

Pulmonary Cyst

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ABSTRACT

Pulmonary cysts occupy an important place among diseases of the chest, and their treatment has to be considered by taking into account the pathologic condition of each disease.

We have reported here the findings obtained through clinical experience of 6 cases of intrapulmonary bronchial cyst, 36 cases of giant bulla, and 127 cases of spontaneous pneumothorax, who were operated on during the past-odd 10 years at this hospital, with regard to the concept, pathologic condition, developmental mechanism, clinical symptoms, diagnosis and treatment of each disease.

Pulmonary cyst is defined as an abnormal space in the lung, and it is observed as a transparent image on roentgenography. From the clinical point of view, pulmonary cyst is broadly classified into alveolar pulmonary cyst and bronchial pulmonary cyst, the former further subdivided into bleb, bulla and giant bulla and the latter into solitary bronchial cyst and multiple bronchial cyst. Although it is placed in the category of benign diseased of the lung, pulmonary cyst often requires surgical treatment. It is important in considering the treatment to study the pathologic condition of each of these diseases. The findings obtained through clinical experience of 6 cases of bronchial cyst, 36 cases of giant bulla and 127 cases of spontaneous pneumothorax operated on at this hospital during the past 10-odd years are reported.

CASES

An outline of the cases is presented in Fig. 1 to 3.

1) Cases operated on for bronchial cyst (6 cases)

The cases were 4 males and 2 females ranging in age from 14 to 35, with an average age of 27 years. the cyst ranged in size from 4×4×4 cm to 9×7×9 cm and was solitary in 5 cases and multi-ocular in 1 case, with affec-

tion of the right lung in 3 cases, the left lung in 3 cases, the lower lobe in 4 cases and the upper lobe in 2 cases. Communication with the bronchus was confirmed in 4 cases and air-fluid-level was presented in 5 cases. One case was air-filled. Four of the 6 cases had had cyst indicated in their early teens and all of those cases were radically cured by operation. the operational procedure employed was lobectomy in 4 cases and cyst excision in 2 cases, with mucosal cauterization additionally performed in one case. Four cases were given this diagnosis histopathologically.

2) Cases operated on for giant bulla (36 cases)

Bulla occupying one third or more of the thoracic cavity were tabulated as giant bulla, and 36 cases-44 lungs were operated on during the past 13 years, comprising 34 males and 2 females, who ranged in age from 14 to 72, with an average age of 47 years, Smoking history was found in 30 cases (83%). As for symptoms, 17 cases had dyspnea and 10 cases were asymptomatic. The site of development of giant bulla was the upper lobe in 34 of the 36 cases (94%). The complications were pneumothorax in 6 cases, intracystic infection in 2 cases, and lung cancer in 2 cases, The operational procedures employed were bullectomy and plication in 37 lungs and lobectomy in 7 lungs.

