

CASE REPORT

Radionuclide Angiographic Demonstration of Systemic Lung Arterialization with Arteriovenous Fistulas

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Radionuclide angiography has diagnosed systemic arterialization of the right lung base in a patient presenting with a basal thoracic bruit. The bruit was due to high flow in the systemic artery and development of arteriovenous fistulas, confirmed by TCT scan and contrast aortography. But the parenchyma of the right lung base appeared normally aerated on the radiographic studies, and Xe-133 ventilation scintigraphy was normal. This case was therefore classified as systemic arterialization of lung without sequestration.

J Nucl Med 24: 228–230, 1983

The value of radionuclide angiography has been reported to demonstrate systemic arterialization in bronchopulmonary sequestration (1,2). The case described here constitutes a particular entity of lung systemic arterialization of a normally aerated and ventilated pulmonary base, presenting with a basal thoracic bruit. Radionuclide angiography achieved the diagnosis, which was confirmed by other investigations.

CASE REPORT

A 32-yr-old black man was admitted after the fortuitous discovery of a continuous bruit heard posteriorly at the right thoracic base. He had no symptoms and was in excellent physical condition. The routine annual chest radiograph had always been normal. He was a sports instructor and professional basketball player.

On admission, the chest radiograph was virtually normal in the postero-anterior projection (Fig. 1A). But the right anterior oblique view showed small ill-defined rounded shadowings in the lower part of the right pulmonary base, especially in the costo-diaphragmatic angle (Fig. 1B). A faint "band-like" density extending below the right hemidiaphragm parallel to the vertebral column was not initially noted, even though it could be considered (a posteriori) as a vascular structure, and the preliminary diagnosis of pulmonary arteriovenous fistulas was made.

A radionuclide angiographic study was performed after intravenous injection of a bolus of 12 mCi of pertechnetate (Tc-99m) with the patient in an upright position. A posterior view was ob-

tained with a gamma camera using a high-resolution collimator. It demonstrated a lack of perfusion of the right lung base during the pulmonary phase. This area was perfused by the radiotracer only during the systemic phase, with activity suggesting hypervascularization (Fig. 2). This suggested a diagnosis of systemic arterialization of the right pulmonary base with poor or absent perfusion from the inferior branches of the pulmonary artery. The activity in images during right basal perfusion tended to spread upwards with time. This could indicate that the venous drainage followed the pulmonary veins. The observed persistent visualization of left-heart cavities and aorta tended to confirm this direction of venous drainage.

The TCT scan without contrast enhancement showed polylob-

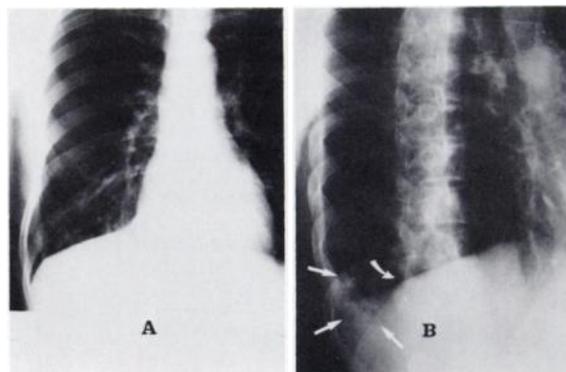


FIG. 1. Chest radiograph. (A) Postero-anterior projection is virtually normal. (B) Right anterior oblique projection shows small rounded shadowings of pulmonary base (straight arrows) and a "band-like" density parallel to vertebral column (curved arrow).

Received July 19, 1982; revision accepted Nov. 15, 1982.

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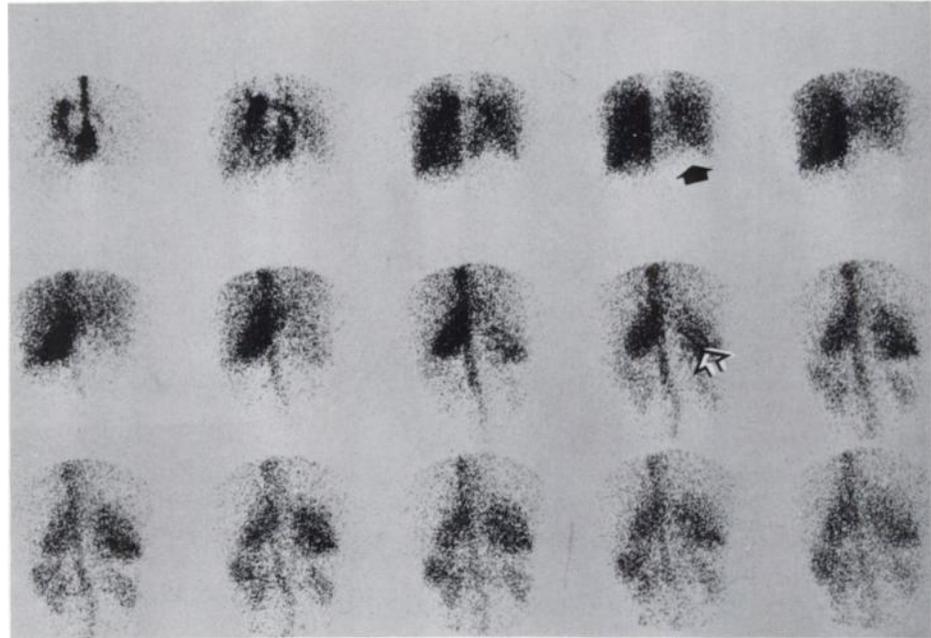


