AIR-CONTAINING CYSTS OF THE LUNG *

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There are many varieties of cysts of the lung which may contain air, but in this paper it is not intended to deal with them all. Only five types, which have some clinical relationship to each other, will be considered. They are (1) bronchogenic cysts, (2) post-infective cysts, (3) infantile tension cysts, (4) anepithelial cysts, and (5) emphysematous cysts.

The cases have been collected from several separate thoracic surgical units, and hence their treatment has been predominantly surgical. This would not have been the case had the source of material been wider.

BRONCHOGENIC AND POST-INFECTIVE CYSTS

PATHOLOGY OF BRONCHOGENIC CYSTS.—Bronchogenic cysts are developmental in origin, they are lined with ciliated epithelium, and the wall may contain smooth muscle, cartilage, mucous glands, or elastic tissue (Fig. 1). The cysts are allied to those which occur in the paratracheal and para-oesophageal regions, and probably arise in much the same way as the enterogenous cysts related to the small gut.

The differentiated elements in the wall are important, as these cysts are liable to become infected; when they do so the epithelial lining may be completely destroyed, but the various constituents of the wall are unlikely to be completely lost. Chronic lung abscesses and post-infective cysts often become lined with ciliated or squamous epithelium when the acute episode is passed (Pryce, 1948), and hence the presence of cartilage, muscle, or glands in the wall of bronchogenic cysts is the only feature by which they may be differentiated with certainty from post-infective cysts.

Bronchogenic cysts may contain air *ab initio*, or they may only develop a bronchial communication after rupture and expectoration of the contents. Unlike anepithelial cysts they never have trabeculae crossing their lumen, although the wall may have a loculated appearance.

PATHOLOGY OF POST-INFECTIVE CYSTS.—When acute lung abscesses have progressed to chronicity they appear as thick-walled, fluid-containing cavities which may eventually become epithelialized. These post-infective cysts may be indistinguishable from bronchogenic cysts, but the walls of the cavities consist of fibrous tissue only (Fig. 2).

Where the original infection has been due to *Staphylococcus aureus* the pathological process is quite different. After the acute pneumonic stage characteristic thin-walled cysts remain. These, unlike chronic lung abscesses, never contain fluid, and the walls may be so thin that they can be mistaken for emphysematous cysts. Also a "check-valve" mechanism is in operation, so that the cysts become distended, and when this occurs in infants the lesion may readily be mistaken for an infantile tension cyst (Fig. 3).

By far the commonest cause of a persistent cystic space after an acute pulmonary infection is staphylococcal pneumonia. In adults this is often a complication of influenza, and before the advent of antibiotics it was frequently fatal (Chickering

FIG. 1.—A section of the wall of a bronchogenic cyst, showing ciliated epithelium, smooth muscle, and mucous glands. $\times 160$.

* This paper comprises the presentations by the authors at the meeting of the Society of Thoracic Surgeons of Great Britain and Ireland in Birmingham, 1952.

FIG. 2.—A section of the wall of a post-infective cyst, showing high columnar epithelium, granulation tissue, and fibrous tissue. $\times 160$.

and Park, 1919). When staphylococcal infection of the lungs occurs in infancy it is more often pyaemic, although the lesions are frequently single. In either age group when the infection is overcome the characteristic cysts remain, although they may eventually disappear after being present for many months.

CLINICAL PRESENTATION.—The clinical features of bronchogenic cysts and persistent post-infective cysts are so similar that the two types may be considered together.

The average age of the 17 patients with bronchogenic cysts was 28 years, nine cysts occurring in men and eight in women. Of the eight cases of post-infective cyst the average age was 23 years, five cysts occurring in men and three in women. Of the total 25 cases, 17 presented with a history of recurrent infection with exacerbation of a productive cough and fever, two with haemoptyses, one with a spontaneous pneumothorax, and two as empyemata, and the remaining three were discovered during routine radiological examinations.

Radiologically these lesions may on occasion appear very similar to emphysematous cysts (Fig. 3a). But the following features make the distinction clear in the majority of cases. There is a visible wall, fluid levels are common, and there is often associated pneumonitis and bronchiectasis. A series of radiographs does not show variation in the size of the cysts. The cysts are never trabeculated, although they may be loculated, they seldom fill during bronchography, and very exceptionally cause displacement of the mediastinum. Many of these features are very different from those of anepithelial and emphysematous cysts (Table II).

TREATMENT.—From a therapeutic point of view the post-infective and bronchogenic cysts may again be considered together, for they are often indistinguishable on clinical and radiological grounds and their course is similar.

