Bronchial and Arterial Anomalies with Drainage of the Right Lung into the Inferior Vena Cava

By NICHOLAS A. HALASZ, M.D., KATHERINE H. HALORAN, M.D.,
AND AVERILL A. LIEBOW, M.D.

Three cases of anomalous pulmonary venous return to the inferior vena cava are presented and 18 previously reported cases are also analyzed. The clinical, radiographic, circulatory and pulmonary function, and anatomic features are carefully detailed. In particular, associated vascular and bronchial anomalies in the lungs were found by preparation of vinylite casts of the pulmonary vessels and bronchial casts; they are of great significance in planning and executing surgical therapy.

INTEREST in major anomalies of pulmonary venous drainage has mounted with the development of methods for accurate clinical diagnosis that have brought the possibilities of corrective surgery into serious consideration. There is a need, therefore, for a careful anatomic study. It is important, also, to determine the incidence and nature of any associated anomalies that might affect therapy.

Anomalous drainage of the pulmonary veins, meaning thereby functional deviation of blood into the systemic venous system, whether by way of a septal defect or otherwise, must be distinguished from anomalous connection, which may imply drainage directly into the right atrium, or some tributary, but may also indicate drainage into the left atrium, by way of an unusual venous channel or postatrial chamber. The basic facts regarding anomalous venous connections of the lungs were collected and excellently analyzed by Brody in 1942. It would appear that total drainage of both lungs into the right atrium or a major tributary occurs half as frequently as partial drainage. In the former, there is always an associated septal defect. With the latter, the drainage is most commonly into the superior vena cava, right atrium, or left innominate vein. In Brody's table, drainage into the inferior vena cava was relatively uncommon, occurring only twice in a total of 65 instances of partial anomalous drainage.

In each of 3 instances of total drainage of the right lung into the inferior vena cava recently observed by the writers, a part of the arterial supply stemmed from the aorta, and there were also associated bronchial anomalies. Not only were there deviations in the pattern of arrangement of the bronchi, but actual abnormalities in structure of a diverticular nature were encountered.

Many of these observations have been reported in isolated instances of this venous anomaly, but that they are commonly associated is suggested by a detailed study. The earlier reports dealt simply with anatomic material. Only since 1949 have more recently developed methods been applied that permit an accurate clinical diagnosis of this condition. In the present report, data from our 3 patients (see Case Reports and figs. 1–27) will be compared with previously published observations (see table 1). Some published accounts are, unfortunately, rather fragmentary. Certain cases have been cited repeatedly, sometimes with the result that various items of information have had to be pieced together from more than one source.

CLINICAL FEATURES

General. Among the 21 patients under survey, approximately two-thirds were male. At least 6 of the group had no symptoms whatever.
Table 1.—Drainage of Right Lung into Inferior Vena Cava, Clinical and Anatomic Data

<table>
<thead>
<tr>
<th>Source</th>
<th>Age</th>
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<th>Symptoms</th>
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Note: Only findings recorded as positive are indicated.

Key: SOB—Shortness of breath
     Dex.—Dextrocardia
     EH—Cardiac enlargement
     PA—Enlargement of main trunk of pulmonary artery

Three had severe associated congenital cardiac lesions, in 1 of whom, with transposition of the great vessels, the drainage of the pulmonary veins into the right heart was actually desirable functionally. Only in a few patients were there symptoms and signs that might be ascribed to the venous or associated bronchial anomalies. Five of the patients had shortness of breath, 1 only upon severe exertion. Our 3 patients had repeated respiratory infections and another, reported by Park,* died of pneumonia early in life.

The most constant finding upon physical examination was evidence that the heart had been shifted to the right. There was, in some instances, electrocardiographic evidence of right ventricular dominance. There was also, frequently, diminution of resonance in the right hemithorax. In the absence of cardiac anomalies, cyanosis was not present except episodically in the first few days of life in some patients. Pneumonic episodes were attended with appropriate physical signs.

Radiographic Findings. In the plain films, the most striking appearance, which should at once suggest the diagnosis, was that of a dagger or scimitar-shaped vascular density more or less parallel to the right cardiac border, but tending to deviate laterally into the lung field near its tapering upper end (figs. 1, 11, 20). The right hemithorax frequently appeared small with narrowed intercostal spaces and a deviation of the heart to the right. The right cardiac border and, in particular, the right atrium, in appropriate oblique views and at fluoroscopy, appeared prominently enlarged, as did the main trunk and left branch of the pulmonary artery (fig. 11). The vascular shadows can be made more prominent by section radiography.

Angiography proved to be useful in demonstrating the opacification of the paracardiac
vascular shadow simultaneously with the pulmonary veins of the contralateral side (figs. 3, 14, 23). The enlargement of the right cardiac chambers was strikingly demonstrated, as were the small right and expanded main and left pulmonary arteries (figs. 3, 13). Evidence of recirculation was the long-maintained opacification of the right cardiac chamber to a time when the pulmonary veins of the left side, and even the aorta, had become opacified (figs. 4, 15, 22, 23). In late films of the angiographic series in 1 of our patients, large arterial vessels were demonstrated to enter the lung from below (fig. 15). The angiographic series have been analyzed to give the temporal sequence of hemodynamic events in each patient (Case Reports).

Bronchography is useful to demonstrate anomalies in distribution and structure of the bronchi. One or more irregular outpouchings were demonstrated in 5 patients, including all 3 of those in the present series (figs. 2, 12, 21). In 2 of our patients the right and left main bronchi were symmetrical, and it was thought that the upper lobe, or both upper and middle lobes, were hypoplastic or absent. However, upon review in the light of the bronchovascular casts prepared in each instance, it was apparent that there was, in fact, a mirror image of the left lung present on the right side (figs. 2, 12). In one other instance, the condition was diagnosed as "hypogenesis."17 In case 3 of the present series, there was a notably hypoplastic eparterial right upper lobe, compensated in part by accessory bronchi from the medial segment of the large middle lobe (figs. 21 and 25).

Functional Studies. Although cardiac catheterization is not necessary to establish the diagnosis, it is of interest in providing confirmatory evidence of the presence of fully saturated blood within the anomalous vein, or at least of an unusually high oxygen saturation of the blood in the right cardiac chambers.

