PULMONARY CYSTS

Review of the Subject, with a Case Report*

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In 1687 there appeared in the *Opera Omnia* by Malpighius a report by Thomas Bartholinus of three cases of what were presumably lung cysts. The original description is in vulgar Latin, and is so quaintly and so naively written that it should be cited in full. The translation is from the Leyden edition and is quite literal.

A new case, and a rare one in mankind, of a boy without the ordinary lung—a case observed by N. Fontanus and favorably commented upon by ourselves—amply confirms the new observation of Malpighius. I shall relate the whole course of this strange case from Fontanus himself, because many learned men, when they have incorporated this phenomenon into their writings, have done so upon the basis of the belief they have placed in my word.

The four-year-old boy died, brought to his end by marasmus. Out of interest in the matter I dissected the body, observing that the liver was above the ordinary size. It had an abscess near the vena cava, which was covered with pus all around about the humped portion; a spleen of good color, and unaffected; no omentum or at least none was readily visible. And strange to relate, there were no lungs, but in their place was a membranous vesicle filled with air. This vesicle was provided with small veins and arose from the rough artery† itself; the vesicle seemed to carry the cooling effect to the heart. Fontanus wrote this to Cl. Plempius on December 20, 1638. He in turn, in his answer of December 29, judged the membranous vesicle to have occupied the place of a lung, since it could receive outside air for cooling the heart by virtue of the fact that it started from the rough artery. But because it did not furnish enough air to the heart, and because the heart could not easily enough be cleansed of its soot, the learned Plempius suspects that therefore the youth fell into this marasmus, the heat of the heart being too intense and almost burning up the body. The illustrious Boyle in his *Digressio* uses the same example drawn from our anatomy to illustrate what he has published, with great intelligence, concerning the lungs—that is, when the lungs lack the fibers with which to distend themselves they quite properly provide themselves with a vesica‡, and are distended by an ample supply of air. From this source I shall borrow a great aid to the observations of

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†trachea.

‡ membrane, bladder.

Malpighius. Nature formed this vesicle from its very origin, even as she forms the original substance of the lungs in all cases, and extended it from the rough artery so that the air, taken in by a continuous duct and path, might be transmitted to this point. According to the observations of Harvey, at the fourth month the membranous lung remains in the fetus without any effusion of blood. Thus nature made no mistake up to this point. But since, on account of the meagerness of the pulmonary vessels the course of the circulation was interrupted (and it has been elsewhere and by others shown that this circulation through the lungs is necessary for the preservation of the heart), cause was given for the marasmus; and to the misfortune of the marasmus the corrupted viscera of the lower belly conspired to add. Like this is the case communicated by L. Nonnus to Zacutus Lusitanus, although it is inadvisedly reported under the heading of those who are devoid of lungs. As a matter of fact, both in this case and in the other the lung was present but the lung vessels* by which a more ample nutriment should have gone to the empty vesicle were imperfect. The lungs of a three-year-old girl, suffering from a long quartan, seemed to Nonnus to have so far decayed that no trace of them appeared but only a membrane full of pussy matter. But on what authority could he know that the lungs had wasted away? It seems much more likely to me that save for the membranous vesicle no lungs ever existed in the child. My suspicion is increased by the fact that she was never overcome by a cough nor did she spit up pus. The pussy matter came from the blood and from the badly affected pulmonary veins, and was customarily disposed of through the heart. Then, when the heart ceased to function with the cessation of life, the matter was gathered in the pulmonary membrane. I have no doubt at all that the patient described by Fabr. Bartolettus and whom he believed to have no lungs, so that, rather, on either side was seen something similar to lung tissue, actually had lungs of this sort. This man undoubtedly had a membranous vesicle for a lung, and naturally so; for the matter stagnating in the thorax was not pus, as from an empyema, but a sort of milky water coming perhaps from the thoracic duct of Pecquet through the heart, or of the nature of our lymph, which, in this case Malpighius praises and approves, comes from the serous vessels which he has observed wandering about the surface of the lungs, and has described very admirably.

For about two centuries following this communication the literature was silent on the subject, and then records of isolated cases of cystic changes in one or both lungs began to appear sporadically. In 1925, Koontz reported the first case in this country. The 108 cases which he collected and reviewed at that time were derived

* Probably bronchioles.

chiefly from the German literature, with a few from the English, French, and Italian. These 108 cases were not all of intrapulmonary cysts, but included also diverticula of the trachea or main bronchi, solitary cysts of the tracheal or bronchial wall, and cysts of aberrant lung tissue and of accessory lobes.

Under the term "congenital cysts of the lung," which captions most of the recent communications, there have been included a wide variety of fluid- and air-containing structures. Many of these are related only in reference to certain gross characteristics and clinical manifestations, but present striking differences on histological examination and are probably entirely dissociated as far as etiology is concerned. Even their congenital nature is in dispute. Because of the lack of uniformity of opinion concerning the nature and origin of these cysts, it is preferable at present to include them all under the non-committal, generic term of "pulmonary cysts," and to delay further subdivision and subclassification until knowledge of their structure and of their relation to infections and to congenital malformations of the lung is more complete and better understood.

Pulmonary cysts have occurred in all ages from the six-month fetus in Meyer's case to the 84-year-old patient reported by Buchmann. They may be single or multiple, small or large, and may involve either one or both lungs. They may be scattered sparsely and irregularly through the pulmonary tissue, or may replace the usual parenchymatous structure of one lobe or of an entire lung.

Anatomical Structure

There is a great variation as to the anatomical structure of these cysts. They may be simple or multilocular. The walls may be everywhere smooth and regular or may be ribbed by small or large fibrous tissue septa. The cysts may contain only fluid, or only air, or a combination of both in varying proportions. The fluid may be thin, clear serous material, but is more usually turbid, mucopurulent, and sterile. Suspended in the fluid there are often minute, solid particles composed of cells which have desquamated from the cyst wall. The original nature of the fluid may be modified by hemorrhage into the cyst, or by infection. In the former case the fluid will be colored red or chocolate, depending upon the stage of decomposition of the blood. In the latter instance the cyst wall will contain thick purulent material which has a very offensive odor and

which strongly resembles pus from a lung abscess or an infected bronchiectasis.