When the cyst presents as a thin-walled uninfected air space, resection is not indicated, as the

FIG. 3.—A radiograph of a child of 6 weeks old with two staphylococcal cysts secondary to an umbilical infection. (Dr. E. Hart's case.)

FIG. 3a.—A radiograph of a bronchogenic cyst with a thin wall and containing no fluid. (Mr. T. Holmes Sellors' case.)

cyst is probably lined with epithelium and infection does not appear to be inevitable. Also the staphylococcal variety of cyst may disappear altogether after a variable length of time.

When, however, the cysts are chronically or recurrently infected and causing a cough productive of purulent sputum, they should be resected together with the affected segment of the lung. Similarly, if the cyst is responsible for a chronic spontaneous pneumothorax, it should be excised as pleurodesis is unlikely to be effective.

None of the 26 patients treated by resection in this series died and the late results were excellent.

INFANTILE TENSION CYSTS

Not all congenital cysts are of the "tension" variety. However, if cysts give rise to symptoms in the first year or so of life, it is usually the result of a positive pressure in them and there may be severe respiratory embarrassment due to mediastinal displacement.

Many cysts in infants have been reported, but histological descriptions have often been inadequate. Some consist of a single cystic space lined with respiratory epithelium, others take the form of a localized emphysema, and yet others lie between the two extremes (Fig. 4). The exact aetiology is obscure, but the tension element must arise from a check valve mechanism (Sellors, 1938). The emphysematous type is lobar in distribution and the check valve mechanism must lie in the lobar bronchus. In one such case Gross

FIG. 4.—A section of the wall of an infantile tension cyst showing the cavity lined by low cuboidal epithelium and widespread rupture of the alveolar walls. $\times 45$.

FIGS. 5 and 5a.—A radiograph of an infantile tension cyst in a child of 5 weeks with gross mediastinal displacement and a little fluid in the loculated cyst. (Mr. T. Holmes Sellors' case.)

(1946) has shown a congenital absence of cartilage. Robertson and James (1951), who first stressed the importance of this group under the title "congenital lobar emphysema," believed that the bronchial obstruction in one case resulted from an abnormally placed vein. A redundant fold of mucous membrane has also been cited as responsible for a valvular mechanism in the bronchus (Royes, 1938).

CLINICAL PRESENTATION.—The three patients in this series were 5 weeks, 11 weeks, and 15 months old. All had dyspnoea and cyanosis; in two life was threatened. Radiographs showed enormous air spaces and gross mediastinal displacement (Fig. 5 and 5a). Those described in the literature are very similar. The lesion was almost invariably fatal until the first successful resection reported by Fischer, Tropea, and Bailey in 1943. In none had there been a history of infection. This is an important point in differential diagnosis from staphylococcal cysts, which require different treatment from the infantile tension cysts (Potts and Riker, 1950).

TREATMENT.—Resection is the operation of choice and should be carried out without delay. In some cases it may be essential to decompress the cyst by aspiration or by temporary drainage before an anaesthetic can be given. This technique has been described and successfully used by Walker, Taggart, and Staton (1948) and Burnett and others (1948). The results of resection have been excellent and the mortality low; all three patients in this series were treated by resection and are now free from symptoms.

ANEPITHELIAL AND EMPHYSEMATOUS CYSTS

PATHOLOGY OF ANEPITHELIAL CYSTS.—There is a group of air-containing cysts of lung which is essentially unassociated with emphysema. As the clinical presentation and the treatment of these cysts is different from that of any other variety, a separate word should be used to describe them. Much of the confusion of thought about "emphysematous" cysts may have arisen from the fact that the group of "anepithelial" cysts has usually been described with them, whereas the latter form a sufficiently clear-cut pathological and clinical entity to justify separate description.

The term "anepithelial" has been used as the cysts have no epithelial lining, and the cells which have often been described as "low cuboidal epithelium" lining the cyst were more probably a collection of macrophages (Fig. 6).

Macroscopically these cysts may reach very large proportions and almost fill a hemithorax; characteristically they are only joined to the lung by a narrow pedicle. The walls are thin and translucent, and trabeculae often cross the lumen. These trabeculae are the remnants of bronchi and blood vessels which have withstood the distending force which first ruptured and finally led to the complete disappearance of the alveoli in the affected area.

The aetiology of anepithelial cysts is uncertain; the fact that they are often found at a younger age than the emphysematous cysts suggests a developmental origin; the coincidence of other congenital lesions in the patients suffering from this condition is also suggestive of this aetiology.

It is possible that an abnormality similar to that causing the infantile tension cysts, but further out in the bronchial tree, may produce a check-valve mechanism and so lead to the development of these large tension cysts in the segment of the lung distal to it.