Some estimate of the magnitude of the recirculation through the right side can be obtained by various methods. If the entire right lung drains into the right side of the heart, and if there are no complicating anomalies on the arterial side, such as accessory pulmonary arteries from the aorta, bronchospirometry will provide information on the relative effective blood flow through the 2 lungs, on the assumption that this is directly proportional to the oxygen uptake of the respective sides. The actual volume of the shunt can be calculated by applying the Fick principle using the oxygen consumption of the normal left lung in the numerator and the difference in oxygen contents between the mixed venous blood in the pulmonary artery and the systemic arterial blood in the denominator. The volume of recirculated blood can then be obtained by subtracting this figure from the right ventricular output, which is obtained by using the total oxygen consumption. Bronchospirometry was performed only in the patient described by Cooke and co-workers.18 The anomalous right lung in this case accounted for 48.8 per cent of the total oxygen consumption, but it must be noted that there was evidence that only the right lower lobe drained into the right atrium and there was, in addition, at least 1 accessory pulmonary artery from the aorta. In this instance, an attempt was made to estimate the volume of the recirculation to the right lower lobe, but the Fick principle was not correctly applied.

In the absence of bronchospirometric data, a range of values for the relative volume of the recirculation can be obtained by applying a mixing formula, for which the following data are necessary, stated in volumes per cent or per cent oxygen saturation:

1. $S = \text{oxygen content of mixed systemic venous blood (before admixture has occurred in the atrium with the blood brought in by the anomalous vein).}$

2. $T = \text{oxygen content of blood in the anomalous vein.}$

3. $M = \text{oxygen saturation of blood in the pulmonary artery (presumably after a thorough mixture of } S \text{ and } T \text{ has occurred).}$

Now let $x =$ the volume of blood arriving from the anomalous vein for every ml. of mixed systemic venous blood. Then, $S + xT = M(x + 1)$. Or $x = \frac{M - S}{T - M}$.

Since $S$ cannot be obtained unless the
relative inflow from the superior and systemic component of the inferior vena cava are known, a very crude approximation can be obtained by assuming the inflow to be equal from these 2 vessels, and averaging the oxygen contents of the superior and inferior vena cavae (from a point distal to the entry of the anomalous vessel). A less spurious way is to calculate the possible range of values by using as \textit{S}, first, superior vena caval oxygen content and then, separately, the inferior vena caval saturation in the formulas given above. On this basis, the patient reported by Mankin and Burchell\textsuperscript{17} (using data supplied in their fig. 2) had recirculation through the right lung amounting to somewhere between 49.8 and 21 per cent of the volume of blood entering the right heart, or of 39 per cent, assuming an equal systemic inflow from the 2 cavae. Mankin and Burchell,\textsuperscript{17} however, estimated the magnitude of this shunt as 30 per cent, on the basis of an unstated method.

Further evidence of recirculation through the right lung in the same patient was obtained by Swan, Burchell, and Wood\textsuperscript{20} from the study of concentration curves of Evans blue dye in the peripheral vessels, when injected into the right and into the left pulmonary arteries respectively. When applied to the same patient, recirculation was estimated variously as 40 and 35 per cent, values that are within the range calculated in the preceding paragraph.

Whatever its magnitude, the volume of blood recirculated through the right lung in this way is a burden on the right heart, of which the output exceeds that of the left by the magnitude of the shunt.\textsuperscript{21} It is of interest that this burden is not usually sufficient in itself to produce cardiac failure, and that most patients with this anomaly can reach adult life.

The blood volume was determined in only 1 patient and was found to be actually less than expected.\textsuperscript{15} This result is surprising, inasmuch as patients with a large left-to-right shunt, as through an arteriovenous fistula, usually have an expanded blood volume.\textsuperscript{22}

The magnitude of the left-to-right shunt resulting from the accessory pulmonary arterial supply from the aorta has not been measured in these patients, but this must constitute a further increase in the work of the right heart. From observations on experimentally induced collateral circulation to the lung, it appears that even considerable transpulmonary shunts from the aorta through the lung do not significantly affect the thickness and capacity of the left cardiac chambers.\textsuperscript{23}

**Anatomic Features**

\textit{Course of the Anomalous Vein.} From its termination in the inferior vena cava at the medial and inferior angle of the lobe, the anomalous vein extends upwards and laterally. It may receive its tributaries as small twigs radially arranged at various levels in the manner of a fir tree, and it may not itself branch so as to permit the delimitation of lobes (figs. 6, 8, 16, 17). On the other hand, the vessel may branch near its origin, as in case 3, so that the usual number of lobes can be delimited by penetrating fissures (figs. 26, 27).

\textit{Arrangement of the Bronchi.} All 3 of the specimens studied display major deviations from the norm in the arrangement of the bronchi. In 2 instances, the pattern is close to that of a mirror image of a left lung (figs. 5, 8, 17). This is suggested even in the bronchogram by the long course and symmetry of the first divisions of the trachea before the upper lobe bronchi are derived. In the casts of cases 1 and 2, it is apparent that the right upper lobe bronchus is hyparterial and that there is a lingular bronchus, but no middle lobe bronchus.

It may be asked whether this arrangement may not represent an upward expansion of lower lobe bronchi, and more specifically those of the superior segments, in consequence of agenesis of the upper and middle lobes. The bronchus interpreted as that of the upper lobe, however, not only possesses branches typical for this lobe, but is, moreover, in both cases 1 and 2, situated anterior to the main trunk of the pulmonary artery, quite the opposite of the relations of a superior segmental bronchus (figs. 7, 17, 18). In case 1, there is an azygos lobe (fig. 9). This is simply a part of the upper lobe separated from the remainder...
by the penetrating mesoazygos, but not otherwise disturbed.\textsuperscript{24, 25}

In case 3 there is an eparterial upper lobe bronchus, although it lacks anterior branches. These are replaced by upward expansions of rami from the medial branch of the middle lobe bronchus (figs. 25, 27).

**Bronchial Diverticula.** In each of the specimens there is a bronchial diverticulum. The largest of these is the bifid blindly ending pouch-like diverticulum of case 1 in the position that the medial basal bronchus might occupy were it present (figs. 5, 7). In case 2, there is a small pit corresponding to the position of the anterior segmental bronchus that is not represented as such, but compensated for by some expansion of the branches of neighboring segmental bronchi (fig. 17). This compensation probably took place early in embryonic development, since all regional pulmonary arteries or veins accompany compensating bronchi. In case 3, the aneurysmal widening of the section of the lateral segmental bronchus of the middle lobe might be considered artifact, were it not clearly demonstrated in the bronchogram (figs. 21, 26, 27). From this expanded region there bronchial branches of the usual size and distribution.