In most of the reported cases a direct communication was found between the cyst cavity and a bronchiole. In those where no communication was found on gross inspection or even by injecting fluid into the bronchial tree at autopsy one cannot say definitely that such communication did not exist. The bronchioles leading into air cysts are often so narrow, distorted, and tortuous that they may not be observed even on careful examination and can be located only by studying serial histological sections of the cyst wall. It is also similarly possible that in some of the fluid-filled cysts where no apparent bronchiole was found such a communication may have actually been present but was so small or so distorted that it permitted only the slow escape of the renewable fluid contents, but never a sudden appreciable emptying of the cyst. Such a structure would fit in with the clinical observations where small amounts of mucoid sputum were expectorated at irregular intervals.

Microscopic examination reveals in most instances a striking resemblance between the structure of the cyst wall and that of the bronchial tree. The inner lining of the cyst is composed usually of a pseudostratified layer of columnar epithelium resting upon a fairly definite basement membrane. It is this layer which produces the smooth, soft, velvety structure so often observed and described in the gross. Outside of the basement membrane is a layer of areolar connective tissue within which are scattered large blood vessels, elastic tissue fibers, collections of smooth muscle, and islands of cartilage, all in varying amounts and proportions. In some instances even mucous glands have been found. There are, however, cases which vary markedly from this usual picture. The inner wall in these instances is made up of a single or of multiple layers of cuboidal cells, with or without cilia, or of flat cells which resemble closely the endothelial lining of blood and lymph vessels. Occasionally, no epithelial layer at all is present, and the inner cyst lining is composed of areolar tissue. It is probable that in these cases an epithelial or mesothelial layer had been originally present, and that it desquamated either in vivo or in vitro. It is obvious that the structure of the wall of the cyst may be modified by its contents and the manner in which it is formed. If, as in the case here reported, the cyst has undergone a rapid enlargement under tension it is unlikely

that its surface could be covered by epithelium even though the original small cyst may have been completely lined by these cells.

The pulmonary tissue adjacent to the cysts may present different stages of atelectasis, varying anywhere from alveoli of normal size and presumably normal function to completely collapsed functionless air sacs. The walls of such shrunken sacs are contiguous, and their alveolar identity is witnessed only by the characteristic presence and arrangement of elastic tissue fibers, and of scattered structures which resemble compressed bronchioles (Oudendal). Frequently the tissue neighboring the cysts bears no resemblance to normal or even much compressed pulmonary tissue, and is made up of connective tissue with no trace of alveolar structure and with a striking paucity of elastic tissue fibers. In all of the above instances the microscopic picture may be profoundly changed by superimposed infection which may completely alter the original histological structure of these cysts and thus mask or distort such anatomical landmarks as could perhaps have been of aid in tracing their mode of origin.

Theories of Formation of Cysts

The term "congenital cyst of the lung" implies that the etiological factor, if not the cyst itself, exists before birth, but this is by no means proved in all cases. Although most writers favor the congenital origin of these cysts, there are still some who feel that they may occur as a result of postnatal, mechanical, or infectious influences. The various theories are presented briefly.

Stoerk believes that there may be at some time in intrauterine life an inflammation of a bronchiole with resultant scar formation, constriction and subsequent occlusion of the lumen of the bronchiole. The accumulation of the bronchial secretions distal to this occlusion would thus result in cyst formation. Such an inflammation, however, may produce only a partial obstruction of the involved bronchiole. In such a case, with the onset of breathing at birth, it is conceivable that a condition may exist where on inspiration, air will be sucked through the narrowed bronchiole, but where on expiration a variable amount of this air will be trapped distal to the point of narrowing. Thus, on repeated respiratory movements, air will slowly accumulate behind the constriction and produce in that area a dilatation of the bronchioles and alveoli. If this progresses long

enough, it will result in the formation of a conspicuous bronchiectasis, or, to use another term, in air-containing cysts. The air may accumulate in this structure until the tension within becomes great enough to resist further introduction and confinement of large amounts of air. A state of equilibrium will thus be set up where the cyst will continue to contain air under tension, but will not further increase in size. The point at which such an equilibrium will be established and the size which the cyst will, therefore, attain are direct functions of the degree and nature of the bronchial constriction. If the obstruction is almost complete and is of a one-way character, it is possible that such an equilibrium may never result. The cyst will then continue to expand and increase in size, and will not only compress the adjacent normally functioning pulmonary tissue, but will also eventually displace and rotate the mediastinal structures to the opposite side. This will in time result in circulatory and respiratory embarrassment which may become fatal. The number of bronchiectatic cavities or air cysts that will be formed in this manner depends upon the number of bronchioles involved in the original prenatal inflammation.

The English authors (Tooth, Bernstein, Box) agree that such cysts are the result of severe inflammation of the bronchioles with subsequent bronchiolar constriction. They feel, however, that this inflammation is not a prenatal one, but is a postnatal infection, a diffuse bronchitis and bronchiolitis such as are found in pneumonia following measles, whooping-cough, influenza, and in debilitated rachitic children. Others (Pollack and Marvin) believe that the constriction is due to neither a prenatal nor postnatal inflammation, but rather to a congenital malformation and distortion of the bronchiole. Sauerbruch maintains that a bronchus, especially in the region of the left lower lobe, may be constricted prenatally as a result of pressure of the adjacent duct of Cuvier. He feels that such infection as has been reported in the English literature may be a secondary and not a primary manifestation.

To explain the already vague and obscure origin of pulmonary cysts, many have introduced another equally vague and little understood concept, atelectatic bronchiectasis. The association between bronchiectasis and atelectasis was made by Reynaud as early as 1835. Fifty years later the theory of atelectatic bronchiectasis was revived and was given a new and lasting impetus by Heller. This condition presupposes that at birth there exists an atelectasis which may be

due to a complete occlusion of a bronchiole by a mucous plug, by aspirated meconium, or may result from a congenital malformation or inflammation of the bronchial wall. After birth, as the thorax rapidly increases in size, there follows in the normal lung a concomitant increase in size and in capacity of the pulmonary alveoli which take up the excess intrathoracic space thus provided. When a large portion of the lung is atelectatic, however, it is argued that the bronchiolar walls expand and dilate to take up this excess space which ordinarily would have been occupied by the now collapsed alveoli. In this way there results the so-called atelectatic bronchiectasis which has been described as giving rise at an early period in life to the usual symptoms of infected bronchiectasis. When such bronchiectases are large, saccular, and uninfected they may form structures resembling lung cysts. Cases have been reported where the mechanism of the formation of the cysts was in general agreement with the above outline, but where the bronchiectasis was believed to have resulted not from the presence of shrunken neighboring alvoli but rather from the congenital malformation or complete agenesis of these alveoli. The congenital nature of the disturbance is identified by the striking absence in the tissue adjoining the cysts of the usual and characteristic elastic tissue scaffolding, which one always finds in atelectatic alveoli, and which is frequently present even after severe infections of the lung.