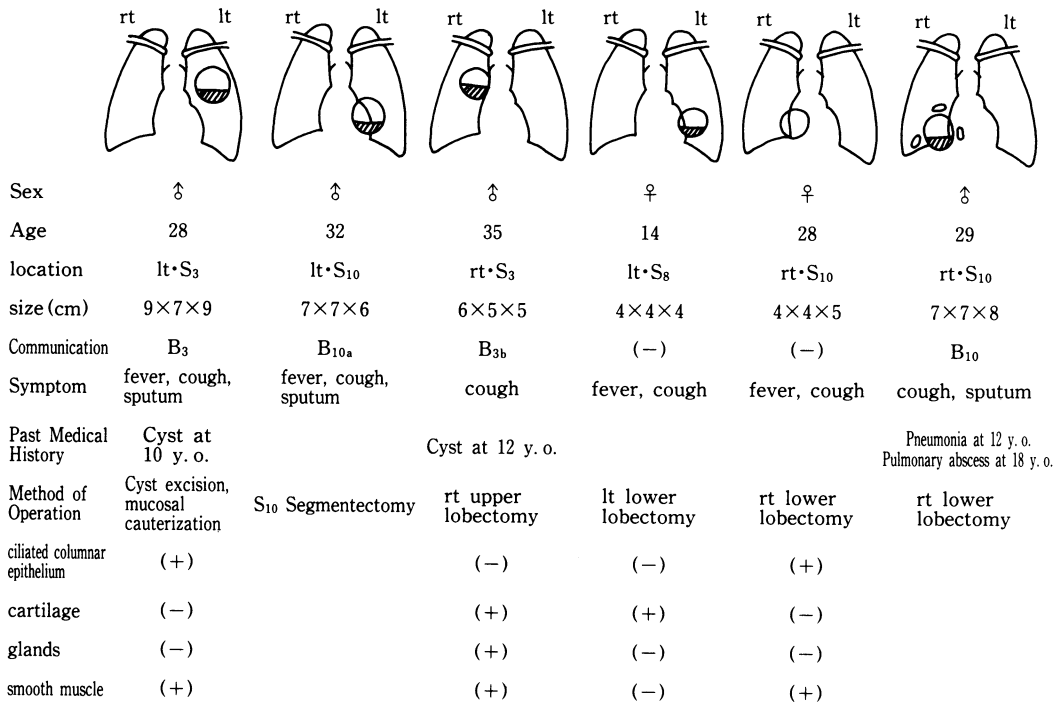


Fig. 1. Intrapulmonary Bronchial Cyst 6 Cases

■ Giant Bulla 36cases
 ♂ 34cases
 ♀ 2cases

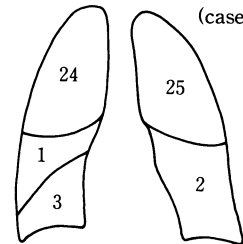
■ Age Distribution (cases)
 ~19 (1)
 20~29 (3)
 30~39 (5)
 40~49 (12)
 50~59 (9)

■ Smoking history (+) 30cases
 (-) 6cases

■ Complications(cases)
 Pneumothorax (6)
 Intracystic infection (2)
 Lung tuberculosis (4)
 Lung cancer (2)

■ Symptoms (cases)
 Asympton (10)
 Dyspnea (17)
 Cough (10)
 Chest pain (6)
 Sputum (3)

■ Location of Giant Bulla at thoracotomy (cases)



■ Method of Operation (44sides)
 Bullectomy & plication 37sides
 Lobectomy 7sides