Although there is no histologic proof, it is probable that these diverticula represent the seat of chronic suppuration that accounted for much of the symptomatology.

**Dextrocardia.** The dextroposition of the heart results simply from the mediastinal shift occasioned by the small size of the right lung.

**Associated Arterial Anomalies.** It is remarkable that, in each of the right lungs of the present series, there were several arteries derived from the aorta as well as a right pulmonary artery of diminished size. The aortic branches were demonstrable in angiograms of 1 patient of the present series (fig. 15), but have previously been noted only at operation or necropsy.

Knowledge of the existence of such vessels is valuable not only in suggesting the presence of left-to-right recirculation, and therefore the relative uselessness in respiration of tissue thus supplied, but also in being forewarned in anticipation of an operative procedure. Since these large arteries often lie within the pulmonary ligaments and in the substance of adhesions, there is a danger that they may be carelessly transected. Such an event may even be fatal.

These arteries substitute for pulmonary arteries of particular segments or subsegments, without overlap. These branches from the aorta, although often beginning their approach to the bronchi from an antihilar direction, yield branches that turn to pursue the direction of and to branch with the smaller bronchial rami, one artery being distributed to each bronchus (fig. 19). They gradually become attenuated with the bronchi to their terminations, in the manner of true pulmonary arteries. These they approximate not only in size, but also in being truly end arteries. There is no evidence that the walls of bronchi are vascularized by them before alveoli appear. In all of these features, they differ from the bronchial arteries, which are generally multiple and plexiform in arrangement and relatively small as compared with the bronchi, the walls of which they richly supply.

**Discussion**

The very fact that bronchial and vascular anomalies appear to be associated would suggest a common factor in their genesis. No ready explanation, however, presents itself.

The embryologic background for the occasional drainage of the lungs into tributaries of the right atrium is considered to be the presence of a common capillary bed for the ultimate pulmonary veins and branches of the cardinal and umbilicovitelline venous systems, seen most clearly at the early stage when the lungs are represented simply as a diverticulum, soon to become tubular and branching, of the primitive foregut.\textsuperscript{26} What is not known are the forces that produce the deviation of drainage, in the present instance unilateral and into the ultimate inferior vena cava. It seems unlikely that small bronchial diverticula could exert any influence upon the drainage of an entire lung or that, on the contrary, the huge anomalous vein could produce these small localized anomalies, while the remainder of the respira-
tory tract remained structurally normal. A vestige of the original interrelationships of the pulmonary and systemic venous systems is the free connection of the bronchial and pulmonary veins in the hilum in the normal lung.

Any aortic blood supply to the lungs is considered, on the basis of embryologic studies, to be derived from the primordial postbranchial plexus of the descending thoracic aorta. In the normal course of development this plexus persists as bronchial arteries. In the anomalous lungs of the present series, perhaps the fault lies in the incomplete development of the pulmonary artery of the left side by terminal accretion from components of the postbranchial pulmonary plexus. If some of the latter should fail to contribute to the growing extension of the sixth arch, but retain their original connections with the aorta, the ultimate structure would be that actually existing in the specimens under study.

Similar large branches of the aorta have been described to supply “sequestrated” pulmonary tissue and indeed the question of sequestration may well arise in view of the diverticular anomalies that have been described. In all 3 instances, however, the anomalous portions of the bronchial tree are not supplied by these aortic branches, but rather apparently normal segments or subsegments. In fact, the present instances supply additional evidence that anomalously developed arteries from the aorta need not result in “sequestration” neither from pressure upon the bronchi nor by any phenomenon of traction, but that they can represent an orderly substitution from the dorsal aorta for vessels normally developed in continuity with the more proximal sixth aortic arch.

Partially, or completely, corrective surgery has been practiced in a few instances of major anomalies of connection of the pulmonary veins. The presence of an associated interatrial septal defect has actually been helpful in some instances in meeting the anatomic problems. Surgical redirection of pulmonary venous blood originally draining into the inferior vena cava from the right lung to the left atrium has been reported only once, in a patient with an associated septal defect.

In this instance, the ingenious solution to the problem was to transplant the anomalous vessel to a higher position in the right atrium opposite the defect, and to isolate by suture as a pocket separate from the right atrium opposite the defect, the transplantation site and defect. No other ready solution presents itself in the case of this particular anomaly, since the vein is not sufficiently long to be connected directly to the left atrium. Grafts to increase the length of a vessel for anastomosis are to be considered, but experimental work is needed to determine whether such grafts would take and function without thrombosis under the low pressure gradient prevailing, and in close relation to the actively beating heart.

Study of the present material would suggest certain additional precautions and indications in surgery. It is customary to occlude the pulmonary artery of a lung before preparing the vein for anastomosis. When a lung is supplied by aortic branches, special precautions must be taken, since a very high pulmonary intravascular pressure may otherwise develop. What the fate of the lung might be were these arteries to be permanently occluded is not known. Pneumonectomy should be considered as possibly the best procedure, since the presence of these vessels implies the existence of a left-to-right shunt, since in a considerable proportion of instances, there are also bronchial diverticula, and since the lung is hypoplastic as well.

Case Reports

Case 1

Clinical History. Immediately following birth and on a few occasions during the first year, there were episodes of cyanosis. A physician noted a cardiac bruit at 1 month of age and found, by fluoroscopy, the heart to be shifted to the right. In early childhood there were 2 bouts of pneumonia preceded and followed by chronic upper respiratory infection with cough productive of thick sputum. When exercising heavily during relatively well periods, the patient was noted to have a thin, pale blue line about the lips and to be mildly dyspneic.

The child was first admitted to the Columbia-Presbyterian Medical Center at the age of 4 years, in 1947. He was noted to be retarded physically and to have a congenital coloboma of the left eye and a loud precordial systolic murmur, best heard over the left third interspace. A roentgenogram of
the chest was considered consistent with atelectasis of the right middle lobe and a shift of the heart to the right (fig. 1). At fluoroscopy, there was marked pulsation of the right heart border. An esophagram was read as negative, but a bronchogram showed a right lower lobe bronchiectatic cavity (fig. 2). There was a right axis deviation in the electrocardiogram. Cardiac catheterization demonstrated higher

**Fig. 1.** Case 1. Plain film. The band-like shadow of the anomalous vein is visible (arrows). The fissure demarcating the azygos lobe is seen as a well defined white line extending to the right and upwards from the mediastinum. The heart has a prominent right border and is dextroposed. The right hemithorax is smaller than the left.

