BRONCHIOGENIC CYSTS OF THE MEDIASTINUM Herbert C. Maier, M.D. New York, N. Y.

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A VARIETY OF CYSTS occur in the mediastinum and the vast majority are congenital in origin.⁶ Some arise as a result of developmental aberration of the primitive foregut. The term bronchiogenic is usually applied to cysts arising from the respiratory system, whereas those derived from the digestive tract are termed esophageal or gastric. There is no sharp line of distinction between the bronchiogenic cysts and those arising from the digestive tube. This paper, nevertheless, will be concerned chiefly with the bronchiogenic group. The subject of intrathoracic cysts arising from the digestive tract has been recently discussed by Schwartz and Williams³⁵ and by Ladd and Scott.²⁰ Bronchiogenic cysts of the mediastinum are closely related to aberrant pulmonary tissue¹¹ and congenital intrapulmonary bronchial cysts.¹⁵ As some of the clinical features as well as the therapeutic problems of bronchiogenic cysts of the mediastinum may differ from the intrapulmonary cysts, it seems advisable to consider the former as a separate group. The intrapulmonary bronchiogenic cysts will not be considered in this paper, as the subject has been previously discussed elsewhere^{22, 23} and reviewed by Pugh.²⁹

Until recently bronchiogenic cysts of the mediastinum have been considered to be rare, but with more frequent roentgenologic examinations of the thorax and the widening scope of thoracic surgery, many more cases of this type are being observed. Although some authors have believed the bronchiogenic mediastinal cyst to be usually asymptomatic, an analysis of all the case reports, both in the pathologic and surgical literature, suggests that a considerable number eventually cause symptoms of varying degree. Occasionally a bronchial cyst of the mediastinum causes death in early life by compression of the trachea or main bronchi.

Bronchiogenic cysts of the mediastinum which do not cause symptoms in early life may be found by chance on roentgenographic examination in adult life, or may be an incidental finding at autopsy. In some instances, however, due to the gradual increase in the size of the cyst with resultant pressure on adjacent structures, symptoms of varying degree may lead to clinical investigation and diagnosis. During a two-year period (1943–1944) five patients with a bronchiogenic cyst of the mediastinum were operated upon by the author on the Thoracic Surgical Service of the Memorial Hospital and three additional cases have been operated upon elsewhere.* The eight cases illustrate various features of bronchiogenic mediastinal cysts and some problems in the surgical therapy of this lesion. In addition to reporting these eight cases, the literature of mediastinal cysts is reviewed and the collected cases have been analyzed.

^{*} Two cases at Lenox Hill Hospital and one case at Kings County Hospital.

ORIGIN OF MEDIASTINAL BRONCHIOGENIC CYSTS

A brief discussion of the embryology of the primitive respiratory tract may clarify the problems associated with the development of bronchiogenic cysts.³ The respiratory tract has a common origin with the esophagus from the primitive foregut. As a result of the lateral invasion of two septa, the foregut is divided into a ventral and a dorsal component. These two masses of cells eventually are separated from one another and the dorsal component forms the esophagus, while the ventral component forms the trachea and major bronchi. The close embryological association of the respiratory tract



FIG. 1.—Bronchiogenic cyst. Appearance of inner aspect after evacuation of fluid contents. Note the thin wall and the numerous trabeculations.

with the primitive foregut indicates the possibility of a close association of developmental anomalies of these two structures.

Bronchiogenic cysts result from an abnormal budding or branching of the tracheobronchial tree.³⁴ If the continuity with the bronchial tree is maintained, the cyst is usually intrapulmonary or in intimate association with the lungs. If the mass of cells becomes separated from the tracheobronchial tree, there may be no continuity with the bronchial lumen. Such cysts gradually increase in size because of the distension produced by the secretion within the cavity.

An analysis of the literature reveals a difference of opinion concerning the

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FIG. 2.—A. Photomicrograph of wall of bronchiogenic cyst. Appearance similar to wall of a bronchus. Ciliated columnar epithelium, mucous glands, cartilage, and muscle fibers are present.

B. Higher magnification of cyst wall clearly demonstrates the ciliated columnar epithelium.

stage in embryonic development at which aberrations occur which result in bronchiogenic mediastinal cysts. Although the wall of a typical bronchiogenic cyst is composed of structures similar to that of the bronchial wall, in an occasional case components of the digestive tract are also found in the same cyst wall.³⁷ The latter finding has been advanced as an argument in favor of the contention that the abnormal development begins at an early stage of the embryo before the separation of the respiratory tract from the primitive foregut. Whereas the majority of bronchiogenic cysts are found in relationship to the trachea or main bronchi, in an occasional instance the cyst may be in close association with the esophagus or even the vertebral bodies. These findings lend support to the contention that in at least some instances the misplacement of cells occurred at a relatively early stage of development.

PATHOLOGY

Bronchiogenic cysts usually appear as a single spherical cystic mass, but the cyst may be lobulated, or occasionally there is a chain of cysts. On section the cysts are often single, but may be multiloculated or contain several noncommunicating cavities of varying size. The inner aspect is frequently trabeculated (Fig. 1). The cyst wall is usually relatively thin, although there may be thicker portions. The cyst is lined by ciliated columnar epithelium and the wall contains mucous glands, cartilage, elastic tissue and smooth muscle (Fig. 2A and B). In some instances the cartilage or mucous glands may be absent. The uninfected cyst usually contains a thick, white mucoid material. Even in the absence of infection the cyst contents may be dirty brown⁴⁰ and sometimes closely resembles thick, purulent material. The cyst only rarely has any actual communication with the lumen of the tracheobronchial tree. Secondary infection may produce an inflammatory process which results in destruction of the lining epithelium and may render definite recognition of the bronchiogenic origin uncertain, especially if cartilage is not present in the cyst wall. The cyst may have a definite attachment to a portion of the tracheobronchial tree, but in some instances, although it is adjacent to a bronchus, there may be only loose areolar tissue between the two structures. Occasionally, a bronchiogenic cyst has no apparent connection with the tracheobronchial tree. If the bronchiogenic cysts of the mediastinum are segregated according to their location, certain embryologic, pathologic, and clinical features characteristic of each group may be noted. Bronchiogenic cysts may be arbitrarily divided into the following groups:

- 1. Paratracheal
- 2. Carinal
- 3. Hilar
- 4. Paraesophageal
- 5. Miscellaneous

1. Paratracheal Group. Several instances of a bronchiogenic cyst attached to the tracheal wall have been reported. A characteristic example of this type

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of cyst is illustrated by Case 1. The cyst is usually attached to the right lateral tracheal wall a short distance above the bifurcation of the trachea. The site of attachment of the cyst is similar to the point of origin of the first lateral bronchus of some mammals. Moreover, diverticuli of the trachea have been reported in this same location.⁷ The evidence suggests that bronchial cysts of the trachea occur as a persistence of a structural characteristic of lower forms. The embryology of the tracheobronchial tree in the pig has been thoroughly studied by Flint.¹⁰ He has demonstrated that the lateral bronchial branch from the trachea ordinarily occurs only on the right side. The instances of bronchiogenic cysts in which the cyst had an intimate association with the tracheal wall have almost all been on the right side. An analysis of these data leads to the conclusion that this type of bronchiogenic cyst is explained on the basis of the rudimentary development of a first lateral bronchial branch from Blackader and Evans⁴ reported a paratracheal bronchiogenic the trachea. cyst which compressed, but was not attached to, the left side of the trachea. and which caused the death of a nine-months-old boy.

2. Carinal Group. A number of cases in which the bronchiogenic cyst was located near or just beneath the bifurcation of the trachea have been reported. In some instances there was a definite strand-like attachment extending to the carina.¹⁸ In other cases the attachment was to one of the larger bronchi, whereas in still others the attachment to the bronchial tree was not definitely demonstrable. The frequency with which small bronchial cysts occur in the subcarinal area suggests the possibility of a relationship with other types of congenital anomalies. The bronchiogenic cysts often have a definite attachment to the carina. In some instances there is also an attachment to the anterior esophageal wall, or the cyst may be in the esophageal wall in this region. The location of these cysts corresponds closely with the most common site of congenital tracheo-esophageal fistula. The frequency of embryologic aberrations in this region is striking. The subcarinal bronchiogenic cysts may well represent a pinching off of a group of cells in the communicating channel between the primitive foregut and tracheobronchial tree. The bronchiogenic cyst arising in the carinal area may cause pressure on either the right or left main bronchus, or both.²⁵ If the cyst is large, it may also press on the lower trachea. The cyst may be relatively small and still cause considerable compression of the air passage, due to its rather confined location. Most of the reports of this type of cyst are to be found in the pathologic literature because the diagnosis was unsuspected before death.¹² The majority of the patients died within the first year of life, due to pulmonary complications secondary to compression of the tracheobronchial tree. Although no successful surgical removal of a bronchiogenic cyst in this location in an infant has been reported to date, the lesion should be amenable to surgical therapy if the correct diagnosis were made. In a discussion of the clinical features of this group of bronchiogenic cysts, the clinical picture which should suggest the possibility of the existence of the lesion will be outlined. Whereas some cysts in the same location, chiefly due to their small size, have been chance findings

at autopsy in late adult life, a large percentage of the cases that have been published have caused serious, if not fatal results from secondary pulmonary complications.

3. *Hilar Group*. The majority of bronchiogenic cysts which are diagnosed in the later part of childhood or in adult life are located in the hilar area and have an attachment to one of the main or lobar bronchi. In some instances the attachment to the bronchus is well defined, whereas in other cases only connective tissue strands bind the cyst wall to the bronchus. As the cyst increases in size, it projects into the pleural cavity.²⁴

Because of its close proximity to the main bronchus, it may cause compression of the main stem or one of the lobar bronchi. The secondary pulmonary changes which may occur will be described under the clinical findings. A considerable portion of a large cyst may be in close proximity to the pericardium and the cyst may project between the various hilar structures of the lung. In one of my patients there was an anomalous distribution of the hilar structures of the right lower lobe and the cyst was intimately associated with these structures and displaced them to a considerable degree. In this case there was an extension of the cyst across the mediastinum almost into the opposite hemithorax.

The bronchial cysts occurring in the hilar area are embryologically closely related to aberrant pulmonary tissue and anomalous accessory lobes of the lung. A review of the subject of aberrant intrathoracic pulmonary tissue has been presented by Friedlander and Gebauer.¹¹

Secondary infection of the bronchiogenic cyst may result in considerable destruction of the cyst wall, rendering identification difficult. Moller²⁷ has reported a case of bronchiogenic cyst which ruptured into the tracheobronchial tree with the development of secondary infection. The cyst was in an unusual location, in that it lay in the anterior mediastinum. At operation the communication between the cavity of the cyst and the bronchial tree could not be demonstrated.

4. Paraesophageal Group. Some bronchiogenic cysts may be in close relationship with the esophagus and have little or no apparent connection with the tracheobronchial tree. In some instances the bronchiogenic cyst is merely in close proximity or loosely attached to the wall of the esophagus. In other instances, however, the cyst is entirely within the walls of the esophagus and bulges into the esophageal lumen, only the mucous membrane of the esophagus covering the inner aspect of the cyst wall. If an intramural esophageal cyst is lined by ciliated columnar epithelium but contains no cartilage within its wall, the bronchiogenic origin may be seriously questioned.^{26, 19} Cysts of this type located in the lower esophagus near the cardia have been reported by v.Wyss,³⁹ Zahn,⁴² Rau,³⁰ Tresp,³⁸ and Westenryk.⁴¹ Robbins³² reported a bronchiogenic cyst within the wall of the esophagus which contained cartilage, and was lined by ciliated columnar epithelium. A number of cases have also been reported in which a cyst lined by ciliated columnar epithelium was found attached to the outer portion of the esophagus. In several of these same cases a well-defined double muscle layer was present in the wall of the cyst.⁹ In one of my cases (Case 2) the cyst extended for a considerable distance parallel with and to the right of the esophagus. There were three separate cyst cavities extending from the level of the thoracic inlet down beneath the arch of the vena azygos to the level of the pulmonary hilum. The paraesophageal location of the cyst in this case, as well as the thick double layer of muscle in the wall, suggested a re-duplication of the alimentary tract. Moreover, no cartilage was found in the cyst wall. Although the lining of the cyst was typical ciliated columnar epithelium suggesting a bronchial origin, the presence of ciliated columnar epithelium lining the esophagus at one phase of embryological development makes the matter inconclusive.

