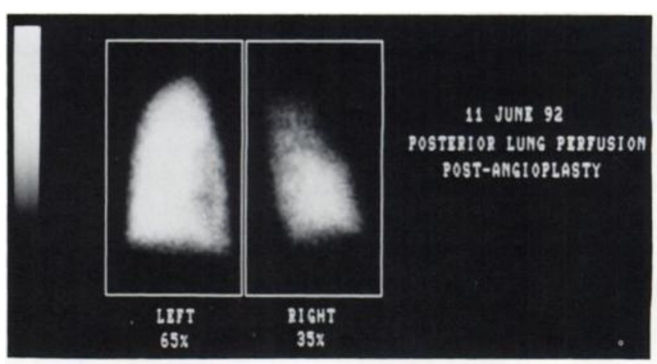

FIGURE 4. Perfusion study post-angioplasty. Perfusion ratio does not improve to unity as the angiogram alone would have suggested.

pulmonary pressures, the flow is increased proportionately, i.e., R = ΔP/Q. In theory, such a vascular bed has the potential to again carry essentially half the cardiac output when relieved of any obstruction at it's origin. In practice, however, the rehabilitated lung will initially receive less than one-half the cardiac output since the nonaffected side has developed capacious vessels and recruited new capillaries. Our experience with this patient demonstrates that in the presence of longstanding unilateral discrete obstruction, perfect relief of the stenosis over the short-term will result in no more than 35% of the cardiac output entering the rehabilitated lung. Interestingly, over a 1-yr period the flows in the right and left lung equalized.

Our patient was studied in the 10-day period postangioplasty, not only to evaluate the result of the angioplasty but also to establish a baseline for this patient so that any restenosis might be measured as a change in perfusion on future scans. We suggest yearly scans initially. No change on the perfusion study for 3 yr might be an indication to perform studies less frequently since this would imply no restenosis is occurring.

Nuclear perfusion scans represent a highly sensitive and specific modality for the evaluation of discrete acquired peripheral pulmonary stenosis both before and after angioplasty. The procedure is cost-efficient and has minimal morbidity. Previous studies have shown MAA perfusion scans to be consistent and accurate (*12,15*) with ±3% accuracy (*23*). Results have been reproducible by different technologists on the same and different scanners (*11*). The cost of the study is about the same as a less sensitive color Doppler study and exposes the patient to little more radiation than the highly insensitive chest x-ray, which can be deferred.

Postsurgical (acquired) peripheral pulmonary stenosis is not an uncommon lesion that can deprive the patient of functional use of the involved lung. Quantitative nuclear perfusion imaging provides a cost-effective, rapid, noninvasive assessment of the stenosis in the patient at risk. In our patient, it was superior to echocardiography, angiography and pressure measurements.

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