**Fig. 2.** Case 1. Bronchogram. The branching of the trachea is symmetrical and the bronchi of the right side are close to a mirror image of the left. On the right there is a lingula (Li) rather than middle lobe (compare with left side). Projecting downwards and medially from the right lower lobe bronchus is a bifid diverticulum (D).

**Fig. 3.** Case 1. Angiogram. The trunk of the pulmonary artery and its right branch (RPA) are both large. An interventricular septal defect may have contributed to this condition. The anomalous pulmonary vein of the right side (arrows) is opacified simultaneously with the left (arrows).

**Fig. 4.** Case 1. There is persistent opacification of the right cardiac chambers and pulmonary vessels at a time when the contrast medium has already reached the aorta (Ao), indicating recirculation of blood through the lungs. The anomalous left pulmonary vein is indicated by arrows.
O₂ values in the right heart than usual, and hypertension in the right heart and pulmonary artery. These observations, taken together with the murmur, were considered to indicate the presence of an interventricular septal defect. The catheter was, unfortunately, not passed into the inferior vena cava. An oblique right apical shadow seen in anteroposterior chest films was interpreted as thickened pleura between upper and lower lobes. At this time the red blood count was 4.7 million, and the hemoglobin was 13 Gm. per cent. Surgery for what was thought to be congenital heart disease and atelectasis was deferred.

Frequent upper respiratory infections and 1 bout of pneumonia occurred between 1947 and 1950. The question of hypoplasia of the right lung was raised. It was decided to admit the patient for resection of the lobe containing the bilobate cystic structure seen on the bronchogram, which was considered to be the source of recurrent suppuration.

Accordingly, he was readmitted in January, 1950. Physical and laboratory findings were as before. At thoracotomy on February 2, 1950, filmy adhesions were encountered in the right hemithorax. An azygous lobe and another single large lobe were found. When the pleural membrane confining the azygos lobe was divided and the lower large single lobe retracted, a large anomalous pulmonary artery was encountered coming into the right hemithorax at the usual level, but looping over the right main bronchus, which, therefore, was h yparterial. No pulmonary veins were present in the usual sites except for a small vein that drained the azygos lobe into the left atrium. However, one vein 1.5 cm. in diameter, another 1 cm. in diameter, and 2 much smaller veins were noted leaving the anteromedial pleural surface of the large lobe near the diaphragm to empty into the inferior vena cava, 4 to 5 cm. below the right atrium. Also in this region were 4 small vessels that were later shown to be arterial; they also ran to the region of the mediastinum in perpendicular fashion. At this point in the exploration the patient became apneic and convulsive and the procedure was terminated without further surgery.

On February 21, 1950, Diodrast was injected and films taken at half-second intervals with a rapid exposure apparatus. The first film showed filling of the superior vena cava and of a portion of the right atrium. A paramediastinal linear shadow in the left side of the thorax was thought to represent an accessory hemiazygos vein or a persistent left superior vena cava. Following filling of the right atrium, the 1/2-sec. films showed filling of the right ventricle as well. The shadows of the atrium and ventricle were superimposed and could not be delimited in this single projection. It was clear, however, in these films, that the area occupied by the right atrium and right ventricle was only a small portion of the cardiac shadow, perhaps 1/4 to 3/5. On the 1 1/4-sec. film, a large outflow tract, presumably pulmonary artery, was visible with its most cephalic portion at the level of the third thoracic vertebra, just to the left of the midline. A normal left pulmonary artery was seen with normal distribution of its vessels. There was a large shadow, considered to represent a greatly dilated right pulmonary artery taking origin from the main pulmonary artery (fig. 3). Filling of the left side of the heart was not clearly seen until the 3 1/4-sec. films, and the shadow of the aorta was clearly seen 1/2 sec. later (fig. 4). The shadow of the aorta appeared normal radiographically. A conspicuous feature of the remainder of the examination was the persistence of filling of the pulmonary vessels. The large anomalous veins seen at the first operation were not discerned as such. The findings were considered to support the roentgen diagnosis of a greatly enlarged right pulmonary artery and interventricular septal defect. The question of arteriovenous fistula was raised and a second exploration was decided upon.

Through the old incision the right lung was again exposed and a thrill noted in the largest of the veins that drained the lung and entered the vena cava. It was thought that either an arteriovenous fistula or simply rapid flow from high to low pressure areas could produce the thrill. The "cystic" zones could not be felt through the lung substance. Because of the anomalous venous drainage of the lung proper, and because of the small size of the azygos lobe, total right pneumonectomy was carried out. It was necessary to tie and cut several small arteries that passed from the aorta directly into the base of the lung.

The cardiac murmur was not altered in the 6 years since operation nor has the heart shadow changed more than to shift slightly farther to the right. Presumably there is still an intracardiac shunt from left-to-right not associated with abnormalities of the left pulmonary vasculature. The patient is retarded in weight and height. He is dyspneic, but not cyanotic. Pulmonary function studies done in February, 1956, demonstrate a bronchospastic element in the dyspnea. There is a family history of asthma, and this is considered as a more important element at present than either the pneumonectomy or the interventricular septal defect.

Anatomic Observations. Thin adhesions covered the specimen. Only a single fissure, that delimiting an azygos lobe, was present. A vinylite plastic bronchovascular cast was prepared. The vessels were injected first, and a radiograph was taken. Then plastic containing 10 per cent lead sulfate was introduced into the bronchial tree and another radiograph was prepared with greater penetration to demonstrate the bronchial tree (figs. 5, 6). After blocks for microscopic section were taken, the tissue was digested away.

After preparation of the cast (figs. 7-10), it was apparent that the lung was anomalous not only in its...
FIG. 5. Case 1. Roentgenogram of cast. The bronchial tree has been filled with plastic containing lead sulfate, and an exposure to yield appropriate contrast has been made. The upper lobe bronchus (UL) divides into superior (S) and lingular (Ll) divisions. The diverticulum (D) is attached just beyond the origin of the lower lobe bronchus (LL).