FIG. 2. Series of 1.3-sec radionuclide angiographic images (posterior view) showing lack of perfusion of right lung base during pulmonary phase (closed arrow) and hyperperfusion of same area during systemic phase (open arrow). Right basal activity then tends to spread upwards. There is persistent visualization of left heart chambers and aorta.

ular densities of the right lung base. A TCT scan was obtained using a bolus contrast enhancement, with scanning timed to coincide with the arterial phase. It showed an abnormal, dilated artery running along the right anterolateral aspect of the vertebrae, arterial structures in the right costo-vertebral angle, as well as flow into the polylobular structures of the pulmonary base (Fig. 3).

Pulmonary arteriography demonstrated a reduced number of inferior segmental branches of the right pulmonary artery (Fig. 4). The rest of the pulmonary arterial tree was unremarkable. Normal pulmonary artery pressures were obtained and there was no evidence of left-to-right shunt.

A contrast aortogram demonstrated a large anomalous artery arising from the aorta, which was considered to be an inferior diaphragmatic artery (Fig. 5). This vessel supplied the right pulmonary base, revealing multiple vascular structures that appeared as arteriovenous fistulas. Large veins were seen but their destination could not be established.

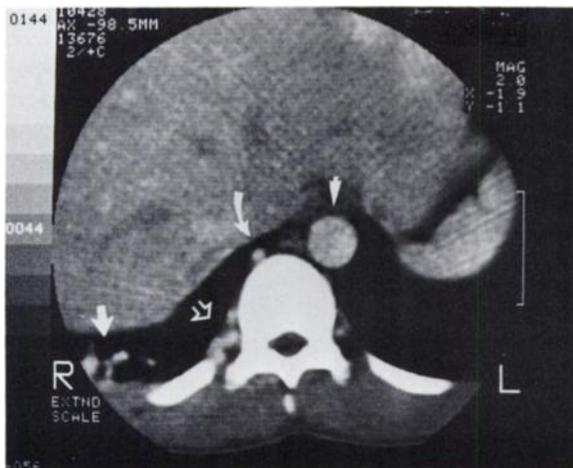


FIG. 3. TCT scan with bolus contrast enhancement shows anomalous artery (curved arrow) perfused at same time as the aorta (arrow head). Also shows arterial structures in right costovertebral angle (open arrow) and contrast enhancement of polylobular structures in right pulmonary base (straight arrow).

A Xe-133 ventilation scintigraphic study was subsequently performed using a posterior view, with the patient sitting against the gamma camera. A single-breath inhalation image was first acquired. After 3 min of rebreathing, an equilibrium image was obtained and then 30-sec washout images were made for 5 min. The three phases of this ventilation study were normal (Fig. 6). A lung scan with Tc-99m albumin microspheres showed perfusion defects of the posterior and lateral segments of the right pulmonary base.

The patient was discharged without treatment, but was requested to reattend regularly.

DISCUSSION

Several congenital pulmonary pathological conditions have been reported concerning anomalous systemic arterialization. The commonest is bronchopulmonary sequestration. There have also

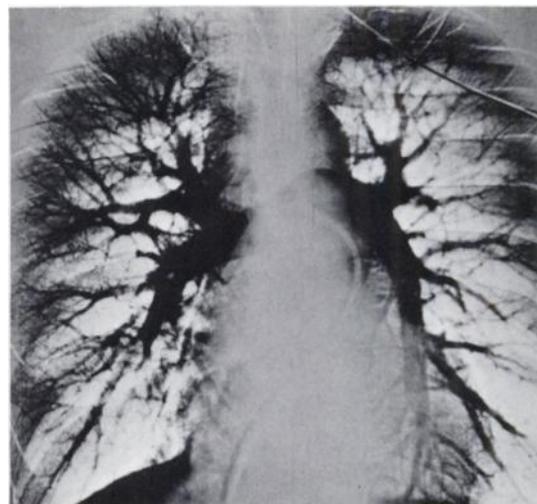


FIG. 4. Pulmonary angiogram shows reduced number of inferior segmental branches of right pulmonary artery.