Fig. 2. Giant Bulla 36 Cases

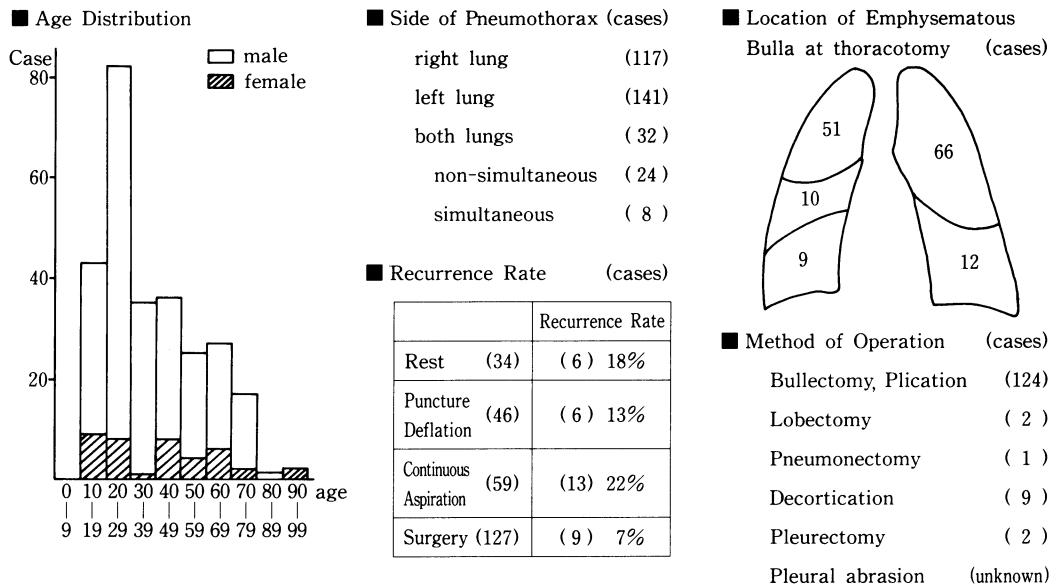


Fig. 3. Spontaneous Pneumothorax 274 Cases
Surgical Operation 127 Cases

3) Cases operated on (127 case) among cases of spontaneous pneumothorax (274 cases)

The frequency of the disease was 232 males and 42 females, at a sex ratio of 5.5 By age, at one peak distribution with the peak in the twenties was presented. By the side in which it developed, it was the right lung in 117 case (44%), left lung in 141 cases (57%), and both lungs in 32 cases (12%), The recurrence rates by rest, puncture deflation, continuous aspiration, and surgical treatment were 18%, 13%, 22%, and 7%, respectively.

At onset of pneumothorax, shadow of cyst was clearly evident on the chest roentgenogram in 34% of the cases, at thoracotomy, almost every case showed emphysematous cyst in the bilateral upper lobes at the apex, The operational procedure employed was bullectomy and plication in almost every case, by which radical cure was effected.

DISCUSSION

Pulmonary cyst has attracted attention ever since it was first reported as a "large bladder of air" by Nickolous Fontanas in 1638. However, opinions of investigators are divided on its definition. Mayer^{4,5)} and Mikami et al take the view

that it is a disease group in which an abnormal space develops in the lung which is not attributable to direct destruction of the lung interstice by inflamation and tumor. Kumagai²⁾ defines it as a gaseous body inconsistent with the regular broncho-alveolar system or a cyst formation in the lung with liquid content or a cystoid structure, numerous other definitions^{3,8,9)} have been reported, and many classifications are found.

Today, pulmonary cyst^{2,3)} is classified from the clinical point of view broadly into bronchial pulmonary cyst¹⁰⁾ and alveolar pulmonary cyst. Bronchial pulmonary cyst is further classified into solitary and multiple, and alveolar pulmonary cyst into bleb, bulla and giant bulla, each of which differs in its pathologic condition and has its clinical characteristics.

During the past 10-odd years, operations were performed at this hospital on 6 cases of bronchial cyst, 36 cases of giant bulla, and 127 cases of spontaneous pneumothorax. a study has been made concerning the concept, developmental mechanism, pathologic condition, clinical symptoms, roentgenological characteristics, diagnosis and treatment of each of these diseases, which will be reported.

1) Concept

Pulmonary cysts are broadly classified into bronchial pulmonary cyst and alveolar pulmonary cyst by the nature of their cystic wall. The two also differ by the site of development, the bronchial cyst occurring mostly in the center of the bronchial system, that is, in the central lung area from the regional bronchus to the terminal bronchus, and the alveolar cyst being characterized by development in the region from the terminal bronchus to the peripheral lung area.

Bronchial cyst¹¹⁾ is a condition where the cystic wall has various levels of normal bronchial components. but it is broadly classified by the site of development into the intrapulmonary bronchial cyst¹⁰⁾ developing in the lung and the mediastinal bronchial cyst¹³⁾ developing in the mediastinum, the former is treated as a pulmonary cystic disease and the latter as a mediastinal tumor. Of concern of this paper, naturally, is intrapulmonary bronchial cyst.

Alveolar pulmonary cyst is subdivided into bleb, bulla and giant bulla. Bleb is interstitial pneumatosis causing dissociation between the elastic plate of the pleura and the marginal elastic plate of the lung. a condition⁶⁾ where the pleura is partially detached from the lung by air. Bulla, on the other hand, is localized pneumatosis beneath the marginal elastic plate of the lung-strictly a change in the lung which grows in units of lobulette and lobule, as regards the mechanism of its development, localized inflammation occurs in the bronchiole and scar tissue forms, whose valve-like action causes the alveolus to expand and become a cyst. Bulla, whose diameter exceeds 1 cm, is a cyst that generally occurs at a deeper region of the lung than bleb. It often protrudes semispherically from the pleura.