5. Miscellaneous Group. Occasionally a bronchiogenic cyst is found in a very unusual location. Two cases have been reported in which the cyst was found within the pericardium.^{17, 28} Rusby and Sellors³³ reported a case of bronchiogenic cyst associated with a congenital deficiency of the pericardium. The patient had symptoms which were attributed to the bronchial cyst. This was removed at operation which was followed by an uneventful recovery. A defect in the pericardium was also present in one of my cases (Case 7). The bronchiogenic cyst was located anteriorly in close proximity to the site of congenital absence of a portion of the upper part of the pericardium. Guillery¹³ found a cyst lying on the anterior aspect of the thoracic spine and extending into the vertebral bodies, together with a smaller cyst of similar type behind the vertebral bodies. In this case no cilia were demonstrated on the cylindrical epithelium which lined the cyst. The muscle layers were more suggestive of the wall of the digestive tract than that of the respiratory system. The infant had died at the age of three months. The portion of the cyst which projected forward from the thoracic vertebrae had caused compression of the bronchi, especially on the right side.

Seybold and Claggett³⁶ reported a presternal cyst lined by ciliated, pseudocolumnar epithelium, which was located in the subcutaneous tissues at the sternal angle. The walls were thin and contained mucous glands, cartilage, vessels and nerves. This cyst bore a striking similarity to a bronchiogenic cyst of the mediastinum, and it was considered that the cyst had migrated into a presternal position. The underlying sternum showed no abnormality.

SYMPTOMATOLOGY

In the absence of infection, the symptomatology of bronchiogenic cysts depends chiefly on the size and location of the mass. In some instances there are no symptoms referable to the lesion, and the presence of the cyst is only demonstrated by a routine roentgenogram of the chest, or is a chance finding at autopsy. In general, the symptoms presented are those produced by compression of the tracheobronchial tree. In some of the patients in whom a bronchiogenic cyst was removed because of the finding of an undiagnosed mass on a roentgenogram, the symptoms were rather vague and not characteristic, so that without a follow-up study it is difficult to ascertain whether or not the symptoms were caused by the cyst. When secondary infection supervenes, the symptoms resemble those of intrathoracic suppuration, particularly those of mediastinal or pulmonary abscess. Occasionally there is prominence of the chest wall in the region of the cyst. The symptomatology in special groups will now be discussed.

I. Paratracheal Group. In the few cases of this type that have been reported, no characteristic symptoms have been present. One case had previously had an empyema on the same side, but at the time of operation ten years later no infection was present. Due to its location, the cyst may cause considerable narrowing of the trachea, as is illustrated in Case I, but the compression is usually insufficient to cause serious obstruction.

2. Carinal Group. When the bronchiogenic cyst is located just beneath the bifurcation of the trachea and causes symptoms in early life, the history may indicate some respiratory difficulty at or shortly after birth. In other cases nothing abnormal is noted until a respiratory infection develops. The natural tendency is to ascribe the symptoms to the pulmonary infection. Sometimes without such an evident precipitating factor, difficulty in breathing may be noted. Clinical and roentgen examination may indicate either obstructive emphysema or atelectasis, depending upon the degree of bronchial obstruction. Wheezing may be present; a croupy cough is sometimes noted. Varying degrees of dyspnea and cyanosis may develop. Progressive respiratory difficulty ensues and frequently results in a fatal outcome.² Analysis of the literature suggests that in those cases in which the subcarinal type of cyst causes symptoms in early life, the mortality rate is extremely high.

In some infants noisy breathing or attacks of cyanosis, especially during crying, may have been noted since birth. At times expiratory stridor is present. If the wheezing has been present for a considerable period of time, an asthmatic condition may erroneously have been considered the explanation of the symptoms. Adams and Thornton¹ have reported a bronchiogenic cyst in this location which first caused symptoms in adult life when secondary infection supervened. Robbins³² reported a bronchiogenic cyst in an 18-year-old male which was located just beneath the carina. This cyst measured 8 x 6 cm. and caused an irritating, non-productive cough apparently due to pressure on the main bronchus. Surgical removal was successfully carried out. Similar cysts have occasionally been chance findings at autopsy in adults.³¹

3. Hilar Group. Bronchiogenic cysts arising in the area of the pulmonary hilum usually project a varying amount into the pleural space on that side. These lesions are often asymptomatic, but when symptoms are present, these are usually due to compression of a portion of the bronchial tree. Dull chest pain, dry cough, wheezing and frequent respiratory infections may occur. In occasional cases, such as in Case 4, moderate bronchial compression over a considerable length of time may cause chronic pulmonary infection which causes a productive cough. This same patient had slight discomfort on swal-

lowing, due to displacement of the esophagus by the cyst. In one of my patients there were no complaints prior to the accidental finding of the bronchiogenic cyst on a routine roentgenogram of the chest. Following removal of the cyst, however, the patient gained considerable weight and felt far better than he had for many years. Undoubtedly in this case the cyst was causing definite although unrecognized disturbance, probably due to the secondary changes in the lung which, however, had failed to cause cough or expectoration.

4. Paraesophageal Group. When the bronchiogenic cyst occurs in close relationship with or within the wall of the esophagus, there may be symptoms referable to that organ. Some dysphagia may be noted. The symptoms are similar to those encountered in benign tumors within the wall of the esophagus. Occasionally the lumen of the esophagus above the lesion is dilated. If the cyst is chiefly outside the wall of the esophagus, the patient may be asymptomatic.

5. *Miscellaneous Group*. In the cases of bronchiogenic cyst of the mediastinum occurring in miscellaneous locations, the only characteristic symptom complex is that which occurred in the two cases of intrapericardial cysts. These patients died suddenly, apparently owing to pressure on a portion of the heart or great vessels.

DIAGNOSIS

Until the last few years a diagnosis of bronchiogenic cyst has rarely been made before operation or autopsy. In some instances the clinical and roentgenologic picture presents nothing sufficiently characteristic to differentiate the lesion from other types of mediastinal tumor. There are, however, certain clinical and roentgenologic features which may lead to either a probable, or even in some instances, a definite diagnosis prior to operation.