FIG. 6. Case 1. Roentgenogram of cast made to demonstrate the vessels before the bronchi had been filled. The two anomalous veins (Va) are shown. The larger traverses the lobe obliquely. In the azygos lobe is another vein which drains normally into the left atrium (Vn). All other vessels shown are arteries.

FIG. 7. Case 1. The cast with diverticulum, labeled for comparison.

FIG. 8. Case 1. The diverticulum removed showing the course of the major veins, labeled for comparison. Arteries (A) are derived in a semicircle from a trunk (not present) that looped over the lobar bronchus.
venous drainage, but in other respects—it had the characteristics of a right-sided left lung, in that a lingula rather than a middle lobe was present, and in that the right pulmonary artery looped over the right main bronchus. A small bifid diverticulum was derived from the inferior division of the right main bronchus. This occupied a position corresponding to that of a medial basal segment. The arterial blood supply of this diverticulum was not apparent, but it may have come from bronchial vessels.

The azygos lobe received bronchial components both from the anterior and apical segmental bronchi. This violation of segmental boundaries by the penetrating mesoazygos is commonly observed. It had its own venous drainage by a vessel that was seen at operation to have drained into the left atrium. Most of the remainder of the lung, including parenchyma supplied both by the upper and lower divisions of the right main bronchus was drained by an enormous vein which was said to have entered the inferior vena cava. There was also a smaller additional venous branch running transversely from the base to the lateral aspect of the lobe, which was said likewise to have entered the inferior vena cava.

In the lateral basal segment, a branch from the aorta, injected with black plastic, was found to have substituted for a branch of the pulmonary artery. This vessel, although it had the course and distribution of a pulmonary artery and the typical relationship of a pulmonary artery to the bronchi, terminated in the pleura in a tortuous plexus of fine vessels, that possessed the characteristics of bronchial arteries. The other branches from the aorta were not injected in this specimen.

**Case 2**

This female child was born on March 16, 1948, after an uneventful gestation. The neonatal period was not remarkable, but toward the end of the third month, she was admitted to a hospital because of episodes of crying and coughing, accompanied by cyanosis of the head and neck. Extensive diagnostic studies were not carried out at this time.

At the age of 6 months, it was found, upon her
readmission to the hospital, that the trachea and mediastinum were deviated to the right. The breath sounds over the right lung were diminished. The heart was thought to be shifted to the right in consequence of the pulmonary difficulty, and to be devoid of intrinsic pathology. The electrocardiogram gave evidence of dextroposition of the heart. The condition was interpreted as hypoplasia of the right lung, possibly with agenesis of the upper lobe. During the preceding 3 months, she had had recurrent episodes of upper respiratory infections with fever. After this admission, the child seemed improved, but remained underweight and continued to have frequent upper respiratory infections.

In January 1954, she was admitted to the New Haven Hospital for further study of the dextroposition of the heart. At this time, vocal fremitus was increased over the right lower chest, but no rales were heard. The heart was found to be shifted markedly to the right by physical examination and fluoroscopy. Upon review of the chest films, a tapering shadow, apparently venous, was seen extending into the right lung field obliquely upward and outward from the region of the inferior vena cava (fig. 11). The point of maximal cardiac impulse was in the right second intercostal space in the midclavicular line. The blood pressure in the arms was 80/50, and in the legs 100/55. A soft, systolic murmur was heard in the precordial region.

The patient was readmitted on June 27, 1955, for consideration of surgical therapy. At this time, the physical findings were the same as those of the previous admission. Bronchography was performed on July 8, 1955 (fig. 12). The left lung appeared normal. It was thought that the right main bronchus was rather long. Actually, it was shown subsequently by anatomic study to be symmetrical with the left. No upper lobe bronchus was identified, and it was thought that there were “no bronchi in the anterior half of the chest on the right.” The pulmonary artery was not discerned. It was thought at this time that there was partial hypoplasia of the right lung, with probable hypoplasia of the right pulmonary artery, and abnormal venous return from the right lung. Angiography was undertaken on July 13, 1955 (figs. 13–15). Moderate enlargement of the right atrium and right ventricle was noted. Branches of the left pulmonary artery in the left lung appeared to be normally distributed. The right pulmonary artery was visualized, but it appeared to be small. The left pulmonary veins emptied into the left atrium, but on the right side all pulmonary veins were collected into a large venous channel that was thought to empty into the vena cava or right atrium. At 1.2 sec., the smaller size of the right pulmonary artery was in sharp contrast with the large trunk and left branch. The pulmonary veins of both sides first became opacified at 2.8 sec. and were sharply outlined at 3.5 sec. The course of the anomalous right pulmonary vein was demonstrated with great

**Fig. 11.** Case 2. Plain film. Clearly shown against the background of the dextroposed enlarged heart is the tapering anomalous pulmonary vein (arrows). The right hemithorax is smaller than the left.

**Fig. 12.** Case 2. Bronchogram. The trachea branches symmetrically. Only 2 major bronchi are present in the right lung. Here there is a lingula, but no middle lobe. Extending laterally above the lingular bronchus (Li) can be seen a small outpouching of the upper lobe bronchus (compare with fig. 17).
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clarity (fig. 14). At 4.2 sec., the aortic arch first became radiopaque. A large artery was seen to ascend from the aorta to the base of the lung at 6.3 sec. (fig. 15). Even at 13.2 sec. there was still some opacification of the right cardiac chambers, evidence of the long persistence of recirculation. A right thoracotomy was performed on July 19, 1955, in the hope that a surgical anastomosis of the anomalous venous channel to the left atrium might be possible. This, however, proved not to be the case. Five large arteries were found to proceed from the aorta into the substance of the right lung at various levels. A bronchial artery also was found along the right main bronchus. The pleural cavity on the right side was obliterated by adhesions. A pneumonectomy was performed.

Anatomic Observations. The gross specimen was lightly covered with the remnants of adhesions. There was no external fissure. A bronchovascular cast was prepared. The anomalous vessels were well seen in a roentgenogram of the cast (fig. 16). The specimen, although derived from the right hemithorax, was seen in the cast (fig. 17) to possess essentially the structure of a left lung, but with venous drainage entirely into the inferior vena cava, and with 6 arteries derived from the aorta substituting for certain pulmonary arteries.