PATHOLOGY OF EMPHYSEMATOUS CYSTS.—Within this group are included the blebs and bullae described by Miller (1927), the "progressive bullous emphysema" of Price and Teplick (1946), "giant cysts," "pneumatocoeles," "cotton candy lung," and a variety of other examples of the same fundamental lesion. The aetiology is similar in them all, but the type and anatomical site vary with duration and the extent of the original lesion.

Microscopically the cysts are precisely similar to the anepithelial cysts; macroscopically the lesions are similar in that they are trabeculated, but they differ in certain essential features. The cysts are either within the pulmonary substance or on the surface but are seldom pedunculated. They are frequently multiple and are associated with emphysematous changes which are visible either in the lobe primarily involved or throughout the lungs.

FIG. 6.—A section of the wall of an anepithelial cyst. The cells partially lining the cyst are macrophages. $\times 160$.

FIG. 7.—A left upper lobe showing gross bullous emphysema, (Mr. D. Barlow's case.)

The fundamental lesion may be the rupture of alveolar walls ; so long as this is confined to groups of alveoli simple emphysema may result, but when an alveolus ruptures into the interstitial planes a destructive process may be set up which sweeps all before it leaving only the remnants of the bronchi and blood vessels (Cooke and Blades, 1952). If this occurs within the lung substance a bullous cyst develops. If it occurs on or near the surface a bleb may be the result. The disease may be confined to one lobe wherein examples of all these processes may be seen—emphysema, blebs, and bullae (Fig. 7). If all the lobes on one side are diseased the term coined by Burke (1937), "vanishing lung," describes the condition. More frequently it is a bilateral phenomenon as in the case described by Allison (1942). The disease is usually slowly progressive, but it may be entirely confined to one lobe.

The aetiology of emphysematous cysts is undecided, but the lesions are almost certainly acquired and degenerative. Cysts may be caused by lesions which lead to partial bronchial occlusion, such as tumours, foreign bodies, or the scarring caused by tuberculous bronchitis. The cysts caused by these conditions are strictly localized ; sarcoidosis, on the other hand, may cause widespread cystic changes which may eventually prove fatal.

The lesion has been attributed to trauma ; infection has also been implicated (Almeyda, 1949), and in some cases the onset does appear to be related to a violent fit of coughing. It is probable that in these patients the pre-existing disease is made apparent by coincidence. Primary lesions of the pulmonary and of the bronchial arteries have also been suggested as the cause of emphysematous

cysts. Korol (1947) drew attention to the frequent co-existence of emphysema and degenerative arterial disease elsewhere. More recently Cudkowicz and Armstrong (1953) have shown very widespread abnormalities of the bronchial arteries in emphysematous patients and it may be that this is in fact the primary lesion.

CLINICAL PRESENTATION OF ANEPITHELIAL CYSTS.—As it is probable that these have a developmental origin it might be expected that they would be discovered in early life, but of the nine cases collected the average age was 32 years, in five men and four women.

TABLE I
PRESENTING SYMPTOMS IN ANEPITHELIAL AND EMPHYSEMATOUS CYSTS

	Anepithelial Cysts	Emphysematous Cysts
Progressive dyspnoea ...	4	14
Spontaneous pneumothorax ...	3	14
Congestive failure ...	—	4
Pain ...	—	1
No symptoms ...	2	6
Total ...	9	39

The commonest presenting symptom (Table I) was increasing dyspnoea due to the mechanical effects of the cyst, but three of these patients had recurrent spontaneous pneumothoraces, two had no symptoms at all, and none had a history of bronchitis. In the few cases where it has been possible to get a series of radiographs over a long period the increase in size had been negligible and it may be that this is the reason why these patients do not develop symptoms until relatively late in life.

CLINICAL PRESENTATION OF EMPHYSEMATOUS CYSTS.—The average age of the 39 patients in this group was higher than in that of the anepithelial cyst group ; it was 40 years: 25 were men and 14 were women. The average age in the women was 35 years, and in the men 45 years, but it must be repeated these patients were derived from surgical centres, and those suffering from the same condition, but of more advanced age, have been excluded, so that the average age of 40 years is far below that of all the patients suffering from bullous emphysema.

The most frequent presenting symptom was dyspnoea, which occurred in 23 of the 39 patients. In 16 the disease was complicated by a spontaneous pneumothorax and there was chest pain in four cases (Table I). Six patients had no symptoms and in only one was there a history of infection. Bronchitis and bronchospasm, which is so

FIG. 8.—Radiograph showing enormous pedunculated cyst arising from left lower lobe.