The concept that cysts are not the result of simple developmental mechanical or infectious influences but are in reality neoplasms was advanced by Stoerk in 1897. He believed they were produced by the neoplastic proliferation of the embryonic endodermal bronchial tissue and called them cystic fetal bronchial adenomata. Löhlein was struck by the similarity of these cysts to the papillary cystic ovarian tumor, and assumed therefore a similar neoplastic basis. Hueter called them congenital alveolar pulmonary adenomata.

Virchow thought these structures were due to a disturbance of the lymphatic circulation of the lung with the resulting formation of a lymphangiectatic cyst. Such cysts would be analogous with the cystic lymphatic hygromata of the neck which are present in early life and which are presumably congenital in origin. Klebs agreed with this explanation, and applied the term lymphangiectasia congenita pulmonum. More recently Anspach and Wollman have revived this theory.

As in every disease of disputed etiology, syphilis here also has

been blamed as the causa morbi. Sandoz thought the cysts resulted from the improper development of the terminal portion of the bronchial tree. This maldevelopment was in turn believed to be due to the presence of syphilitic infection within the lung. He supported his view by calling attention to the fact that pulmonary cysts had been described in cases where known systemic syphilitic infection was present. Oudendal thought that prenatal pleural adhesions might be responsible for the formation of cysts. He suggested that syphilis was the cause of these adhesions.

A critical analysis of these theories is difficult because relatively little detailed information is available. There are, however, a few incongruities which are at once apparent even on casual perusal of these explanations. The evidence in favor of syphilis as the sole offender is entirely inconclusive, and barely suggestive. The fact that such cysts have been found in known syphilitics can certainly not be interpreted, as it has been by Sandoz, as meaning that the cysts represent one of the stigmata of the infection.

It is rather difficult, also, to conceive of the formation of these cysts as a result of either atelectatic bronchiectasis or of bronchiectasis due to alveolar agenesis. Certainly in the cases of acquired atelectasis due to the presence of a mucous plug, a foreign body, or a neoplasm such as is seen in later life, there is no tendency to bronchiectatic or cyst formation. The bronchi and bronchioles do not dilate appreciably to take up the excess space in the thorax. Instead, there is a shift of the heart and mediastinum toward the involved side, and an elevation of the diaphragm on that side. It is difficult to attribute a different reaction to atelectasis in a newborn or a young child where the same mechanical principles obtain within the thorax. It is extremely unlikely that in a newborn child the mediastinum should be so rigid and so fixed as to prevent the shift of its contents to the side where perhaps an entire lobe is collapsed. However, even if it were so rigid, it is improbable that the remaining, normal, elastic alveoli should not participate in taking up the slack produced by the atelectatic lobe, and that the more rigid bronchial tubes should alone dilate to the extent of the formation of large air cysts. Heuer and his collaborators have demonstrated in dogs that when all but one lobe of both lungs is removed, the remaining lobe expands until it fills the space originally occupied by all seven lobes. That the normal pulmonary alveolar structure can take care of even large "dead spaces" in the thoracic cavity has been

shown again by Rienhoff and others in cases of pulmonectomy as well as in the case here reported. These facts throw doubt not only upon the theory of the formation of lung cysts from atelectatic bronchiectasis, but also upon the entire subject of atelectatic bronchiectasis itself. The conception that bronchiectasis may occur early in life as a result of neighboring atelectasis has taken a strong foothold in the literature, and, whether justly or not, is accepted more or less as a truth. In those cases where autopsy revealed the presence of bronchiectasis and of adjacent diffuse atelectasis, it has been assumed that the former was a result of the latter. Is it not just as likely, or perhaps even more likely, that the bronchiectasis was the primary factor, and that the atelectasis resulted from the pressure of the dilating bronchi? The cause of such a primary bronchiectasis is, indeed, not known at present, and it may be the same as that which, when acting in greater degree, results finally in the formation of lung cysts. It seems possible that those cases of unilateral bronchiectasis which produce symptoms in early life and which have been included under the classification of atelectatic bronchiectasis are really cases of primary unilateral pulmonary air cysts which have become secondarily infected.

The theory proposed by Virchow that these cysts are interstitial and represent cystic lymphatic hygromata is a very plausible one, but can be applied to only a very small number of the cysts described. The lining of such a hygroma must be made up of a single layer of flat endothelial cells. It is true that occasional cysts present this structure, but certainly the majority of them are lined by cuboidal or columnar epithelium which may or may not have cilia. This latter and more frequent structure favors a bronchial rather than a lymphatic origin.

That all pulmonary cysts are neoplastic is possible, but very unlikely. Any malignant nature is fairly definitely ruled out by their histologic structure and by the fact that they have apparently been present for many years without evidence of local invasion or of distant metastases.

That some pulmonary cysts are certainly congenital in origin is borne out by the discovery of these structures in fetuses and in premature still-births. The relatively minute amount or the complete absence of anthracotic pigment in the walls of some cysts has been interpreted as proof of their congenital nature. This criterion, although advocated repeatedly with great conviction, is not entirely

reliable. The amount of pigment deposited in the walls of the cysts is a function of the amount of air circulating through the cyst and the power of absorption of its wall. Thus a congenital cyst having a communication with a bronchus that permits the entrance and circulation of air may after many years store up a large amount of pigment in its wall. Conversely, a cyst acquired early in life when there is very little pigment anywhere in the lung may communicate with a bronchus through a narrow opening which allows only very slight circulation of the dust-laden air. In this instance the cyst, even though an acquired one, may remain relatively free of pigment. In both congenital and acquired cysts the anthracotic pigment deposit must be less than in the adjacent lung by virtue of the tremendous difference in the absorptive surface of the cyst wall from that of normal pulmonary tissue occupying the same volume.

The many theories above presented concerning the etiology of these cysts evidence our lack of knowledge of their true nature. Many of the proposed mechanisms are only theoretical possibilities which have as yet not been satisfactorily observed or demonstrated to take place. Although it is conceivable that they may produce cystic changes, still one cannot attribute with definiteness and finality the mode of formation of all of these different types of cyst, or of even one small isolated group to an unobserved and entirely theoretical set of circumstances. It is indeed possible that pulmonary cysts all arise as a result of a single common process, whatever its nature may be, and that the numerous variations in location, size, structure, and contents are only the result of subsequent mechanical, chemical, or infectious influences. It is more reasonable, however, to suppose that pulmonary cysts are not essentially different from cysts of any of the other parenchymatous organs of the body, and that like these cysts they may be congenital, infectious, mechanical or neoplastic in origin. The scant knowledge of the finer structure of these cysts and of their evolution even after they are recognized does not permit at present a definite, final opinion or classification.