I. Paratracheal Group. As the clinical findings in this group are not characteristic, the roentgen examination would seem to be the sole evidence upon which a diagnosis might be made prior to operation. Bronchiogenic cysts would have to be differentiated from other tumor masses in the upper mediastinum lying in close proximity to the trachea. A substernal thyroid, a thymic tumor, an intrathoracic hygroma, a serous cyst and an aneurysm are the chief lesions to be considered in a differential diagnosis. A substernal thyroid often surrounds the trachea to a greater extent than a tracheal cyst, which is usually located entirely to the right of the trachea. The borders of the mass as seen radiographically are generally more sharply defined in a cyst, as contrasted to a soft tissue mass such as a thyroid adenoma. Both lesions, however, may show calcification.

A thymic tumor may have a less well defined border on the roentgenogram, usually projects bilaterally from the mediastinum and is located more anteriorly. The upper limit of a paratracheal cyst will usually be above the level of the clavicle on the roentgenogram, whereas a thymic tumor begins below the clavicle and extends further downwards. Differentiation of a bronchiogenic cyst attached to the trachea from an intrathoracic hygroma or serous cyst may offer real difficulty. If aspiration is performed, thick white or yellow mucoid material may be obtained from the bronchiogenic cyst in contrast to the clear watery fluid in the hygroma or serous cyst.

An aneurysm, particularly of the innominate artery, might give a somewhat similar roentgen appearance. Angiocardiography may definitely establish whether the mass is of vascular origin. An aneurysmal sac filled with a laminated clot, however, will not be delineated by the opacified blood, and therefore this diagnostic procedure does not always give positive information, particularly if the degree of opacification is somewhat unsatisfactory.

2. Carinal Group. The diagnosis of the subcarinal bronchiogenic cysts, especially when producing symptoms in infants, has apparently been very difficult in the past. As mentioned in a discussion of the symptomatology, the picture is essentially that of a bronchial, or occasionally lower tracheal, obstruction. It is usually erroneously assumed that the obstruction is produced by an inflammatory process within the air passages, whereas actually the pulmonary infection is secondary to bronchial compression by the cyst from without. A roentgenogram of the chest may give no indication of the presence of a mediastinal tumor because the cyst is relatively small and is completely hidden in the mediastinal densities. If the possibility of a cyst were borne in mind, an oblique film might be helpful in delineating the mediastinal mass, provided the superimposed densities from the pulmonary infection.

A clue to the true nature of the lesion may be obtained by bronchoscopic observation in certain cases. If bronchoscopy shows no endobronchial lesion but external compression of the lower trachea, or one or both main bronchi, a mediastinal tumor of some type must be ruled out. Other lesions, however, may also cause extrinsic pressure on the lower trachea. Congenital anomalies of the large vessels may be associated with obstruction to trachea or esophagus, or both. Operative intervention might be advisable in either situation.

3. *Hilar Group*. Apparently the most frequent preoperative diagnosis in patients with bronchiogenic cyst reported in the literature has been mediastinal dermoid. A point which should be of considerable aid in differentiating these two lesions is that the dermoid tumors arise in the anterior mediastinum, whereas the bronchial cysts are more frequently located in the posterior mediastinum. A few dermoid cysts have been reported as arising in the posterior mediastinum, but on reviewing these cases, most of them are found to be instances of bronchiogenic cysts rather than mediastinal dermoid.¹⁴ In some instances bronchiogenic cysts have been confused with neurogenic tumors. On the lateral roentgenogram neurogenic tumors are seen to be located in the costovertebral portion of the thorax, whereas the bronchiogenic cysts are in the posterior mediastinum anterior to the vertebral bodies.

Robbins³² has recently reported the roentgenologic findings in several cases of bronchiogenic cysts. He points out that bronchiogenic cysts may move

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with respiration and also may change shape with respiration, indicating their cystic nature and relationship with the tracheobronchial tree. This finding may be of aid in differentiating bronchiogenic cysts from certain other mediastinal tumors. Dermoid cysts have a rather stiff wall and usually do not change contour with respiration. Included in Robbins' report are several cysts in which the histopathologic findings do not definitely demonstrate the origin of the cyst. The author included these cases because he thought they may be of value in linking the other cases of so-called simple or unclassified cysts of the mediastinum into one group. Whether these other cases are actually of similar origin to true bronchiogenic cysts cannot be stated at this time. In two of the cases the cyst was located in the anterior mediastinum in close relationship to the pericardium and diaphragm. In neither of these cases was the cyst wall entirely typical of a bronchiogenic cyst, and the radiologic and histologic findings suggested the possible diagnosis of pleural cyst of the type discussed by Lambert.²¹ Also included in the report of Robbins are cases of bronchiogenic cysts within the substance of the lung and an intrapulmonary cyst-like lesion secondary to chronic abscess, which are not to be confused with the cases considered in the present paper.

Rarely a mediastinal bronchiogenic cyst perforates into the lung, with resultant bronchial communication. Then the roentgenologic and clinical picture simulates either a mediastinal or pulmonary abscess. As bronchiogenic cysts are closely related to aberrant pulmonary tissue, a cyst-like cavity lined with respiratory epithelium may be formed as a budding from the bronchial tree. Harrington¹⁵ has reported a case in which the azygos lobe consisted of a sac similar to the wall of a bronchiogenic cyst. There was a small bronchial communication with the upper lobe. The connection with the tracheobronchial tree permitted the partial evacuation of the cyst contents and resulted in the presence of a fluid level within the cyst cavity at one roentgenographic examination. A few other instances of bronchiogenic cyst of the mediastinum with a fluid level have been reported, but this finding usually indicates that the cyst is intrapulmonary.

4. Paraesophageal Group. A bronchiogenic cyst may be in close association with the esophagus, or even the cardiac portion of the stomach. The lesion must be differentiated from solid tumors of the esophageal wall.