FIG. 8a.—Radiograph of the same patient after removal of the cyst. (Case of Mr. D. Barlow.)

frequently associated with emphysema, was recorded in only nine of these patients.

The final stage of the disease is the development of cor pulmonale. This occurred in four patients and it is significant that in two of these there was a history of bronchitis. Donald (1953) has stated that congestive failure when it occurs in emphysematous patients is almost invariably precipitated by bronchitis and bronchospasm; it seems probable therefore that this train of events is also responsible for the development of cor pulmonale in patients suffering from emphysematous cysts.

Radiologically the anepithelial and emphysematous cysts can be grouped together. They have a very thin, hairline wall, although an area of compressed lung may appear to form a thicker wall in some cases; in others no boundary at all may be visible on the film (Fig. 8 and 8a). In the latter event the cyst appears as an area of increased translucency. Occasionally it may be difficult to differentiate the cysts from spontaneous pneumothoraces. If the lung edge cannot be seen it may normally be assumed that a cyst rather than a spontaneous pneumothorax is the cause of the increased translucency, but it may be quite impossible to differentiate lobar emphysema from an emphysematous cyst.

Infection of the cysts is rare, so fluid levels and associated pneumonitis are uncommon, but in the case of emphysematous cysts, which may be multiple and bilateral, there is often radiological

TABLE II
COMPARISON OF RADIOLOGICAL FEATURES OF
EPITHELIALIZED AND UNEPITHELIALIZED CYSTS

Characteristics	Bronchogenic and Post-infective Cysts	Anepithelial and Emphysematous Cysts
Wall	Marked around whole circumference	Hairline or none
Fluid levels and pneumonitis	Frequent	Rare
Displacement of mediastinum	Never	Common
Associated emphysema ...	Rare	Frequent
Other	Occasional loculation	Trabeculation

evidence of emphysema elsewhere in the lung. Finally, trabeculation is common (Table II).

TREATMENT

Before any decision can be taken as to the correct treatment in patients suffering from emphysematous cysts it is important to decide the cause of the dyspnoea (Baldwin, Harden, Greene, Cournand and Richards, 1950). It may be due to the mere mechanical interference with respiration, to advanced "alveolar insufficiency," or to a combination of both these elements.

PULMONARY FUNCTION TESTS.—The object of pulmonary function tests is to determine the degree of emphysema in the remaining lung and to estimate to what extent this is the cause of the disability, as mechanical interference with respiration can be dealt with surgically whereas alveolar insufficiency cannot.

In the majority of patients suffering from emphysematous cysts it may be safely assumed that the communications between the cysts and the bronchi are small; so small that they may be disregarded when pulmonary function tests based on gas-mixing techniques are employed. Therefore if it is found that there is evidence of diminished mixing efficiency, a high residual air/total lung capacity ratio and a large "poorly ventilated space," these readings are certain to be due to lung abnormality rather than to the presence of the cysts.

Characteristically these cases have a reduced maximum breathing capacity due to interferences with the mechanics of respiration, and evidence of emphysema of varying degree in the rest of the lung. (Table III shows a typical series of figures.)

TABLE III
RESULTS OF PULMONARY FUNCTION TESTS* ON A PATIENT AGED 48

Total lung capacity ...	...	3.37 litres
Inspiratory capacity ...	...	1.42 "
Functional residual air ...	...	1.95 "
Expiratory reserve ...	...	0.28 "
Residual air ...	...	1.67 "
Residual air : total lung capacity ratio (49.5% : <45%)		
Mixing efficiency ...	24.25% (normal, 80-100%)	
Poorly ventilated space ...	350 ml. (" 0-200 ml.)	
Poorly ventilated space/functional residual air ...	18% (" 0-10)	
Maximum breathing capacity ...	36.4 litres/min. (normal, 70)	

* The residual air/total lung capacity ratio is a little high, the mixing efficiency is grossly impaired, the poorly ventilated space is abnormally high, and the maximum breathing capacity is low.

TREATMENT OF ANEPITHELIAL CYSTS.—When these lesions are causing symptoms resection is the treatment of choice. The cyst can usually be removed from the lung without difficulty. Of the nine cases in this series treated in this way, all have been symptom free for periods up to three years after this operation and none have developed signs of emphysema elsewhere.

When the cysts are unassociated with symptoms an expectant policy may be justified, as they do not necessarily increase in size and infection is rare. The only justification for resection is the possible risk that the presence of the cyst may lead to the development of emphysema, or that if an unrecognized spontaneous pneumothorax develops it may be lethal.