Symptoms

The clinical manifestations of these cysts are extremely variable and depend upon the size, location, and contents of the cysts, the nature of the communication with the bronchial tree, the presence or absence of infection, and the possible rupture of the cyst wall into

the pleural cavity. Many of the uninfected cysts have been asymptomatic and were discovered incidentally at autopsy. Others have produced symptoms simulating tuberculosis, including even hemoptyses. The most typical and perhaps pathognomonic symptom-complex of lung cysts is that observed in infants and young children. This is manifested by repeated sudden attacks of dyspnea and cyanosis associated with the signs of tension pneumothorax and mediastinal shift. The cause of this abrupt attack is attributed to the rupture of an air sac into the pleural cavity. It is probable that in some cases, as in the one here reported, there is not a rupture of a cyst and tension within the pleural cavity, but rather extreme dilatation of, and marked tension within the intact cyst itself. Such a sudden increase in size of the cyst can be produced by the further constriction, as a result of a bronchitis, of the already narrow tortuous bronchial orifice. The child may die in the first of these attacks, or may survive it only to have recurrences at irregular but fairly frequent intervals. Unless relieved by thoracentesis, one of these attacks will sooner or later result in a fatal issue. The clinical manifestations of infected cysts are those of lung abscess or infected bronchiectasis. Large infected cysts at the periphery of the lung have been mistaken for and treated as pleural empyemata. Like the benign mediastinal tumors, the fluid-filled cysts may produce no symptoms at all or may cause pain in the chest, cough, dyspnea, cyanosis, and perhaps circulatory disturbances as a result of pressure upon the mediastinal structures.

Diagnosis

An accurate diagnosis of lung cysts is difficult. As stated above, the only typical history is that found in young children who have sudden periods of respiratory and circulatory embarrassment. The other symptoms that have been observed are not at all characteristic of lung cysts, and are found in a variety of intrathoracic lesions.

The findings on physical examination are extremely variable because of the great diversity in the attributes of the cysts. Positive findings, which may be completely absent in the case of small cysts, may serve to reveal only the presence of an air- or fluid-containing structure within the thorax. Such findings, however, in no way identify the nature of this structure. On the other hand, X-ray examination of the chest often reveals a typical picture. There are

in the lung fields small or large annular shadows whose density is less than that of the adjacent pulmonary tissue. These annular shadows may be situated deeply within the lung parenchyma or may be in the periphery. Stereoscopic examination reveals an absence of lung markings within these structures. A mediastinal displacement may or may not be present. Cysts filled with fluid present similar annular shadows but of increased rather than decreased density. Cysts which contain both fluid and air present the characteristic fluid levels.

Bronchoscopic examinations may demonstrate a definite constriction of the bronchus leading to these cysts. In some instances the corresponding bronchus could not be located at all. Attempts to inject lipiodol into these structures have in some cases been unsuccessful because of the small tortuous communications with the bronchioles, or because, as some maintain, of the absence of such a communication. In Eloesser's and Wood's cases there was observed a puddling of the lipiodol in the cysts. A similar picture may be obtained when the lipiodol is injected directly into the cyst through the chest wall (Ballon, Singer, Graham). The shape and confines of these cysts can then be obtained by taking roentgenograms in different positions.

Lung abscess, infected bronchiectasis, pleural empyema, and pulmonary tuberculosis should be considered in the differential diagnosis of infected lung cysts; large, peripheral air-containing cysts should be differentiated from pneumothorax; cysts filled with fluid may be mistaken for intrathoracic neoplasms or parasitic cysts. There may be difficulty in distinguishing infected pulmonary cysts from lung abscess or bronchiectasis not only clinically but even at autopsy, especially since these cysts are believed by some to be secondary pulmonary infections and suppurations. Infected peripheral lung cysts may reveal a history, physical findings, and roentgenograms which may be indistinguishable from those of pleural empyemata. Eloesser reported a case in a boy 20 years of age who for ten years was believed to have chronic empyema, and in whom the diagnosis of lung cyst was made only at operation after the lesion was seen. Neuhoff reports a similar instance where a sterile, fluid cyst was mistaken for an empyema and where the correct diagnosis was made only at operation. Eloesser states that in empyema the shadow is usually wedge-shaped with the base toward the chest wall. The shadow produced by an infected lung cyst, on the other

hand, is circular or rectangular. It is apparent that a differentiation based only upon the shape of the shadow is not reliable.

Large air-containing cysts may be mistaken for pneumothorax cavities. If the cysts are multilocular and have fibrous septa between them, the roentgenogram resembles a pneumothorax with scattered pleural adhesions. The walls of the cyst and the fibrous septa, however, present curved sinuous shadows which connect not only the visceral with the parietal pleurae, but which also fuse with each other, and which may run from one point on a pleural surface to another point on the same pleural layer. The pleural adhesions seen in a pneumothorax cavity on the other hand, are straight, taut, and so-called "fiddle-string" in type. Moreover, they do not bridge across different parts of the same pleural layer but extend from the visceral to the parietal pleura. In tension pneumothorax the pleural adhesions become straighter and more tense. When the tension is increased within the cysts, on the other hand, the shadows cast by the cyst walls remain curved and annular. Wilson, Pollack and Marvin, and Wood were able to identify a large lung cyst by inducing an artificial pneumothorax. In this way it was possible accurately to outline the cyst wall and to demonstrate its independence of the pleural cavity. This is a valuable diagnostic procedure, but is applicable only when the pleural surfaces are not fused. It is possible that some of the cases of spontaneous pneumothorax which have been attributed to tuberculosis are cases of lung cysts which have either ruptured into the pleural cavity or have themselves suddenly become so large as to simulate a pneumothorax cavity.

Treatment

The statement made by Miller in 1926 that "radical cure is manifestly out of the question" is probably not true today. The better understanding of the principles involved in the dynamics of respiration and the recent development of more adequate operative technic have permitted a successful attack upon intrathoracic lesions which was not possible only a decade ago. Nevertheless, the number of cases that have received treatment of any kind is relatively small.