A considerable number of cases are on record in which a cyst lined by ciliated epithelium has been found within or in close association with the wall of the esophagus. In the majority of instances no cartilage has been found in the cyst wall, and therefore it is questionable whether they should be classified with the bronchiogenic group. Those cases without cartilage might be considered as having resulted from the pinching off of the cells from the esophageal wall itself, since the esophagus is lined by ciliated columnar epithelium at one time in its embryological development. The paraesophageal cyst lined by ciliated columnar epithelium should be differentiated from paraesophageal cysts which are lined by mucous membrane similar to that of the stomach or small intestine. Paraesophageal cysts lined by gastric mucosa may

secrete pepsin and acid which result in ulceration and even erosion of adjacent structures. Examination of the contents of the cyst may aid in the differential diagnosis of a paraesophageal bronchiogenic cyst from a thoracic gastric cyst. Both types can bear a similar relationship to the esophagus, and it would not always seem possible to make a differential diagnosis on the basis of clinical and roentgen findings alone. An analysis of the literature suggests that the thoracic gastric cysts are far more likely to lead to symptoms and surgical intervention in early life than the paraesophageal bronchiogenic group. Schwartz and Williams³⁵ collected ten cases from the literature and added two of their own. Half of the cases of thoracic gastric cysts caused symptoms or death within the first year of life, and only two of the 12 cases were over four years of age at the time of operation or autopsy. Probably the gastric mucosa secretes more rapidly and leads to more rapid enlargement of the cyst, with secondary compression of the lung. The mediastinal cysts of enteric origin usually project from the right side of the mediastinum, and although the bronchiogenic cysts are also more common on the right side, there is not the overwhelming predilection for the right side as is seen with those of enteric origin. No obvious sex predominance has been noted in either group. Robbins³² reported a case of a small bronchiogenic cyst which was located in the wall of the esophagus and appeared as an intramural, extramucosal lesion. The mucosal folds of the esophagus were preserved over the lesion and the mass was noted to move up and down with respiration and upward during the act of swallowing. The motion of the mass was similar to that of the esophagus. At operation the cyst was removed from the wall of the esophagus, and histopathologic examination showed respiratory ciliated epithelium, smooth muscle and cartilage in the wall of the cyst.

TREATMENT

In a monograph on mediastinal tumors published in 1940. Heuer and Andrus¹⁶ state that they had found in the literature 25 cases of mediastinal cysts. This group of cases apparently includes both bronchiogenic and esophageal cysts. In 12 instances an operation had been undertaken for possible removal of the cvst, but the tumor was excised in only 8 of these cases. In 5 instances the cyst was extirpated at the first operation and in 3 its removal followed drainage and marsupialization. Operation in the remaining 4 patients consisted either of drainage of the cyst or its partial removal. All but one of the reported cases in whom complete removal of the cyst was possible recovered, but one of them had a persistent bronchial fistula. The authors also reported a personal case in which recovery followed excision of the cyst. During the past few years several reports on the surgical extirpation of bronchiogenic mediastinal cysts have appeared. The recent increase in routine roentgenographic examination has been responsible for the finding of a large number of these cases in which the patient was asymptomatic. Bronchiogenic cysts of the mediastinum were considered rare a few years ago, but are known today to be one of the most common tumors of the mediastinum.

Adams and Thornton¹ reported three cases of bronchiogenic cyst of the mediastinum treated successfully by surgery. Brown and Robbins⁵ analyzed 12 cases of mediastinal cyst from the Massachusetts General Hospital. In 6 of the 12 cases the bronchiogenic origin of the cyst may be considered definite on the basis of the histologic appearance of the cyst wall, and in one additional case the gross findings at operation leave little doubt about the diagnosis. In the remaining cases the cyst wall contained no definite bronchiogenic elements.

It is often difficult definitely to diagnose the exact nature of a mediastinal tumor preoperatively. As mediastinal tumors in general should be removed if proper facilities are available and no contraindication exists, a failure to make a correct preoperative diagnosis is not necessarily a great disadvantage. Whereas a considerable number of the bronchiogenic cysts which have been reported in the literature caused few if any clinical symptoms, a sufficiently large percentage cause complications, especially secondary respiratory infections, to warrant surgical removal. Naturally, if the patient's general condition is such as to increase the hazard of surgical intervention, it may be best to merely observe the patient closely if it is considered quite certain that the lesion is a benign cyst.

The author recommends a posterolateral transpleural approach because of the impossibility of ascertaining preoperatively the various attachments of the cyst. A posterolateral approach permits access to all parts of the pleural space and mediastinum. It may not be possible to remove the cyst intact. If the patient has manifested no signs of infection preoperatively, and if the findings at operation do not suggest suppuration, opening of the cyst and evacuation of its contents in the course of its excision would not seem to be hazardous. One wonders whether some of the empyemas following the opening of a bronchiogenic cyst reported in the older surgical literature may not have been due to other causes than opening the cyst.8 In three of our patients the cyst was opened at operation and there was gross contamination of the pleural space, but no pleural infection ensued. In one patient, (Case 4) it would have been impossible to remove the cyst intact. Before the cyst was opened, it was felt that complete removal might not be possible because of the marked extension of the cyst through the posterior mediastinum to the contralateral side, and because of its close relationship to the pericardium. After the cyst had been opened and evacuated, with collapse of the walls, it was possible to define the limits of the cyst more accurately, and then remove the cyst wall by combined sharp and blunt dissection.

As the cysts are on a congenital basis, the possible presence of other anomalies must be borne in mind. In one patient (Case 4), there was an anomalous pulmonary vein to the right lower lobe which had to be carefully dissected from the cyst wall. The inferior pulmonary vein was many times its normal length and encircled the lower border of the cyst. If this vein had been injured a lobectomy would have had to be performed, in addition to the

removal of the cyst. Similar anomalies of the vascular system may be found in cases of intrapulmonary bronchiogenic cysts.

If complete removal of the cyst wall seems hazardous, it would seem permissible to leave a small portion *in situ*. Adams and Thornton¹ have utilized silver nitrate to destroy the remaining epithelial lining. Incomplete removal of a bronchiogenic cyst is not as likely to lead to complications as partial removal of gastric or dermoid cysts.



FIG. 3.—Case I: Paratracheal type of bronchiogenic cyst. Note that upper border of mass does not descend from cervical region in manner commonly seen with substernal extension of a thyroid.

In the paratracheal bronchiogenic cyst, injury to the tracheal wall must be carefully avoided. In our case the cyst had a common wall with the trachea over an area of approximately 2 cm. with a direct continuation of cartilaginous rings from the tracheal wall into the wall of the cyst. These small cartilaginous fragments were removed level with the tracheal wall, but a very small portion of the cyst wall was actually left adherent to the trachea. In this instance it was not deemed advisable to apply any escharotic to destroy the residual lining because of the danger of denuding the cartilage, and the development of a chondritis. Although there were some recesses in the attachment between cyst and trachea, careful probing failed to reveal any continuity with the lumen of the trachea.



FIG. 4.—Case 2: Bronchiogenic cyst projects laterally above right hilar region. Considerable scoliosis due to hemivertebrae is present.