Spontaneous pneumothorax has come to be recognized as a cystic disease of the lung because it is frequently found to occur in relation to emphysematous cyst and almost every case is caused by rupture of bleb or small bulla. Bleb and bulla are only to be distinguished by histologic study at the optic microscopy level⁶⁾ and because the two are not infrequently found in the same specimen. it would seem there is little meaning making a strict distinction .

Giant bulla is defined as a large cyst occupying one third or more of a unilateral thoracic cavity. as regards the dissimilarity of biant bul-

la and bulla. one view has it that they are transitive and another that they are altogether different conditions, and there is still no established opinion at present. Considering such facts as that cases of biant bulla are remarkably fewer than cases of spontaneous pneumothorax, that cases of giant bulla are infrequently complicated by pneumothorax, that there is a difference in onset age, that there is a close relationship to smoking, and that giant bulla itself is a risk factor¹²⁾ for the development of lung cancer, might there suggest that bulla and giant bulla are two different conditions?

2) Cause

As regards the cause of bronchial cyst, some take the view that it is congenital and some that it is acquired, but more seem to support the former at present. The view has it that a series of abnormalities occur in the process where the trachea and the esophagus, which originate in the foregut embryologically, separate, and an undifferentiated piece of tissue strays in, grows and become a cyst. Alveolar pulmonary cyst, on the other hand, is regarded as a change occurring in the postnatal growth process of the lung and is thought to be acquired. The view is that a localized inflammation occurs in the bronchiole which develops a scar tissue that acts like a valve and a large cyst develops by the check valve mechanism.

The 6 cases we experienced had developed at age 14 to 32, at age 27 on the average, and the time of detection of the cyst was in the early teens in 4 of the 6 cases, at ages 10, 12, 14, and 12. This may perhaps be a finding suggesting that bronchial cyst is congenital.

The 36 cases of giant bulla occurred at age 14 to 72, at age 47 on the average. Those aged 40 or over accounted for three fourth of all the cases, and 30 of the 36 cases (83%) had smoking history. This disease was often found by chance roentgenologically in healthy asymptomatic individuals and it was sometimes found to have developed in lung that was normal before. These findings suggest that giant bulla is acquired. a review of the 274 cases of spontaneous pneumothorax¹⁵⁾ showed that the incidence of the disease peaked in the twenties. Also in 10 cases comprising 8 families, this was found to have occurred within the family, and this gives us to feel that hereditary factor is not to be

negated as the causative factor of spontaneous pneumothorax.

3) Pathologic condition and clinical symptoms.

Considering the pathologic condition of each of these diseases, giant bulla is a disorder comprising obstructive ventilation disturbance due to emphysematous change and compression of the adjoining lung due to a space occupying lesion. Spontaneous pneumothorax is collapse of the lung associated with rupture of emphysematous cysts. Intrapulmonary bronchial cyst may be said to be intrapulmonary infection due to intracystic infection and cyst-bronchial fistula.

Accordingly, the clinical symptoms of each differ. Giant bulla often takes an asymptomatic course and is not infrequently found by chance by X-ray. When the bulla becomes large, symptoms, such as dyspnea, chest pain and cough are found, which are due to decreased respiratory surface and compression of the adjoining lung. However, the size of the cyst and the symptoms are not necessarily proportional. and it is said that giant bulla is rarely complicated by intracystic infection and pneumothorax, all of the 36 cases experienced by the authors were operated on, and among the many cases that were clinically advanced, 10 were asymptomatic cases, six cases were complicated by pneumothorax and 2 by intracystic infection. attention should be paid here to the fact that 2 cases of giant bulla were found to be complicated by lung cancer. It is said today that giant bulla is complicated by lung cancer¹²⁾ and also that presence of giant bulla itself is a factor to cause lung cancer. While giant bulla is placed in the category of benign disease, closer observation is believed to be necessary in regard to development of early occult carcinoma.