Simple aspiration of air to relieve intrathoracic tension and a mediastinal shift has been reported in this country by Anspach and Wollman, Croswell and King, and Wood. In the nine-week-old

child described by Anspach and Wollman a single aspiration of 150 cc. of air from the chest temporarily relieved the symptoms, but resulted in no change of the mediastinal displacement. Respiratory difficulty again set in, and the child died. Croswell and King performed repeated aspirations of air from the chest of a three-year-old child with a large air cyst in the left lung. The child recovered from his respiratory difficulty and, at the time of their report, had been symptom-free for two years. Wood was able to relieve the respiratory embarrassment by a single removal of air from the chest and by limiting the activities of the patient.

Miller and Lillienthal combined aspiration of the chest with the subsequent insertion through the chest wall of a one-way "flapper valve" tube. The former author reported a case of a female child five weeks of age who had had attacks of cyanosis and dyspnea since the age of two weeks. She was found to have a right-sided pneumothorax and a displacement of the mediastinum to the left. Aspiration of air released the pressure, but it soon re-accumulated. A one-way valve tube was, therefore, inserted into the pleural cavity, and was removed after one week. At the time of discharge from the hospital the mediastinal shift was improved, but the pneumothorax was still present. The attacks of cyanosis and dyspnea recurred, however, soon after she had returned to her home. She died after five months during one of the attacks. Lillienthal reported the case of a woman of 54 years with a large fluid-containing cyst in the left upper lobe. Twenty-four hours after the aspiration of some of the fluid she became dyspneic and presented a marked displacement of the heart and mediastinum. A "flapper valve" tube, inserted into the cyst, served to relieve the symptoms completely. Removal of the tube led to almost immediate respiratory embarrassment. At the time of his report in 1929, the patient was quite well, but was still carrying the one-way tube in her chest.

In one of Wood's cases, a large cyst in the right lung was opened through a thoracotomy wound and the bronchial orifices leading into these cysts were cauterized. The patient was well for ten years and then died of peritonitis. Roentgenograms of the chest at that time showed no change in the appearance of the solitary cyst on the right side. A smaller cyst was noted in the left upper lobe. The mediastinum was still displaced to the opposite side. At autopsy water was injected into the trachea but did not enter the cyst on the right side whose bronchial openings had been previously cauterized.

In discussing Wood's paper, Singer stated that he had attempted cauterization of the bronchial openings in one case, but he did not indicate the route or the result. Adams suggested cauterization of the bronchial orifice with 35 per cent silver nitrate, but no mention was made of any case in which it had been tried.

Harrington completely excised in one stage a large fluid-filled cyst which occupied an azygos lobe. The patient, a woman of 37 years, made an uneventful recovery and has been apparently entirely well since. Sauerbruch was able to cure four cases of lung cysts by resection of the cyst or of the involved lobe. Melchior successfully resected a fluid-filled tracheo-bronchial cyst in an eight-year-old girl.

Neuhoff operated upon a woman of 30 years of age with a diagnosis of chronic empyema. At the first stage the lower lobe was packed off in order to permit the formation of pleural adhesions. The empyema cavity, which turned out to be a large lung cyst with sterile mucopurulent contents, was marsupialized at the second operation. The cyst gradually diminished in size, and at the time of his report in 1928 had a capacity of only 10 to 15 cc.

Eloesser reported a case in a boy 20 years of age with a history suggesting chronic empyema of ten years' duration. At operation he uncapped and marsupialized what were apparently multiple, infected cysts of the left lower lobe. After the infection had subsided, he dissected the cysts from the adherent chest wall, diaphragm, and pericardium, and completely removed them. This was essentially a left lower lobectomy. He closed the bronchial stump, but it soon reopened. At the time of discharge from the hospital the bronchial fistula was small, but still draining. The general condition of the patient was very much improved. A large, solitary air cyst was present in the left upper lobe, but it was asymptomatic and was not treated.

In general, the nature of the treatment depends upon the symptoms produced by the cysts. Those cysts which are silent and which are discovered on routine examination require no treatment. Although it is true that they may not always remain quiescent, but may become infected or may lead to intrathoracic pressure disturbances, still the risk of their exploration and resection seems at present to be greater than is the possibility of the occurrence of subsquent untoward symptoms.

The cysts which are encountered in children with attacks of

dyspnea and cyanosis present definite indications for treatment. The tension pneumothorax and the mediastinal displacement can be relieved easily by the simple introduction of a needle and the aspiration of air from the tense pleural cavity or cyst. It must be emphasized, however, that this is only a temporary treatment. Such a patient, if permitted to go home, is likely to have further similar attacks, which may end fatally. In those instances where the cysts involve an entire lung, an exploratory thoracotomy should be performed with the purpose of either cauterizing the bronchial communications or of completely removing that lung. The latter, if technically feasible, is preferable. Cauterization through a bronchoscope or through a thoracotomy wound appears to be of doubtful value as a therapeutic procedure even in uncomplicated and uninfected air cysts. The narrow and tortuous bronchial communications frequently cannot be located. Moreover, even if they are found, there is no assurance that the cauterization will result in the complete occlusion of such communications. It is possible that this procedure may serve only to narrow these orifices even more, and thus make it possible for air to be more easily trapped and retained within the cysts.

If the cysts do not involve an entire lung it is advisable to introduce a catheter within the cyst and to apply negative pressure. This will relieve the intrathoracic tension, and will in addition tend to re-expand and to over-expand the collapsed normal lung so that it again fills the major portion of the hemithorax. If no pleural adhesions form between this re-expanded lung and the chest wall, subsequent removal of the catheter may again lead to a re-accumulation of air within the cyst. The tension of this air will continue to increase, the mediastinum will be displaced and there will be a recurrence of symptoms. In such a case an open thoracotomy should be performed and the pleural surface roughened by physical or chemical means. The lung, again expanded by negative pressure applied to an indwelling intrathoracic catheter, will become adherent and fixed to the chest wall. The negative pressure should not be discontinued until it is felt that such adhesions are firm enough to withstand any possible pressure from the cysts. As a result of such treatment the lung may expand to fill the hemithorax, and may compress and obliterate the already narrow and tortuous bronchial communications. If the expanded lung does not fill the hemithorax, the lobe containing the cysts should be resected. The dead space

left in the chest may be filled in by further expansion of the remaining normal lung, by artificially paralyzing the phrenic nerve, or by local thoracoplasty.