In operations for mediastinal tumors it is a great advantage to get complete early expansion of the lung. For this reason all the residual air is aspirated from the pleural space through a catheter as soon as the chest wall has been rendered air-tight by closure of the muscles of the thoracic wall. The catheter is then withdrawn as a mattress suture is tied. In some instances closed drainage for a day or two may be desirable.

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Most of the serious complications in the small number of surgical procedures that have been carried out for bronchiogenic cysts occurred in the case reports of earlier years and were related to the technic of thoracic surgery in general at that time, rather than to any particular problems encountered in bronchiogenic cysts. Seven of my eight patients had an uneventful postoperative course. One patient, who had mild symptoms of hyperthyroidism prior to operation as well as considerable pulmonary infection from bronchial compression, had a rather stormy postoperative course. At operation the cyst wall was dissected from a large part of the pericardium. This operative manipulation, superimposed on a mild hyperthyroidism, apparently precipi-



tated auricular fibrillation and flutter several days postoperatively. The cardiac abnormality rapidly responded to digitalis, and the patient had an otherwise uneventful course.

CASE REPORTS

Case 1.—R. W., #67865, male, age 26 years. Eight years prior to admission a routine chest roentgenogram had shown widening of the upper mediastinal shadow which was interpreted as an enlarged thymus. Subsequent roentgenograms showed little increase in the size of the shadow, until four months before admission to Memorial Hospital when another film showed a definite increase in the size of the mediastinal density. The patient did not complain of any chest pain, cough, expectoration, hemoptysis or dysphagia. There had been no change in weight and his general health was good. Physical examination

was essentially negative. Roentgenograms of the chest in postero-anterior projection (Fig. 3) showed a shadow of considerable density projecting from the right upper mediastinal shadow. The shadow began a short distance above the upper margin of the clavicle and had a slightly irregular but sharply defined border, and extended down to the level of the vena azygos. The outer border of the shadow projected laterally several centimeters beyond the normal mediastinal density. The lateral film showed the density to occupy the upper posterior mediastinal area. There was considerable narrowing of the tracheal air column, especially from the anterior aspect. Roentgenogram of the esophagus revealed no abnormality and no compression by the tumor mass. Angiocardio-graphic studies showed no obstruction or displacement of the superior vena cava, and indicated that the mass was not an aneurysm. Bronchoscopic examination revealed only



FIG. 6.—Case 3: Small bronchiogenic cyst projects from right side of mediastinum just below level of inner end of clavicle.

compression of the trachea from the right lateral and anterior aspect. The basal metabolic rate was minus 16.

Operation.—In the upper posterior mediastinal region there was bulging of the mediastinal pleura caused by an underlying tumor mass. The mass felt cystic with areas of calcification. The mass extended from the level of the thoracic inlet down to and slightly behind the vena azygos. It extended from the vertebral column to the innominate artery anteriorly. The tumor mass was loosely adherent to the superior vena cava and vena azygos and also rather closely associated with the vagus nerve. The mass, which measured approximately 10 x 6 cm., was adherent to the right lateral wall of the trachea throughout the greater portion of its extent, but was continuous with the tracheal wall

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for a distance of about I centimeter only. In this area the trachea and cyst wall had a common partition and cartilaginous plaques could be felt extending from the trachea into the base of the cyst. The wall of the cyst was thin and the cyst contained thick, white, non-odorous mucoid material. There was no sign of inflammation. The right lung appeared normal. The entire cyst was removed, with the exception of the small portion which had its wall in common with that of the trachea. No sinus into the trachea could be demonstrated with the aid of a fine probe. Although it was realized that the small portion left attached to the trachea had an epithelial lining, it was deemed inadvisable to apply any caustic because of danger of necrosis of the cartilage of the tracheal wall. The postoperative course was uneventful. Subsequent roentgenograms showed the tracheal air



FIG. 7.—Case 4: A. Postero-anterior roentgenogram reveals area of density merging with right heart border. Note infiltration in right lower lung field.
B. Lateral film shows mass in posterior mediastinum.

column to be of normal dimensions. Three years after operation the patient was asymptomatic.

The surgical specimen consisted of a cystic structure with thin walls containing many irregular pieces of cartilage. Most of the cyst wall measured only 2 mm. in thickness. There were trabeculations in the interior of the cyst (Fig. 1). Microscopic examination revealed that the wall of the cyst resembled the wall of a bronchus. The cyst was lined by ciliated columnar epithelium. The wall contained mucous glands, cartilage, smooth muscle and elastic fibers (Fig. 2).

Case 2.—T. M. #70313, male, age 23 years. Patient had apparently been well until three years previously, when he noted marked weakness and pallor. On examination a very severe anemia was found. A gastrointestinal series was done, but no evidence of peptic ulceration was discovered. A roentgenogram of the chest, however, revealed a mass in the right posterior mediastinal area. Five blood transfusions were given with resultant correction of the anemia. Because of the roentgenologic findings, a diagnosis of probable lymphosarcoma or Hodgkin's disease of the mediastinum was made at that

time, and the patient received radiation therapy. A total of 2,600 roentgens through two ports was given without significant change in the size of the tumor mass. Neither at that time nor in the following three years did the patient complain of any cough, expectoration or chest pain. There had been no dyspnea or difficulty on swallowing. Curvature of the upper thoracic spine had been present since infancy. The patient was referred to Memorial Hospital in 1943, three years after the mass in the chest was first discovered.

Physical examination revealed a well developed and nourished young male. There was rather marked scoliosis of the upper thoracic spine. Physical examination of the heart and lungs was essentially negative. Roentgenograms of the chest showed an irregular, somewhat rounded mass projecting from the right side of the mediastinum (Fig. 4). Lateral film showed the mass to be in the posterior mediastinum close to the esophagus. A barium study revealed no displacement of the esophagus. There was scoliosis of the upper thoracic spine due to hemivertebrae.

Operation.—Located in the posterior mediastinum, beginning at the level of the 7th rib and extending upward beneath the arch of the vena azygos to the thoracic inlet, was



FIG. 8.—Case 4: Photomicrograph of cyst wall shows ciliated columnar epithelium, smooth muscle, and mucous glands.

a well encapsulated, lobulated, cystic tumor mass measuring about 10 cm. in its greatest length. The tumor consisted of three apparently separate cystic areas which were attached to each other (Fig. 5). The cystic tumor was not attached to the trachea, but was more closely associated with the right lateral esophageal wall, although there was actually no definite attachment to the esophagus. The tumor was removed after division of the azygos vein. The postoperative course was essentially uneventful. The patient has been well for three years.