The right stem bronchus was hyparterial (figs. 17, 18). There were 2 main bronchi, an upper and a lower lobe bronchus. The superior division of the upper lobe bronchus divided into 3 branches of which the anterior was a blindly ending wedge-shaped rudiment. Others comprised the apical and posterior branches. Rami of the latter extended into anterior segmental territory. The inferior division had the structure of a lingula and extended almost to the base of the lobe on the medial aspect. It was here fused within the same pleural membrane as the lower lobe. The lingular bronchus had a small superior and a large inferior branch that sprang from the main lingular stem. The length of the latter was 1.3 cm.

The lower lobe bronchus, after a course of less than 1 cm., yielded a superior segmental bronchus and a common basal bronchus. The latter divided almost symmetrically into an anteromedial basal and common trunk for the lateral and posterior basal segments. These 4 segmental bronchi were of almost equal size.

Fig. 13. Case 2. Angiogram. 2.0 sec. The small size of the right pulmonary artery (RPA) and the enlarged pulmonary arterial trunk and left pulmonary artery are shown. The cavity of the right ventricle is also enlarged. Prominent interstices among the trabeculae carneae are visible as finger-shaped projections extending medially from the opacified right ventricular chamber.

Fig. 14. Case 2. Angiogram at 4.2 sec. The anomalous pulmonary vein of the right side (Va) and the pulmonary veins of the left (Vn) are simultaneously opacified.

Fig. 15. Case 2. Angiogram at 7.1 sec. Ascending from the aorta (Ao) is seen one of the accessory pulmonary arteries (XPA). The opacification of the vessels on the right side is maintained in part by recirculation through the pulmonary vessels and in part by the left to right shunt resulting in transfer of contrast medium from the aorta to the right lung through the accessory pulmonary arterial branches.
The arterial supply consisted of a main pulmonary arterial trunk and of 6 "accessory pulmonary arteries" derived from the aorta. The pulmonary artery looped over the left main bronchus and supplied all of the lung in a fashion typical for that of the left pulmonary artery, but in mirror image, except for those portions that were supplied by accessory pulmonary arteries. These comprised portions of the lingular and medial, anterior, and lateral basal segments, and all of the posterior basal. All of the accessory pulmonary arteries, except the lingular, swept laterally from the posteroinferior angle of the lower lobe. The largest of these vessels was one that supplied the lateral basal segment. The relations of these vessels to the bronchi were of interest; although the major arterial trunks approached the bronchi from the periphery of the lung, the actual bronchial branches became applied to the bronchi in a fashion characteristic of pulmonary arteries. None of these arteries had the spiral anastomosing characteristics of a bronchial artery, despite their aortic origin (fig. 19). There was no overlap in the territory supplied by the pulmonary and accessory pulmonary arteries.

The anomalous vein had 2 major tributaries, both of which ran obliquely upwards and forwards from a point near the posterior and inferior corner of the lobe. The smaller branch passed laterally into the fork of the left main bronchus where it drained the apical and a part of the posterior segments and the
Fig. 18. The relation of the main trunk of the pulmonary artery to the bronchi in a "right sided left lung" (as seen in cases 1 and 2 of present series) is shown in the lower half of the figure. The normal status is indicated for comparison in the upper half. In the former, the branching of the trachea is symmetrical, the stem bronchus is long, and the upper lobe bronchus is hyparterial. It is evident that the superior segmental bronchi of the lower lobe (B₆), in contrast with the upper lobe bronchus, is posterior to the trunk of the pulmonary artery in both the normal and anomalous lungs.

The larger branch swung upwards and laterally to the bifurcation of the left lower lobe bronchus and drained the remainder of the lung, i.e., the entire lower lobe and those parts of the upper lobe not drained by the other major branch (figs. 16, 17).

Case 3

Clinical History. This female Negro child was admitted to Babies Hospital on August 24, 1955, when 6 weeks old for wheezing since the age of 2 weeks. The patient was born without difficulty after an uneventful 9-month pregnancy. The neonatal period was normal until 2 weeks of age. At that time, the parents noted slight wheezing, particularly when the patient was lying on her abdomen. About 2 weeks later, a slight cough developed in the morning. She was taken to a pediatrician who, upon radiographic examination, noted "right upper lobe atelectasis."

There was no cyanosis or retraction and the cough and wheezing remained essentially unchanged. On admission, physical examination revealed an alert, active baby in no distress. The left anterior chest appeared to protrude, but both sides were resonant to percussion. Breath sounds were markedly decreased in the right upper lung field. No rales were heard. The heart sounds were strong and there were no murmurs. The blood count and urinalysis were normal. Fluoroscopic examination showed a mediastinal shift to the right, and the diagnosis of congenital hypoplasia of the right lung was made. Absence of the right pulmonary artery was queried. Upon reviewing the plain roentgenogram there was seen a separate right paracardiac shadow that represents the anomalous pulmonary vein (fig. 20). A tracheobronchogram showed a generally small caliber of the right bronchial tree with a "cavity" in the position usually occupied by the right middle lobe (fig. 21). The impression was that there was hypoplasia of the right middle lobe. Exploratory thoracotomy was considered not to be indicated at the time. The patient was discharged to have an angiocardiogram at a later date.

The second admission was on September 25, 1955, at 3 months of age for further study. In the interval, the child had had cough or stridor only on crying and there had been no cyanosis. An esophagram was normal. An angiocardiogram was interpreted as
showing hypovascularization of the right lung consequent to congenital hypoplasia of the arteries. The main pulmonary artery and right and left branches were seen, but it was thought that most of the blood was carried through the left lung. The sequence of hemodynamic events was traced in a review of the angiograms (figs. 22, 23). At 3.2 sec., the right ventricle and left pulmonary artery were opacified, but there was only initial filling of the right pulmonary artery. At 5.4 sec., not only the pulmonary arteries, but also the veins, were visible, including the distal end of an anomalous right pulmonary vein. The aorta became opacified at 6.5 sec., but there was still evidence of recirculation through the right side of the heart, which persisted to 8.7 sec., but faded thereafter.

The third admission was on November 30, 1955. Wheezing had persisted at home. Two weeks prior to admission, she had an upper respiratory infection for which she was treated with penicillin. Feeding and weight gain had continued to be good.