Cysts which are filled with fluid, and which are indistinguishable from intrathoracic neoplasms should be explored, and if possible, removed. Infected cysts which present the same symptoms as those of chronic lung abscess or infected bronchiectasis are subject to the same principles of treatment as those of so-called chronic nonspecific pulmonary suppuration. The treatment of large infected peripheral cysts is similar to that of chronic encapsulated empyemata with communicating bronchopleural fistulae. Adequate drainage must first be instituted. After the infection subsides, cauterization of the bronchial orifices with closure of the wound by muscle pedicle grafts may be carried out. If the cysts are too large to be obliterated in this way the lobe containing them may be resected.

Case Report

T. P. No. A41109. A white schoolboy eight years of age was referred to the New Haven Hospital on August 8, 1934, by Dr. A. S. Brown of Waterbury, Conn. The past history was negative except for measles at the age of six. There was no history of any previous respiratory difficulties.

On April 5, 1934, the patient developed a pneumonia of the right lower lobe, followed shortly by a right pleural empyema. Pneumococcus type I was grown from both the sputum and the pleural fluid. After two preliminary thoracenteses at the Waterbury Hospital the empyema cavity was drained by intercostal thoracotomy and insertion of Carrel-Dakin tubes. He made an uneventful convalescence. The wound drained for three months and finally closed. For four weeks prior to admission he had increasing dyspnea associated with cyanosis after only moderate exertion. Otherwise, his general condition appeared to be good.

Physical examination at the time of his hospital admission revealed a well-developed and well-nourished white boy of eight years, who did not appear to be acutely ill. The temperature was within normal limits. The pulse rate varied between 110 and 140 per minute. The respirations were somewhat labored and varied from 20 to 40 per minute. There was no orthopnea. There was slight but definite cyanosis of the lips and nail-beds. There was no incurvation of the nails of the fingers or toes. The trachea was deviated markedly to the left side. The chest was symmetrical, but full and rounded. The respiratory excursions on the right side were diminished. A wide livid scar was present just below the angle of the right scapula, indicating the site of the recent thoractomy wound. The findings

in the chest were those of marked tension pneumothorax on the right with a corresponding extreme shift of the trachea, heart, and mediastinum to the left. The percussion note on the right side was everywhere tympanitic. This tympany extended well over to the left border of the sternum. The breath sounds were absent, and there was no transmission to the chest wall of the spoken or whispered voice. No succussion splash was present. On the left side the note on percussion was resonant, the breath sounds broncho-vesicular in character, and the transmission of the spoken voice normal. There were no râles. The heart lay apparently entirely within the left hemithorax. The apex beat was in the seventh interspace in the posterior axillary line. The rate was rapid but regular. A soft systolic murmur was heard at the apex. The remainder of the examination was negative. The blood count and urinalysis were not unusual. The blood Kahn was negative.

Roentgenograms:*

(1) April 11, 1934; sixth day of illness. The entire right hemithorax was obscured by a dense homogeneous shadow of ground-glass appearance. The lung markings on this side were barely visible. The left lung shadow was not unusual. There was a moderate displacement of the heart and trachea to the left side. A definite bulge in the mitral area of the cardiac shadow was present. A diagnosis of massive pleural effusion, right, and mitral configuration of the heart was made.

(2) April 12, 1934; seventh day of illness. An X-ray film of the chest taken after the removal by thoracentesis of 300 cc. of purulent fluid from the right pleural cavity demonstrated a uniform increase in density in the lower half of the right hemithorax. A pneumothorax was present along the axillary margin of the right apex. Here the lung border was smooth and was retracted from the chest wall for a distance of about 1.5 cm. No definite fluid level was apparent in this film. There was a slight improvement in the position of the heart and the mediastinum.

(3) April 20, 1934; fifteenth day of illness. The heart and trachea were in approximately their normal positions. The left lung shadow presented no unusual features. A marked pneumothorax was present on the right side, and the right lung was collapsed to at least one-half of its usual size. The lower portion of the right hemithorax was obscured by a dense shadow which probably represented fluid. Scattered through the parenchyma of the collapsed right lung there were a few oval-shaped areas of decreased density.

(4) May 12, 1934; five weeks after onset of illness. The pneumothorax cavity on the right side was increased in size. The pleura at the

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* Those up to August 28, 1934, were obtained through the courtesy of the Waterbury Hospital.

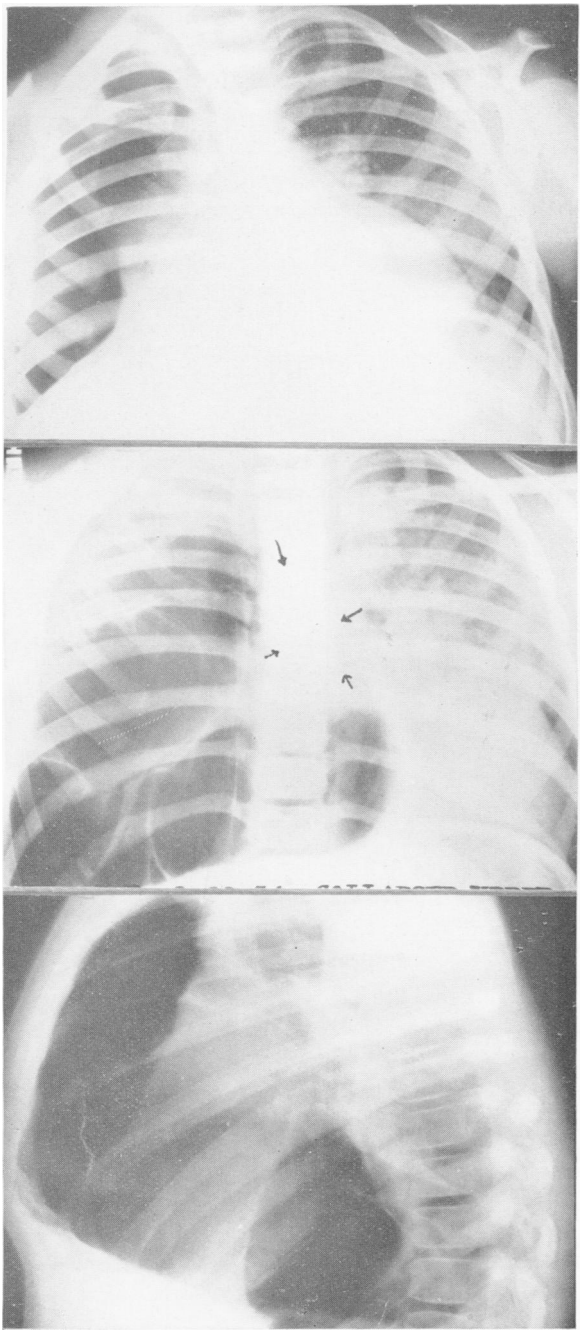

FIG. 1. 5/12/34. Pneumothorax, cyst formation, and collapsed lung.