The specimen consisted of a cylindroid-shaped tumor measuring 9 cm. in length and 3 cm. in diameter. There were two areas of constriction, so that the specimen had the appearance of being composed of three separate portions. The outer surface showed a fairly smooth capsule. On section, the mass was seen to consist of three definite, apparently non-communicating cysts. All three cysts were filled with semi-viscid, brownish, slightly oily material. The lining of the middle cyst and the upper cyst was smooth and shining, while the lower was finely granular. The cyst wall was firm and white, and

varied from 1 mm. to 1 cm. in thickness. Microscopic examination of the cyst wall showed all the component structures of a bronchial wall except cartilage.

Case 3.—L. F., #70359, male, age 18 years. Two months prior to admission a roentgenogram of the chest had shown a small, rounded density projecting from the right side of the mediastinum. The patient's only complaint was a slight, dry cough which he had had for several years and which he attributed to smoking.

Physical examination revealed a well developed and nourished young male. General examination was essentially negative. No thoracic abnormality could be detected by percussion or auscultation. Roentgenogram of the chest revealed a small, rounded shadow projecting out from the mediastinal density on the right side opposite the level of the arch of the aorta (Fig. 6). The lung fields were clear. On the lateral film the mass could be vaguely outlined in the posterior mediastinum.



FIG. 9.—Case 5: A. Postero-anterior roentgenogram reveals mass in upper portion of right hilar region. Infiltration of the right upper lobe is evident. B. Lateral roentgenogram reveals mass in posterior mediastinum.

Operation.—A small multilobular, cystic mass was found just below the vena azygos and attached to the right main bronchus by a small pedicle. The cystic mass was 'oosely imbedded in the posterior aspect of the right upper lobe, from which it could be separated with ease. The cyst was grayish-white in appearance and the wall was very thin. The entire cystic mass was removed without difficulty. There was no communication between the cyst and the lateral aspect of the right bronchus, to which it was attached. The patient's postoperative course was uneventful. Patient has been well for three years, except for a slight cough.

The specimen consisted of a multilobulated mass 5×5 cm. Cut section showed the cyst to contain fluid which was jellied by previous fixation. The cyst was only a few millimeters in thickness. Microscopic examination of the cyst wall showed the various components of a bronchus.

Case 4.—S. B., #70577, female, age 33 years. This patient complained of increasing productive cough of two years' duration with mucopurulent sputum which was frequently blood-tinged. Slight dysphagia had been present for six months. For two months there

had been some pain in the right posterior thoracic region. Physical examination revealed a fairly well developed and nourished negress who did not appear acutely ill. Slight exophthalmos was present and the thyroid was symmetrically enlarged and firm. There was slight tremor of the hands. Examination of the lungs revealed some diminution of breath sounds at the right base posteriorly with occasional rales. The heart was slightly overactive, but no murmurs were heard. Roentgenogram of the chest showed a rounded density projecting from the right lower mediastinal shadow continuous with the density of the right cardiac border. There was scattered infiltration in the right lower lung field (Fig. 7A). On a lateral film the mass was seen to lie in the posterior mediastinum (Fig. 7B). Barium studies showed the esophagus to be displaced toward the left by the mass, but there was no obstruction. The basal metabolic rate was plus 10. It was thought that the patient had a bronchiogenic cyst with compression of the bronchial tree and secondary pulmonary infection with probable bronchiectasis. Bronchoscopy was not done preoperatively because of the patient's refusal following an unsuccessful attempt elsewhere. It was thought the patient had mild hyperthyroidism, but insufficient to contraindicate the thoracic operation as the primary surgical procedure.



FIG. 10.—Case 6: A. Roentgenogram shows double bronchiogenic cyst projecting from right upper mediastinum.

B. Tomograph shows more clearly the outline of two cysts.

Operation.—Avascular adhesions were present over the lower lobe. A cystic tumor mass occupied the posterior mediastinal area. Considerable difficulty was encountered in freeing the cyst from the inferior pulmonary vein which was elongated and closely associated with the wall of the cyst. During mobilization of the cyst it was ruptured and the contents removed by suction and sponge. The collapsed cyst wall was then dissected free by sharp and blunt dissection. The cyst wall varied from I mm. to 4 mm. in thickness. Microscopic examination revealed findings typical of a bronchiogenic cyst (Fig. 8).

Two days postoperatively the patient developed auricular fibrillation which was treated by digitalization and Lugol's solution. Auricular flutter occurred for a brief period. It was thought that the cardiac complication was due to a combination of mild hyperthyroidism and extensive operative manipulation in the region of the auricles. The cardiac rate became normal within a few days. The patient also had considerable cough and expectoration, undoubtedly due to the preoperative pulmonary infection. The

patient was discharged on the 16th postoperative day with the wound well healed. A thyroidectomy was performed later. During the three years since operation the patient has had a productive cough. A resection of the bronchiectatic portion of the lung has been recommended.

Case 5.—J. L., 73999, male, age 35 years. In March, 1943, this patient had a left-sided pleurisy. A respiratory infection developed in January, 1944, following which he had frequent cough with moderate expectoration but no hemoptysis. In March, 1944, he had dyspnea on exertion. A routine chest roentgenogram, taken at an army induction center, revealed a mass in the right paramediastinal area. Patient was admitted to the hospital in May, 1944. Physical examination revealed a somewhat undernourished male who did not appear ill. Physical examination of the chest was essentially negative. Roentgenogram showed a right paramediastinal area of density with infiltration in the adjacent lung



FIG. 11.—Case 7: A. Roentgenogram shows sharply demarcated mass projecting from left hilar region.

B. Lateral roentgenogram shows anterior location of cyst.

(Fig. 9A). On lateral view the density was found to be in the posterior mediastinum (Fig. 9B). There was another small area of infiltration in the right upper lobe separate from the main mass. Fluoroscopy showed no pulsation of the mass. Bronchoscopy was negative.

Operation.—A bronchiogenic cyst of the mediastinum extending into the right upper lobe was found. Evidence of considerable inflammation was present and there was a secondary bronchiectasis of the right upper lobe. The cyst and upper lobe were removed. The postoperative course was essentially uneventful. Within seven months after operation the patient had gained 30 pounds in weight, so that he weighed more than at any previous time. Roentgen-ray showed good expansion of the remaining portion of the right lung.