TREATMENT OF EMPHYSEMATOUS CYSTS.—Before a rational approach can be made to the treatment of these cysts it is essential to ascertain whether the disease process is necessarily progressive leading to the involvement of both lungs or whether it may remain confined to one lobe. Also it is important to know whether removal of a grossly diseased area may arrest the progress of the disease in segments left behind. Until

many more of these patients have been subjected to resection and have been followed up for long periods, neither of these questions can be answered with certainty. On the first point, patients have been encountered with gross emphysematous cysts in one lobe only, with no evidence of abnormality in the rest of the lung. Secondly, Abbott, Hopkins, and Guilfoil (1950) believe that the removal of "trigger areas" can prevent the advance of the emphysematous process.

The 28 patients who were subjected to operation have been followed up for varying periods up to five years. The factor which influenced the prognosis decisively was the presence of bronchitis. Of the 21 patients in whom bronchitis was not noted, the majority became symptom free after operation and showed no evidence of advance of the disease, whereas in the seven patients who had a history of bronchitis only one was free from symptoms after operation. Also two of the patients who were considered unfit for operation had bronchitis and have subsequently died from the disease (Tables IV and V).

TABLE IV
RESULTS OF OPERATIONS

Condition	Without Bronchitis	With Bronchitis
Symptom free ...	17	1
Unimproved ...	1	3
Operative death ...	2	1
Late death ...	1	2

TABLE V
INDICATIONS FOR RESECTION IN EMPHYSEMATOUS CYSTS

Indication for Resection	No. of Cases
Progressive dyspnoea ...	13
Spontaneous pneumothorax ...	9
Congestive heart failure ...	3
Pain ...	1
No symptoms ...	2
	28

The indications for resection in this series are shown in Table V. But as it is uncertain whether resection can be actually curative, we suggest the indications for surgery should be (1) increasing dyspnoea, (2) recurrent or chronic spontaneous pneumothorax, and (3) recurrent infections.

To deal with the last two first: infection is uncommon and in no case in this series was it an indication for surgery. Spontaneous pneumothorax whether recurrent or chronic is unlikely to be treatable by pleurodesis and resection is indicated. Increasing dyspnoea is the major indication for surgery in these patients and the type

and feasibility of any operation depend on the cause of the dyspnoea (Baldwin and others, 1950). If it is predominantly caused by mechanical interference with respiration due to the size of the cysts and the positive pressure within them, and particularly where the disease is confined to one lobe, resection of the cysts is the treatment of choice. More frequently lobectomy or even pneumonectomy has to be carried out (Table VI).

TABLE VI OPERATION FOR EMPHYSEMATOUS CYSTS				
Cystectomy	...	...	...	10
Pneumonectomy	...	...	...	3
Lobectomy	...	...	...	15

If the dyspnoea is due to a combination of mechanical interference with respiration and of the loss of function associated with the destruction of alveoli (alveolar insufficiency) considerable symptomatic relief may be obtained if the mechanical cause of the dyspnoea can be treated. Naclerio and Langer (1947) have described a technique wherein most of the cyst wall is removed and the tiny pulmonary communications are carefully oversewn; the loss of functioning lung is thus reduced to a minimum. Alternatively, if the disease is too widespread to justify resection, an attempt may be made to prevent further enlargement of basal cysts by the performance of a phrenic interruption (Allison, 1947) (in the few cases in this series in which this was tried, no improvement followed) or such cysts may be punctured and deflated by means of Monaldi tubes (Head and Avery, 1949).

When the disease has become so far advanced that a cor pulmonale has developed, surgery is unlikely to be helpful, and, of the four patients in this series with this complication, two died at operation, one died without operation, and one was slightly improved. Similarly patients who have well established bronchitis are unsuitable for surgical procedures.

At the other end of the scale, there are many patients with emphysematous cysts of the lungs who are entirely symptom free, and it is doubtful whether they should be operated on unless the disease shows incontrovertible signs of progression.

SUMMARY

Seventy-five cysts of the lung have been collected from thoracic surgical units.

Five varieties have been discussed: bronchogenic cysts, post-infective cysts, infantile tension cysts, anepithelial cysts, and emphysematous cysts.

An attempt has been made to correlate the pathological, clinical and radiological aspects of each type of cyst.

A group of "anepithelial" cysts has been separated from the emphysematous cysts as they are thought to constitute a separate clinical entity.

The treatment of each group is described; the importance of differentiation between mechanical interference with respiration and alveolar insufficiency in emphysematous cysts is discussed.

The relative infrequency of bronchitis complicating emphysematous cysts in this series is noted and the bad prognosis associated with its presence is stressed.

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