FIG. 2. 8/28/34. Collapsed upper lobe (arrows) and multiple distended cysts.

FIG. 3. 8/28/34. Lateral view showing numerous cysts.

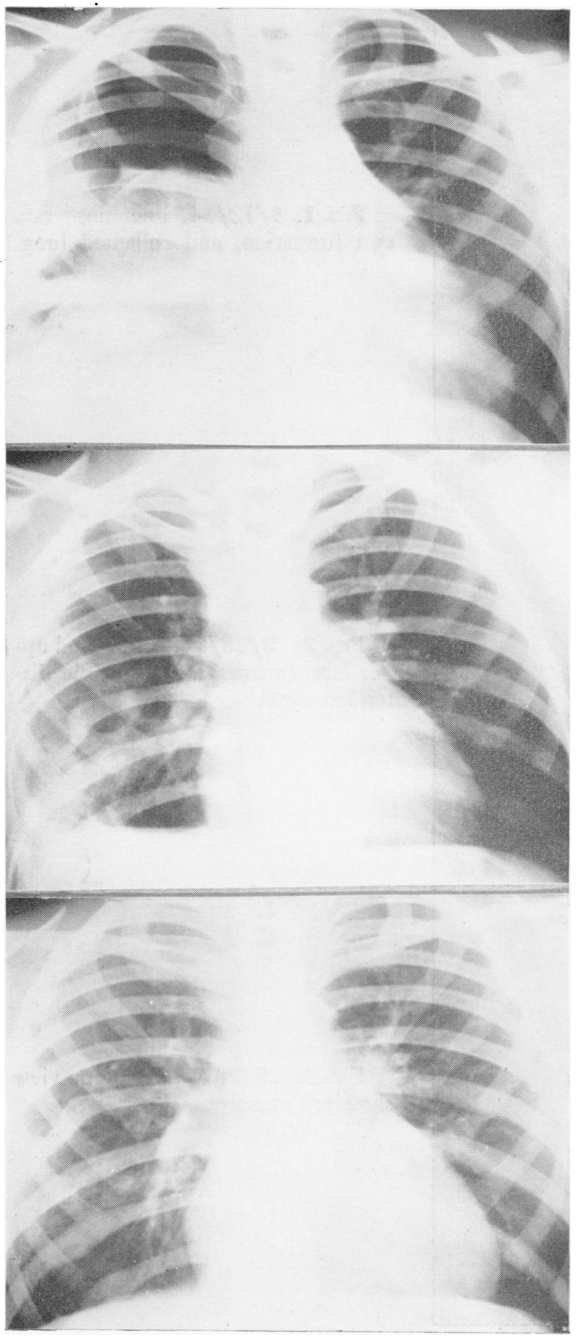

FIG. 4. 9/13/34. One week after operation, showing expanding upper lobe.

FIG. 5. 10/8/34. Four weeks after operation, showing lung fully expanded.

FIG. 6. One year after operation.

right costophrenic sinus was thickened but there was no evidence of any effusion within this pleural cavity. The right lung was collapsed to at least one-fifth of the normal size. Extending into the pneumothorax cavity from approximately the region of the right hilus there was an oval-shaped, air-containing, cystic area which measured 7 x 4 cm. The borders of this area were smooth and delicate but very distinct. There were definite, radially arranged bronchial markings projecting into this area and extending to within a short distance of its lateral border. The shadow of the lateral limits of the collapsed right lung presented a break in its continuity over a distance of 4 cm. This defect was occupied by the oval-shaped air cyst just described. The left lung presented no abnormalities. There was no shift of the mediastinum (Fig. 1).

(5) August 28, 1934. Stereo and lateral roentgenograms of the chest (Figs. 2 and 3) taken at the New Haven Hospital on the day of admission revealed a marked deviation of the trachea to the left. The cardia was approximately 1 cm. to the left of the left lateral border of the sixth dorsal spine. There was a pronounced herniation of the mediastinum toward the left side. The heart lay entirely within the left hemithorax with its apex just within the left lateral costophrenic sinus. There was in addition some rotation of the heart. The left pulmonary shadow was without note. On the right side, and projecting over into the left hemithorax, there were several very large, cystic, air-containing cavities. These were arranged in a rosette fashion around what was presumably the hilus. The walls separating these cysts from each other were represented by thin oval and circular shadows of increased density. At the level of the seventh rib posteriorly these septa were somewhat increased in number suggesting an interlobar division. There were no fluid levels. Obliquely overlying the bodies of the sixth and seventh dorsal vertebrae there was a shadow of uniformly increased density measuring about 5 x 2 cm. This shadow was roughly rectangular in outline and was thought to represent a completely collapsed lobe of the right lung. The apical and mediastinal pleura was thickened. Similar but slightly less pronounced findings were noted in the X-ray films taken at the Waterbury Hospital on August 24, 1934.

Shortly after his admission to the hospital a needle was inserted into the right hemithorax through the fourth interspace in the midaxillary line. The initial pressure was at a mean level of + 12 cm. of water. The oscillations with inspiration and expiration were shallow. The removal of one liter of air served to reduce the pressure to a mean of + 4. This pressure was quickly re-accumulated, however, and reached again a mean of + 12, where it remained stable. Another liter of air was removed with the same result. The needle was then introduced through two other areas, one anteriorly and one posteriorly. In each instance the initial mean pressure was + 12, and in each instance the withdrawal of relatively large amounts of air resulted in only a very temporary reduction of the intrathoracic tension.