Pathologic examination showed the wall of the cyst to be largely replaced by granulation tissue, but a respiratory type of epithelium and areas of squamous metaplasia were also found. The excised lobe revealed bronchiectasis.

Case 6.—H. S., Lenox Hill Hospital #108799, male, age 30 years. One year prior

to admission a mass was discovered in the paramediastinal region on a draft board roentgen-ray. Subsequent roentgenographic studies showed a double mass projecting out from the right side of the mediastinum. Patient had no cough or sputum, no chest pain, and his weight was increasing.

Examination revealed a robust male without abnormal physical findings. Fluoroscopy showed a density extending out from the right mediastinal area, which on lateral projection was in the posterior mediastinum. Roentgenograms showed two smooth, oval shadows of increased density, each about 5 cm. in diameter, in the posterior portion of the right upper lobe near the mediastinum (Fig. IOA and IOB). These shadows were in close contact with each other. Their anterior surfaces were in close relation to the right main bronchus. Bronchoscopy showed no evidence of narrowing of the trachea or bronchi but the bronchus of the right upper lobe was somewhat larger than normal.



FIG. 12.—Case 8: Large rounded area of density is present in the left upper portion of thorax.

Operation.—A dumb-bell shaped cystic mass was palpable in the hilar region of the right upper lobe. A small portion of this cystic mass could be seen on the posterior aspect of the hilum, but the major portion of the mass was covered by the medial portion of the right upper lobe with which it was intimately associated. The cyst projected for a considerable distance into the pulmonary parenchyma, although there was a line of cleavage between the cyst and the normal pulmonary tissue. The cyst was fairly thinwalled, had cartilaginous plaques within it, and contained non-odorous, greenish-gray, thick material. There was a strand-like thickening in the pulmonary tissue extending from the upper portion of the larger cyst to the apex of the lung, where there was a smaller cyst about 2 cm. in diameter which contained material similar to the larger cyst. Otherwise the pulmonary tissue of the upper lobe appeared relatively normal. The middle

and lower lobes were air-containing and appeared normal. The cysts were excised and the patient's postoperative course was uneventful.

Microscopic examination revealed the inner surface of the cyst to be partially lined with tall, ciliated columnar epithelium supported by a congested stroma which was richly infiltrated with round cells. Embedded in the stroma was an occasional small mucous gland. Portions of the cyst showed marked fibroblastic proliferation, and was densely infiltrated with inflammatory cells, including a few multinucleated giant cells of the foreign body type. In the deeper layers of the wall there was extensive fibrosis and a perivascular focal round cell infiltration.

Case 7.—R. D., Lenox Hill Hospital #109712, female, age 22 years. On a routine chest roentgenogram taken during a physical examination, a mass was found in the left



FIG. 13.—Case 8: A. Roentgenogram taken after aspiration of cyst contents and replacement with air and small amount of lipiodol.

B. Lateral view of the same.

mediastinal region. The patient never had any symptoms referable to the thorax. Physical examination was essentially negative. Roentgenogram of the chest showed a rounded density projecting from the left side of the mediastinum slightly above the hilar region. The mass was of homogeneous density and the margins were smooth (Fig. 11A). A lateral film showed that the mass was located in the anterior mediastinum (Fig. 11B). Preoperative diagnosis was dermoid cyst.

Operation.—A large, rounded, relatively thin-walled cyst occupied the upper anterior aspect of the left pleural space, projecting into it from the mediastinum. The wall of the cyst contained cartilaginous plaques. The attachment of the cyst was near the anterior aspect of the hilum of the left lung in close association with the left main bronchus but not actually attached to the bronchus. Anterior to the attachment of the cyst in the mediastinum there was a congenital defect in the pericardium measuring approximately three centimeters in diameter. This defect was just posterior to the pericardiophrenic vessels and phrenic nerve. Through the defect in the pericardium the main pulmonary artery and the tip of the auricle could be seen. No inflammatory adhesions were present. No other anomalies were noted. The bronchiogenic cyst was excised intact. The defect

in the pericardium was partly closed with the flap of mediastinal pleura which had been dissected from the cyst. The postoperative course was uneventful. There was no accumulation of fluid in the pericardial sac postoperatively, as determined by roent-genograms.

Cut section of the excised cyst revealed a relatively thin-walled sac filled with brownish, thick, mucilaginous material. Several trabeculae were present within the cyst.



FIG. 14.—Case 8: Roentgenogram after surgical removal of the bronchiogenic cyst. Note well expanded left lung.

Microscopic examination revealed a lining of ciliated columnar epithelium. The underlying stroma contained mucous glands, fragments of cartilage, bundles of smooth muscle and islands of lymphoid tissue.

Case 8.—J. J., female, age nine months. This baby was admitted to Kings County Hospital because of dyspnea and bouts of cyanosis since birth and frequent respiratory infections. A diagnosis of unresolved pneumonia of the left upper lobe had been made. Chest roentgenogram, however, revealed a shadow suggesting a large cyst (Fig. 12). A

needle was introduced into this region and thick, gelatinous, yellow material, which was sterile on culture, was aspirated. At a second aspiration of the cyst some air and a small amount of lipiodol were introduced and further roentgen-rays taken. (Fig. 13A and 13B). A diagnosis of bronchiogenic cyst was made.

Operation.—A large, thin-walled cyst which was attached by a small pedicle to the mediastinum near the anterior end of the interlobar fissure close to the phrenic nerve was found. A structure like a small bronchus could be felt in the mediastinal pedicle of this cyst, but this ended blindly and was not in close association with the remainder of the tracheobronchial tree. The cyst was excised and the postoperative course was uneventful. Microscopic examination revealed the characteristic findings of a bronchiogenic cyst. The lung expanded well after operation (Fig. 14).

SUMMARY

Bronchiogenic cysts of the mediastinum result from the faulty development of elements of the primitive foregut. The cyst wall resembles that of the bronchus. The symptomatology depends chiefly on the size and location of the cyst. The various clinical pictures associated with the lesion are discussed in relation to the site of the cyst. Bronchiogenic cysts are a common type of mediastinal tumor. Surgical excision is usually indicated, although a considerable percentage of the patients are asymptomatic when the lesion is first discovered on a roentgenogram. Eight illustrative cases are reported.

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