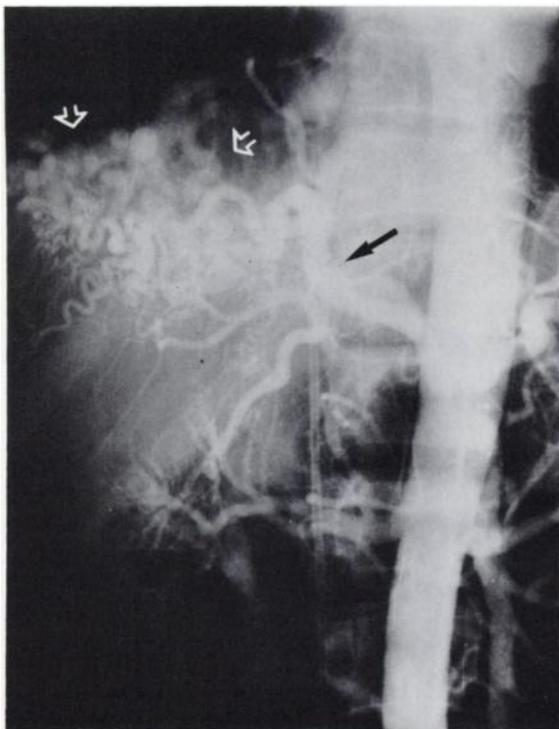


FIG. 5. Contrast aortography shows dilated anomalous artery (closed arrow) arising from aorta. It supplies arteriovenous fistulas (open arrows) in right pulmonary base.

been described the scimitar syndrome and "systemic arterialization of lung without sequestration" (3,4), which is the rarest form.

The "sequestration complex" (4) as first described by Pryce et al. (5) includes several clinico-pathological findings, which have in common an anomalous systemic arterialization of the lung, with atresia, or hypoplasia of the pulmonary artery. The most common form of the sequestration complex is bronchopulmonary sequestration. This congenital malformation consists of a nonfunctioning portion of lung, most often a lower-lobe segment, completely (extralobar) or incompletely (intrapulmonary) separated from the rest of the lung. Characteristically the sequestered segment is a closed system not connected to the normal bronchial tree, but communication may develop as a result of infection in the sequestered portion of the lung (6). The rarest form of the sequestration complex consists of an anomalous systemic arterial supply of a normal unsequestered lung region. Very few cases have been reported (3,4). This last entity was thought to be present in our patient, since the pulmonary base was normally aerated without any anomalous parenchymal density on the chest radiograph and TCT scan. These studies only showed rounded opacities representing arteriovenous fistulas and a medial vascular structure that corresponded to the dilated anomalous systemic artery, arising from the abdominal aorta. Furthermore, normal ventilation of the right pulmonary base was demonstrated by the Xe-133 ventilation scan.

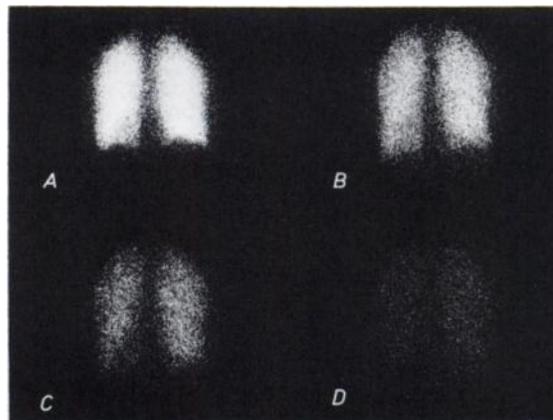


FIG. 6. Xe-133 ventilation scintigraphy (posterior view) shows normal ventilatory function of right pulmonary base. (A) Single-breath inhalation image. (B) Equilibrium image. (C and D) Wash-out images obtained respectively at 0-30 sec and 30-60 sec.

Hence, our observation can be considered as a case of systemic arterialization of lung without sequestration. The radionuclide angiogram indicated the diagnosis: there was a lack of right basal activity during the pulmonary phase. This was later explained by the absence of the most of the inferior segmental branches of the pulmonary artery, which was revealed by pulmonary angiogram. Conversely, there was intense right basal activity during the systemic phase of the radionuclide study. This was due to extensive systemic arterialization and the presence of arteriovenous fistulas as demonstrated by the contrast aortogram.

The case reported here is particularly interesting for the unusual mode of discovery of the lung arterialization. It presented as a continuous bruit in an asymptomatic patient. Radionuclide angiography and ventilation scintigraphy gave the final diagnosis, which was confirmed by invasive vascular radiological studies.

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