Operation: September 6, 1934; rectal avertin and local novocaine anesthesia; right exploratory thoracotomy. A segment of the right eighth rib was removed subperiosteally in the region of the posterior axilla. When the inner periosteal layer was incised two distinct membranes were encountered which were adherent to each other but which could be separated without difficulty. These membranes, which were thought to be the parietal and visceral pleurae, were incised along the length of the wound and the hemithorax thus opened. A remarkable picture was encountered. The hemithorax was occupied by five large, air-containing cystic structures, which were partially divided from each other by thin septa. The largest of these cysts measured about 15 cm. in diameter. The cyst walls were delicate, velvety, and velamentous in character, and easily transmitted light. There had apparently never been any appreciable inflammatory process within these cavities. Coursing in the cyst walls were many large blood vessels which were usually grouped in threes, viz., a relatively large artery in the center and two adjacent concomitant veins. On the medial surfaces of each of the cyst walls there was at least one oval-shaped orifice through which air hissed into the cyst cavity with each inspiration. These ostia, which were undoubtedly bronchiolar openings, seemed to function like "flapper valves" which opened during inspiration but collapsed on expiration. The blood vessels lying in the septa between the adjacent cystic compartments were ligated, and these septa then divided. Thus the five independent cysts were converted into a single large cavity which filled the entire right hemithorax and which extended for a short distance beyond the midline. At this time the upper lobe, which was completely collapsed well down to the hilus, was seen. A number 20 F catheter was placed into the dependent portion of the large cyst and the wound carefully closed in layers around it.

The immedate postoperative course was stormy. The pulse remained high. The respirations were very labored and rose to a level of 50-60 per minute. The temperature reached a maximum of 104.2° F. Negative pressure was immediately applied to the catheter and this pressure gradually increased. It was only when a level of — 18 to — 20 cm. of water was reached that the respiratory embarrassment ceased. It was felt that with this amount of negative pressure there could be removed from the hemithorax at least as much air as he was able to suck into it through the multiple bronchiolar orifices observed at operation. In this way an equilibrium was established. On the fifth postoperative day the temperature slowly began to drop. By the tenth day it reached normal limits where, except for a few temporary rises, it remained until he was discharged. Concomitantly the respiratory rate fell to an average of 26 per minute, and the pulse dropped to a level of 100-110 per minute.

Repeated physical and roentgenographic examinations demonstrated a slow re-expansion of the right upper lobe and a gradual shift of the heart and

mediastinum toward more normal positions (Fig. 4). The negative pressure was maintained at a constant level of — 18 cm. of water for a period of three weeks, at the end of which time the right upper lobe had expanded and completely filled the right hemithorax. The apex beat was felt in the fifth interspace in the left mammary line. The trachea was in the midline. A bronchial fistula was still present, however, at the site of the catheter. At this time the negative pressure was discontinued and the catheter slowly removed over a period of one week to prevent too rapid closure of the skin edges of the wound (Fig. 5). Six weeks after the operation the bronchial fistula was closed and the wound completely healed. He was discharged on October 30, 1934, about eight weeks after operation. He was then entirely symptom-free and his general condition was excellent.

Microscopic examination of a portion of the cyst wall which was removed at operation revealed some loose vascular connective tissue scattered within which were small extravasations of blood in a state of organization. No pulmonary or bronchial tissue was noted. There was no definite epithelial or mesothelial cyst lining. The drainage from the catheter was mucopurulent in character. On smear many polymorphonuclear leukocytes were seen. Repeated cultures were negative.

Unfortunately, no roentgenographic studies of this patient's chest prior to the present illness are available. The exact sequence of events, therefore, is difficult to unfold. It is felt that he probably always had small air cysts in his right middle and lower lobes and that these cysts were congenital in origin. The bronchioles leading to such cyst "anlagen," however, must have been of such a caliber as to permit the easy passage of air into and out of these cysts. Because of this direct and unobstructed communication, the cysts had never become sufficiently infected or distended to produce symptoms. In April he developed a right-sided lobar pneumonia and pleural empyema. After three months the drainage ceased, and, as far as could be determined by physical examination, the right lung was completely expanded. It is believed that concomitant with the pneumonia there was an associated low-grade bronchitis. The organization of this bronchiolar inflammatory process slowly led to a progressive contraction and distortion of the lumina of the involved bronchioles, until a stage was reached where the passage of air through their narrowed openings became difficult. Thus it is conceivable that repeated respirations led to a slow damming back and accumulation of air within the cysts. The increasing tension of this trapped air resulted in a tremendous expansion of

the cysts with the concomitant collapse of the uninvolved right upper lobe and with a marked shift of the heart and mediastinum into the opposite hemithorax. Apparently an equilibrium was reached when the intracystic air tension reached a mean level of + 12 cm. of water. At this point the pressure was great enough either to prevent the further income of air or to expel with each expiration the same amount of air that the cyst received during inspiration. The above outline of the formation of the lung cysts in this case is purely theoretical and is unfortunately not subject to absolute proof at the present time.

At operation the multilocular cysts were converted into a single large cavity by the division of the septa between them. Negative pressure at a level of —18 to —20 cm. of water was applied in an effort to restore the mediastinum to its normal place and to re-expand the collapsed right upper lobe. This re-expansion was so complete that after 3 weeks this lobe filled the entire right hemithorax and apparently completely collapsed the walls of the large cyst. It was hoped that as a result of the operative manipulation and trauma an inflammatory reaction would be set up in the walls of the cyst which would lead to the formation of adhesions between their now collapsed contiguous walls. (That such an inflammatory response was actually taking place was evidenced by the sterile mucopurulent fluid which drained through the catheter.) Such adhesions, if sufficiently diffuse, would obviously entirely obliterate the cyst. It is felt that this has been accomplished. It is now two years since the operation and the patient is perfectly well and symptom-free. It is not anticipated that he will have any further difficulty from this source.

Summary

Pulmonary cysts include a wide variety of fluid- and air-containing structures which are found involving one or both lungs in all ages. The etiology of these structures is obscure, and many theories have therefore been advanced to explain their genesis. It is believed that pulmonary cysts, like those of the other parenchymatous organs of the body, do not all arise as a result of a single, common mechanism, but are either congenital, inflammatory, mechanical, or neoplastic in origin. The clinical manifestations are protean, and the correct diagnosis is frequently made only at operation or at autopsy. The treatment, as reported in the literature, has been

varied according to the conditions encountered. It has included simple aspiration of air through a needle with or without the insertion of flapper-valve tubes, marsupialization of the cysts, resection of the cysts, lobectomy, and cauterization of the communicating bronchioles.

A new therapeutic procedure is reported in a patient suffering from a tension pneumothorax with multiple, large, air cysts in the right middle and lower lobes. The bronchial openings communicating with the cysts were made so large and numerous that air could not be withdrawn rapidly enough through a needle. An open thoracotomy was performed at which time the walls between the cysts were divided, and a shrunken upper lobe disclosed. High negative pressure served to collapse the cysts completely and to expand the upper lobe until it filled the hemithorax. This negative pressure was maintained until the lung became adherent. Physical examination, as well as roentgenograms, revealed normal lung fields two years after the operation. There has been no recurrence of symptoms.

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