Bleb and small bulla are in almost all cases asymptomatic, and they evidently give rise to no problem clinically so long as spontaneous pneumothorax does not occur. Also, because these emphysematous cysts tend to occur frequently and the lesion often extends over both lungs, there is a high possibility of developing bilateral pneumothorax. Further, these fragile cysts are frequently found to recur by conservative treatment, which often makes thoracotomy unavoidable. By the cases we experienced, bilateral spontaneous pneumothorax was found in 12% of all cases. Recurrence rate was 18% by conser-

vative treatment and 7% by surgical treatment. On the other hand, intrapulmonary bronchial cyst, differing from mediastinal bronchial cyst¹³⁾, often showed the symptoms of infection prominently. whether it be solitary or multiple, there is at first no communication with the bronchus, often the symptoms are presented when the liquid filled cyst ruptures into the bronchus and communication with the bronchus develops. Mizote et al⁷⁾ tabulated 107 cases and reported that 85% of their intrapulmonary bronchial cyst cases presented the infection symptoms of mild fever, cough and sputum. Six of our cases had cough, sputum and fever, and 5 of them showed air-fluid-level. Communication with the bronchus was confirmed in 4 cases.

4) Roentgenologic characteristics

As regards the roentgenologic characteristics of bronchial cyst, they were generally observed to be well defined unilocular cyst, smooth of surface and round or oval in shape. They are reported to occur frequently in one lung lobe, especially in the lower lobe. Roger et al¹¹⁾ reported occurrence in the lower lobe in 21 among 32 cases (66%). They also classified the cysts roentgenologically into 3 types: 1) homogeneous water density, 2) air-filled, and 3) air-fluid-level, and reported that as many as 20 of the 32 cases (63%) presented air-fluid-level. In our cases, 4 of the 6 cases showed the cyst in the lower lobe, and 5 of them were unilocular cyst, and 5 cases presented air-fluid-level.

Giant bulla, which has a hair-like thin wall, is observed as a convex, well-defined avascular area in the hilum of the lung, Observed as simple or multiple, it occurs frequently in the upper lobe, In our cases, the lesion was found in the upper lobe in 34 of the 36 cases (95%), extending over both lungs in 18 of the 36 cases (50%).

Small bulla and bleb cannot readily be detected roentgenographically when spontaneous pneumothorax develops. a definite shadow of the cyst is seen only 90 of 211 cases (34%) roentgenographically. This is believed to be attributable to the fact that the emphysematous cyst, the cause of pneumothorax in almost all of the cases existed in the upper lobe, especially at the apex of the lung, and the cyst moreover was small.

5) Diagnosis and treatment.

It goes without saying that clinical symptoms,

past medical history and roentgenographic findings are important in making the diagnosis, Bronchial cyst, whose definitive diagnosis is based on histopathological study of the cyst wall, generally occurs frequently in young people under 30 years of age with a history of pneumonia and bronchitis in childhood, and the chief complaints of this disease are in most cases cough, sputum and fever. Roentgenographically, it is frequently observed as a unilocular spherical cyst associated with air-fluid-level as previously mentioned. reportedly, bronchial cyst is often associated with infection, with the chance of infection being great where there is communication with the bronchus, and sometimes the same symptoms as pulmonary abscess are presented.

Because an infected cyst reportedly cannot be expected to be cured by conservative treatment and operation becomes difficult once infection occurs, surgical pulmonectomy is believed to be the best method of treatment, Lobectomy was performed on 4 and cyst excision on 2 of our cases, in one of which the residual mucosa was cauterized with an electrome.

Rarely the cyst in the lower lobe is nourished from the aorta, in which case the disease is regarded as intrapulmonary sequestration¹⁾. Attention should be paid especially to abnormal blood vessels during the operation.

