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Radionuclide Angiographic Diagnosis of Bronchopulmonary Sequestration

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Bronchopulmonary sequestration (BPS), a congenital malformation that usually presents as a chest mass in childhood, may be identified by its characteristic primary derivation of pulmonary blood supply from the systemic circulation. Five children with BPS were evaluated by radionuclide angiography from 1970 to 1974. In each instance the systemic origin of the vascular supply was correctly indicated. In those lesions where the artery originates below the hemidiaphragm, the aberrant source, when identified as such, provides a characteristic radionuclide appearance of BPS. The scimitar syndrome may be indistinguishable from BPS with this technique.

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The diagnostic evaluation of a chest mass in the pediatric age group poses a problem because of the variety of congenital abnormalities that may be encountered. Diagnosis, particularly in the infant, assumes greater importance as surgical correction of congenital anomalies, such as bronchopulmonary sequestration (BPS), becomes more common. Intrusive or potentially dangerous diagnostic procedures have often been required to evaluate such abnormalities since accurate preoperative definition of blood supply is needed to reduce the hazard of hemorrhage during arterial ligation (*1*) and to permit a planned surgical approach to the lesion.

The roentgenographic diagnosis of BPS depends on the availability of facilities for special studies, such as pneumoperitoneography (*2*), tomography (*3*), bronchography (*4*), and contrast angiography (*5*). Chest masses in children have been evaluated by radionuclide angiography with ^{99m}Tc as pertechnetate (*6*), and bronchopulmonary sequestration has been diagnosed with this technique (*7*). Prosin et al. described a radionuclide technique using ^{133}Xe, ^{99m}Tc-labeled iron hydroxide particles, and ^{113m}InCl to evaluate this condition in an adult (*8*). The present report describes further experience in the use of the single-radionuclide technique for evaluating BPS.

MATERIALS AND METHODS

Between 1970 and 1974, two female and three male children with BPS were studied with radionuclide angiography at the Children's Memorial Hospital. Their ages varied from 2 days to 11 years (Table 1). The patients were sedated with a Demerol–Phenergan–Thorazine mixture when necessary (*7*) and a peripheral vein was injected with ^{99m}Tc as pertechnetate, 100 μCi/lb. A bolus effect was obtained by rapidly flushing the syringe with normal saline.

Dynamic radionuclide imaging followed immediately with a scintillation camera equipped with a high-sensitivity collimator and data storage device. The lesions were defined from chest roentgenograms and the patients were specifically positioned to display these areas to their best advantage. The stored images were retrieved and displayed on Polaroid films for selected time intervals which enabled best visualization of all phases of the vascular perfusion to the abnormal area and the surrounding lung.

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TABLE 1. SYMPTOMS AND FINDINGS IN FIVE PATIENTS WITH BPS

Case No.	Age	Sex	Symptoms	Chest x-ray findings	Surgical diagnosis	Origin of arterial supply
1	2½ weeks	F	Fever with rapid breathing and wheezing	Ill-defined homogeneous density in left base	BPS (extralobar)	Abdominal aorta
2	2 days	F	Difficulty in breathing since birth	Poor aeration of right base with haziness and nonaeration of apex	BPS in monolobate right lung	Abdominal aorta
3	6 days	M	Fever with apneic spells	Right lower lobe consolidation	BPS	Aorta
4	22 days	M	Fever and poor feeding	Consolidation of left lower lobe and right upper lobe	BPS (extralobar)	Aorta
5	11 years	M	A density was seen in the left base near costovertebral angle on routine workup for rapid breathing	Density behind the heart in the left base	BPS with bronchogenic cyst	Thoracic aorta

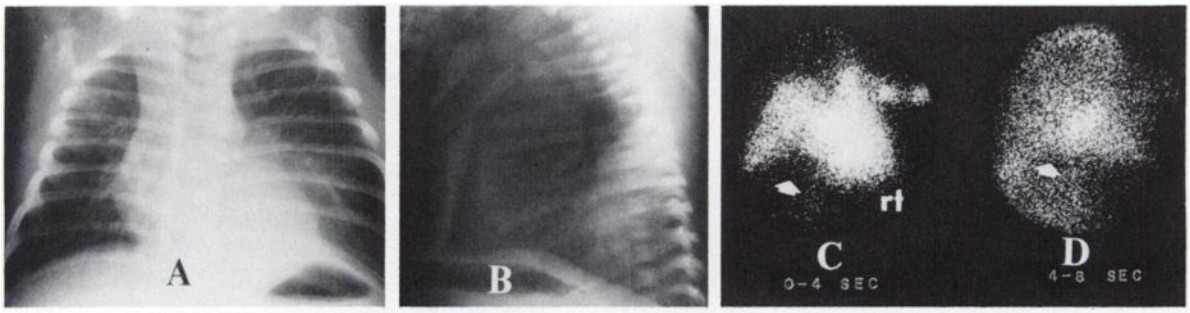

FIG. 1. (A and B) Soft-tissue mass in left lower lobe (arrows). (C) Radionuclide angiogram, posterior projection, during pulmonary phase showing nonperfused area of left base (arrow) corresponding to soft-tissue mass. (D) Perfusion to this area occurs rapidly during early systemic phase (arrow) of radionuclide angiogram. See Case 1 in Table 1.