At physical examination the temperature was 98.6°F, pulse 94, and respirations between 22 and 40. The patient was well developed and well nourished, active, and in no distress. The head was normal. There was a moderate preponderance of the left
anterior chest. The heart sounds were of good quality and were heard on both sides of the sternum. There were no murmurs. A biphasic wheeze was heard throughout the chest and there were no rales. The breath sounds were heard equally well in both lung fields. There was a large reducible umbilical hernia which protruded 4 cm., and had a circumference of 6 cm. The liver and spleen were not palpable.

It was thought that the congenital anomaly of the lung would jeopardize the patient’s future. Since repeated upper respiratory infections were beginning to occur, it was thought that surgery was indicated. The patient underwent a right pneumonectomy on December 2, 1955. At operation the right pulmonary veins veins drained into the inferior vena cava. The right middle lobe and right lower lobe received arterial blood from the abdominal aorta, entirely or in part. A systemic artery to the upper portion of the lower lobe which arose from the thoracic aorta curved around and compressed the esophagus. The right pulmonary artery was seen to supply at least the small upper lobe and a portion of the middle lobe. There was herniation of a portion of the right lower lobe through the foramen of Bochdalek.

On the sixth postoperative day, roentgenographic examination revealed a normal left lung. There was the expected mediastinal shift to the right. The left lung field was still clear, and she was eating well.

On December 16, the fourteenth postoperative day, the patient was noted to be breathing with some degree of laryngeal obstruction. During examination, she became apneic and cyanotic. She responded to oxygen and artificial respiration and was placed in a croupette. In view of the possibility that this episode might be related to distortion of the left main bronchus secondary to a marked mediastinal shift, a right pneumothorax was performed by the injection of 50 ml. of air into the right pleural space over a period of 20 min. After this, aeration seemed better, but there remained a large laryngeal obstructive component.

Roentgenograms then revealed that the heart was shifted back to its preoperative position. There was a questionable infiltrate in the left upper lobe. A murmur was noted to the left of the sternum during the morning. During pharyngeal suction, she developed marked respiratory distress characterized by an epilephytic seizure with a tonic spasm of her chest and apnea. Artificial respiration was maintained by an endotracheal tube, but it was not possible to achieve good exchange. A tracheotomy was done, but the child’s heart beat had ceased.

At necropsy, the most significant lesion was massive mediastinal emphysema. There was also a small subdiaphragmatic abscess. No significant changes were noted in the left lung except for edema. The liver had a large caudate lobe. An adrenal rest was found in the right kidney. The left kidney was somewhat misshapen.

Anatomic Observations. The right lung measured 11 cm. from apex to base and 8 cm. from the rounded posterior border to the sharp anterior margin. On the lateral aspect, 2 fissures were seen to delimit the boundaries of 3 lobes. There was an oblique fissure that lay somewhat more posteriorly than usual and demarcated a rather small lower lobe from the remainder of the parenchyma. The second fissure was shorter than the first, and was not transverse, but rather slanted upwards as well as anteriorly from its origin in the major oblique fissure. As a consequence, the “upper lobe” was much larger than usual, comprising approximately one third of the total mass of pulmonary substance. This appearance suggested that parenchyma ordinarily aerated from the right upper lobe bronchus was supplied by branches of the middle lobe bronchus.

On the hilar aspect, the large size of the middle lobe was especially obvious. The bronchial stump was rather high in the hilum. An almost transverse keel could be seen within the bronchial trunk, which measured only approximately 3 mm. from the level of transection to the level of the first subdivision. This keel divided the trunk into an “upper lobe” branch and the common trunk of the “middle” and “lower” lobes.

The single trunk of the transected pulmonary artery lay anterior to the bronchus.

A single large vein emerged on the medial aspect at the angle of junction of the middle and lower lobes in a position low and rather anterior even in comparison with that of the lower lobe vein in the normal lung.

Five “accessory pulmonary arteries” were present; all were on the medial and posterior aspects of the lung and were said to have taken origin in the subdiaphragmatic portion of the aorta. For convenience in description, they were numbered in a clockwise manner, beginning with number 1, a small artery that entered just below the main bronchus. Number 2 was a large vessel entering along the posterior margin halfway between the bronchial stump and the diaphragmatic margin. Just beneath this vessel was number 3, a smaller artery pursuing almost the same direction. Artery number 4 entered the lung, bending horizontally and forward, approximately 1 cm. above the diaphragmatic border. Artery number 5 was a large vessel that made an entrance in close relationship to the draining venous trunk, but just anterior to it. The arteries that it was possible to inject with black plastic comprise only numbers 2, 4, and 5.

The lung was the seat of interstitial emphysema, most marked on the lateral aspect of the lower lobe anteriorly. Most of the dissecting air could be seen within the pulmonary septa and beneath the pleura. Some air was apparently within the lymphatics.

A bronchovascular cast was prepared (figs. 24, 25). Roentgenograms of the cast demonstrated the anomalous distribution of the bronchi and the
presence of an outpouching in the lateral ramus of the middle lobe bronchus that then yielded branches of normal size (compare figs. 21 and 24). These roentgenograms emphasized the bronchial distribution that was seen in relation to the vessels in the cast after digesting away the tissue (figs. 26, 27).

As suspected from the external appearance, the “upper lobe bronchus” had a distribution corresponding, somewhat irregularly, to the posterior and apical segments. The first branches of the upper lobe bronchus were not derived until 1 cm. from its origin. Here there was a division into B3a (superior ramus of posterior segment) and the common branch of B3b (inferior ramus of posterior segment) and B1 (apical segmental bronchus).

The common trunk of the “middle” and lower lobes had a length of 3 mm., before dividing into “middle” and lower lobar trunks.

The “middle lobe” bronchus, after a course of 5 mm., divided into medial and lateral branches. The medial was much larger. It yielded 2 successive branches that aerated the region of the anterior segment. Three millimeters beyond the origin of the lateral segmental bronchus there was a fusiform aneurysmal outpouching 1.2 cm. long and approximately 4 mm. in diameter.

The lower lobe bronchus was 8 mm. long. No superior segmental bronchus was derived from this trunk. The first branch was the medial segmental; below this was the common basal trunk which had a length of 1.4 cm. before dividing into B8 (anterior segmental) and the common trunk of B9 (lateral segmental) and B10 (posterior segmental). Accessory subsuperior bronchi were yielded by this common trunk, and also as posterior branches of all of the basal bronchi.