Diagnosis of spontaneous pneumothorax is made by chest roentgenography, this disease occurs frequently in young males, and chest pain, dyspnea and cough are frequently observed as the incipient symptoms. the principle of treatment is reinflation of the lung and prevention of recurrence. when reinflation is not gained by conservative treatment or when the disease recurs, surgical thoracotomy should be positively performed. Further, for cases of bilateral simultaneous pneumothorax and cases of bilateral non-simultaneous pneumothorax, bilateral simultaneous thoracotomy should be performed without hesitation. Among the 127 cases we experienced who underwent operation, bilateral simultaneous thoracotomy¹⁴⁾ was performed on 8 cases. (6.3%).

Diagnosis of giant bulla is made by chest roentgenography. As a roentgenological characteristic, it is observed as an avascular area surrounded by a hair-like shadow as mentioned

earlier. Although differentiation from complete pneumothorax is difficult when the cyst is very large. pneumothorax shows a shadow of the collapsed lung with a round contour centering the hilum of the lung, while the cyst presents a long narrow shadow of the hilum. A bulla occupying one third or more of a unilateral thoracic cavity often compresses the adjoining lung and presents various symptoms, but there are many case also where the cyst is asymptomatic and is detected by chance in an examination. A giant bulla rarely presents complication by pneumothorax or intracystic infection, but operation should be performed positively for cases with clinical symptoms, cases with a tendency to grow, and cases complicated by pneumothorax or infection or lung cancer. Fundamentally, bullectomy and plication are performed, but lobectomy is also performed according to the case.

REFERENCES

1. **Culiner, M.M.** 1968, Intralobar bronchial cystic disease. "the sequestration complex" and cystic bronchiectasis. *Diseases of the Chest* **53**: 462-469.
2. **Kumagai, N.** 1961. Pulmonary cyst. (Japanese). *Touhoku Med. J.* **63**: 247-268.
3. **Maier, H.C.** 1941. Pulmonary cysts. *Am.J.Surg.* **54**: 68-81.
4. **Mayer, E. and Rappaport, I.** 1952. Developmental origin of cystic bronchiectatic and emphysematous changes in the lungs. *Disease of Chest* **21**: 146-160.
5. **Mayer, E. and Rappaport, I.** 1953. Clinical observations and interpretations of abnormal air spaces in the lungs. A new concept of their origin. *J.A.M.A.* **153**: 700-703.
6. **Miller, W.S.** 1926. A study of the human Pleura Pulmonalis: Its relation to the blebs and bullae of emphysema. *Am.J. Roentgenol and Rad. Therapy* **15**: 399-407.
7. **Mizote, H. et al** 1973. Bronchogenic cyst. (Japanese). *Surg. Treatment* **29**: 131-142.
8. **Moersch, H.J. and Clagett, O.T.** 1947. Pulmonary cysts. *J. Thoracic, Surg.* **16**: 179-199.
9. **Naclerio, E. and Langer, L.** 1947. Pulmonary cysts. *Surgery* **22**: 516-524.
10. **Ofoegbu, R.O.** 1982. Intraparenchymal bronchogenic cysts in adult. *Thorac. cardiovasc. Surgeon.* **30**: 298-310.
11. **Rogers, L.F. and Osmer, J.C.** 1964. Bronchogenic cysts. A review of 46 cases. *Am.J. Roentgenol.* **91**: 273-283.
12. **Stoloff, I.L., Kanofsky, P. and Magilner, L.** 1971. The risk of lung cancer in males with bul-

- lous disease of the lung. Arch. Environ. Health. 22: 163-167.
13. **Sumimoto, R., Takahashi, T., Hayashi, K. and Kamata, T.** 1985. Mediastinal Bronchial cyst. a review of four cases. Med. J. Hiroshima Univ. 33: 189-194.
14. **Sumimoto, R., Takahashi, T. and Hayashi, K.** 1985. A review of bilateral thoracotomies. Med. J. Hiroshima Univ. 33: 407-414.
15. **Sumimoto, R., Takahashi, T., Yoshioka, S. and Hayashi, K.** 1985. A study of 274 spontaneous pneumothorax. Med. J. Hiroshima Univ. 33: 883-890.