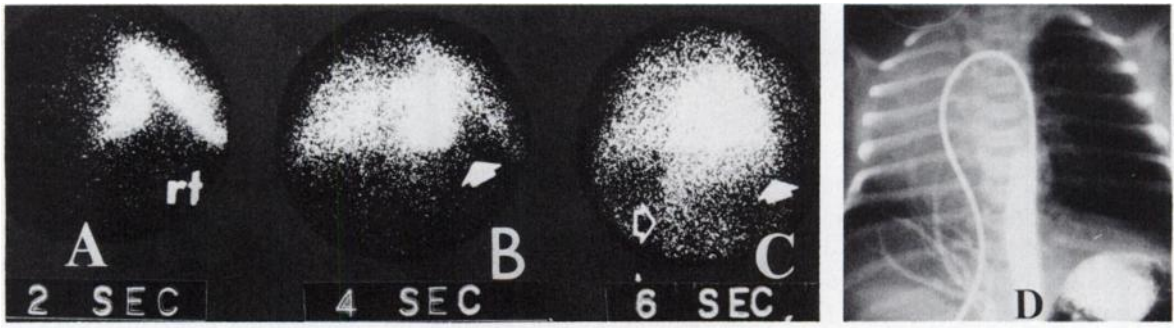

FIG. 2. (A) Radionuclide angiogram, posterior projection, shows initial circulation through right side of heart. (B) Pulmonary phase, with nonperfused base of right lung (arrow). (C) Perfusion of right lung base during systemic phase (closed arrow). Blood vessel supplying area arises from abdominal aorta (open arrow). (D) Contrast arteriogram illustrating primary systemic blood supply to area of bronchopulmonary sequestration. See Case 2. (Reproduced by permission of Ref. 7.)

RESULTS

In all cases of BPS, no perfusion was noted in the area of the roentgenographic abnormality during the pulmonary phase. Subsequently, as the radionuclide entered the left ventricle and aorta (systemic phase), rapid perfusion to the abnormal areas was seen, indicating predominance of vascular supply from the systemic circulation (Fig. 1). In one child (Fig. 2), the aberrant blood supply was seen arising infradiaphragmatically from the abdominal aorta, a finding almost pathognomonic for BPS.

This dynamic sequence in a patient with a chest

mass provides presumptive evidence of BPS. The diagnosis was confirmed surgically in all of these children, three of whom underwent surgery without contrast-angiographic studies.

DISCUSSION

Bronchopulmonary sequestration may be classified as intra- or extralobar (*4*). The extralobar type involves an ectopic lobe of the lung, situated above or below the diaphragm and enclosed in its own visceral pleura. The intralobar type involves a malformation of a perfused, but often unventilated, lung segment sequestered within the normal lung without its own visceral pleura. The distinctive feature of both types is the systemic arterial blood supply that often arises from the descending thoracic aorta or the abdominal aorta.

Bronchopulmonary sequestration frequently presents in childhood as a respiratory problem that may be life-threatening. The children are often very ill and are not favorable candidates for extensive roentgenographic procedures, such as pneumoperitoneography, bronchography, or contrast angiography. These techniques carry minimal but well-known morbidity, such as air embolism, increased respiratory difficulties, and circulatory overload.

The radionuclide diagnosis of BPS depends on the demonstration of nonperfusion of the lesion during the pulmonary phase of the radionuclide angiogram, followed by a rapid perfusion of the area during the early systemic phase. Alterations in circulatory dynamics associated with pneumonia or such neoplasms as neuroblastoma also can show decreased or absent perfusion during the pulmonary phase. However, the appearance of perfusion during the first few seconds of the systemic phase is more suggestive of BPS since there is a direct rather than a collateral blood supply. Other abnormalities may also have a rapid systemic arterial blood supply. Three specific conditions of importance in the differential diagnosis are intrathoracic kidney, hepatic herniation through the diaphragm, and anomalous pulmonary venous drainage (scimitar syndrome). Conditions involving abdominal organs herniated into the thorax are easily identified with radionuclide renal imaging and liver–spleen imaging. The scimitar syndrome, however, may be indistinguishable from BPS with radionuclide techniques and, indeed, BPS may even be a component of the syndrome (*4*).

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