The trunk of the pulmonary artery divided into an upper lobe branch and a common trunk that supplied the remainder of the lung. The upper lobe branch divided after a course of 3 mm. into branches that corresponded exactly to those of the major bronchi as described for this lobe.

The common trunk within 2 mm. of its origin yielded a small artery that supplied most of B4 (lateral segment of middle lobe), then, after a course of 1.2 cm., it yielded a large branch to B5 (medial segment), which supplied also those branches that represent the B2 (anterior) segment of the normal lung. There was a third, minute artery derived from a short common trunk with a B8 branch that supplied the medial rami of B4. The common basal branches otherwise followed the bronchial pattern closely.

The anomalous arteries derived from aorta were given the same numerical designations used in the external description of the specimen. Artery number 1 (not injected) may, in fact, have been a bronchial artery, since there appeared to be no bronchial distribution devoid of true pulmonary arterial supply in the region of its entrance into the lung.

Fig. 24. Case 3. Roentgenogram of cast (medial aspect). Lead sulfate was incorporated in the plastic to bring the bronchial tree into contrast. There is a large fusiform aneurysmal outpouching (D) of the lateral segmental bronchus of the middle lobe. The branches continue from this laterally in their usual caliber. The 3 rami of the upper lobe bronchus can be distinguished (B1 = apical; B2 = superior ramus of posterior segmental; B3 = inferior ramus of posterior segmental). ML = middle lobe bronchus. LL = lower lobe bronchus.

Fig. 25. Case 3. Posteroanterior roentgenogram of cast. The 3 branches of the hyparterial right upper lobe bronchus are shown more clearly, and in relation to the upper lobe stem (UL). The medial (B4) and lateral (B5) branches of the middle lobe bronchus are also shown.
Artery number 2 supplied portions of B5, as did number 3. The distribution of the latter was presumptive, since number 3 was not injected. Artery number 4 was injected and supplied distal portions of B7 and B8. Artery number 5 supplied a part of the B5 segment. Arteries number 2 through 5, all derived from the subdiaphragmatic portion of the aorta, had the characteristic distribution of pulmonary arteries. The course of each was not plexiform, but instead followed the branchings of the bronchial ramus. They were end arteries, in the same sense as the pulmonary arteries, and their size in relation to the bronchus was similar. There was no evidence that they yielded any blood supply to the bronchial
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FIG. 27. In a lateral view of the cast, 2 branches of the upper lobe bronchus can be seen. The courses of the major venous trunks are displayed. At the termination of one of the accessory pulmonary arteries (indicated by an asterisk) the vessel assumes a pigtail tortuosity, characteristic of pulmonary arteries carrying blood at excessively high pressure. (Key as in fig. 26).

tree until the alveolated portions were reached. Their distal branches (fig. 27) were marked by a "pigtail" tortuosity characteristic of high pressure, as seen in the pulmonary arteries in most instances of transposition of the great vessels.

The single large vein was formed by the confluence of many branches resembling a candelabrum. There were 2 main trunks: an anterior trunk passed obliquely upwards and laterally to drain the upper and middle lobes; the other trunk drained the lower lobe and was formed by the union of a vessel, V7, that served chiefly the medial segment, with the common trunk draining the basal segments. Portions of adjacent segments were, of course, drained by these vessels, as is characteristic of pulmonary veins in general. Several minute branches also entered the various trunks close to their confluence.

Two blocks of tissue were taken from each of the 3 lobes for microscopic sections. In the alveoli, there were occasional large mononuclear cells. There was dissection of the interstitial connective tissue by gas. Focal hemorrhage was present within many of the alveoli, and some contained fibrin. In one margin there was apparent necrosis of the alveoli, interpreted to be the result of clamping during operative removal. This also was thought to account for the hemorrhage and fibrinous material within the alveoli. There were no changes suggestive of an organizing pneumonia in the sections studied. A few bronchioles that were present had well-preserved epithelium, despite the injection procedure.

SUMMARY

Observations upon 3 instances of anomalous pulmonary venous return into the inferior
vena cava are presented in comparison with previously published material. This lesion is often asymptomatic and compatible with long life, but is sometimes associated with shortness of breath and symptoms of chronic bronchopulmonary disease. In those patients subjected to the most detailed study, there were often found additional anomalies of bronchial distribution (such as the presence of a mirror image of the left lung on the right side, or cystic diverticula of the bronchial tree), and accessory pulmonary arteries from the descending thoracic aorta. Bronchographic and angiographic technics are useful in bringing such associated anomalies to light. These features should be considered with reference to the usefulness of the right lung and its freedom from chronic supplicative disease in planning corrective or extirpative surgery.

**Summario in Interlingua**

Es presentate—e comparate con previamente publicate materiales—observationes colligite in 3 casos de anomalie retourno pulmonovenose verso le vena cave inferior. Le lesion es frequentemente asymptomatic e non incompatible con longevitate, sed a vices illo es associate con dyspnea e symptomae de chronic morbo bronchopulmonar. In le patientes in qui le plus detaliate studios esseva execute, il esseva frequentemente possibile constatar anomalies additional de distribution bronchial (per exemplo le presentia al laterale dextere de un imagine specular del pulmone sinistre o cystic diverticulæ del arbore bronchial) e accessori arteriae pulmonar exiente ab le descendente aorta thoracique. Technicas bronchographie e angiographie es utile in le detection de tal associate anomalías. In elaborar planos pro chirurgia corrective e extirpative, le mentionate punctos debereaa esser considerate con referentia al utilitate del pulmone dextere e a su libertate ab chronic morbo suppurative.

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As an animal Body consists not only of a wonderful texture of solid Parts, but also of a large proportion of Fluids, which are continually circulating and flowing, thro' an inimitable Embroidery of Blood-Vessels, and other inconceivably minute Canals: And as the healthy State of an Animal principally consists, in the maintaining of a due Equilibrium between those Solids and Fluids; it has, ever since the important Discovery of the Circulation of the Blood, been looked upon as a Matter well worth the enquiring into, to find the Force and Velocity with which these Fluids are impelled; as a likely means to give a considerable Insight into the animal Economy.—Reverend Stephen Hales, B.D., F.R.S. From the Introduction of: Statical Essays. Vol. II, London, 1733.
Bronchial and Arterial Anomalies with Drainage of the Right Lung into the Inferior Vena Cava
NICHOLAS A. HALASZ, KATHERINE H. HALLORAN and AVERILL A. LIEBOW

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