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Bronchogenic cysts: a review of 20 cases

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Abstract *Objective.* All cases of bronchogenic cysts treated in our center are analysed in order to define its clinical and pathological features. These data are used to determine whether surgical treatment in all cases is justified or whether there is a place for conservative treatment.

Methods. We retrospectively studied the medical records and pathology reports of all patients with bronchogenic cysts ($n=20$) referred to our clinic between 1975 and 1993.

Results. Fourteen patients (70%) were asymptomatic. Six patients had symptoms because of cyst-related complications (infection or compression). In only 15 patients the diagnosis was established preoperatively. In the other five cases a solid tumour

was suspected. All patients were treated by either thoracotomy ($n=19$) or thoracoscopy ($n=1$). The diagnosis was confirmed by histological examination. One cyst turned out to be degenerated into a squamous cell carcinoma.

Conclusions. We conclude that both the risk of developing cyst-related complications and the even smaller risk of malignant degeneration justify surgical treatment in all cases. [Eur J Cardio-thorac Surg (1996) 10:393–396]

Key words Bronchogenic cyst · Clinical · Pathology · Computed tomography · Surgery · Malignant degeneration

Introduction

Bronchogenic cysts can be found about everywhere in the thoracic cavity but most commonly develop along the tracheobronchial tree. They are believed to be congenital and to result from supernumerary or abnormal branching of the tracheobronchial tree during development. In adults most of these cysts are asymptomatic. They are found on routine chest radiographies. Only complications produce symptoms: infection of either the cyst or the surrounding lung tissue, hemorrhage or compression of adjacent organs (esophagus, bronchus or heart).

The aim of this study is to record the clinical and histological features of all bronchogenic cysts, surgically treated at our institution. These data are used to determine whether surgical treatment in all cases is justified.

Patients and methods

Over an 18-year period (1975–1993) 20 patients were operated on for bronchogenic cysts. The medical records and pathology reports of all patients were thoroughly reviewed. There were 11 female, and 9 male, patients. The mean age was 35.6 years (range 19–70). Age distribution is shown in Fig. 1. All patients had a chest radiography. Additional investigations consisted of a computed tomography (CT) scan ($n=17$), bronchoscopy ($n=13$), barium contrast study ($n=4$), standard tomography ($n=2$) and echocardiography ($n=1$). Nineteen patients underwent resection of the cyst by either left ($n=7$) or right ($n=12$) posterolateral thoracotomy. Recently one mediastinal cyst was treated by thoracoscopy. All mediastinal cysts could be completely resected, including two paraesophageal cysts with extension into the esophageal wall, requiring local esophageal myotomy. One hilar cyst, located between the upper lobe bronchus and the superior pulmonary vein, was treated by pneumonectomy because frozen section showed malignant degeneration. The intrapulmonary cysts were removed by either lobectomy ($n=2$), segmentectomy ($n=1$) or wedge excision ($n=3$). The postoperative period was uneventful in all patients. The mean hospital stay was 14.6 days (range 11–13).

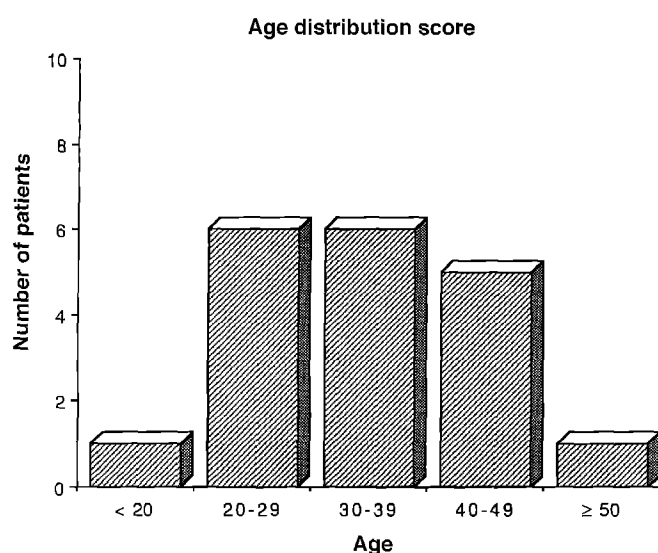


Fig. 1 Age distribution score of patients with resected bronchogenic cysts

Results

Symptoms

The majority of patients (70%) were asymptomatic. Infection was the most common symptom among the patients who had symptoms (Table 1). In the group of patients with pneumonia ($n=3$) there were two mediastinal cysts, one of which communicated with the respiratory tree; a lung abscess, on the other hand, was caused by an intrapulmonary cyst, also communicating with the tracheobronchial tree. Dysphagia and persistent coughing were both caused by a mediastinal cyst.

Location and diagnosis

There were 14 mediastinal and 6 intrapulmonary cysts. The mediastinal cysts are classified according to Maier [7] into five groups: paratracheal ($n=0$), carinal ($n=7$), hilar

($n=1$), paraesophageal ($n=3$) and miscellaneous (3 pre-vertebral). The intrapulmonary cysts ($n=6$) were more frequent on the right side ($n=5$) than on the left ($n=1$). Three cysts in this group communicated with the tracheobronchial tree.

In 15 patients (75%) the diagnosis of a bronchogenic cyst was established preoperatively. The presence of a cyst was suspected on standard chest radiography when an air-fluid level was seen ($n=2$). Computed tomography revealed the correct diagnosis in 12 cases (71%). The other three cysts were diagnosed preoperatively by either echocardiography ($n=1$) or standard tomography ($n=2$). Barium swallow was performed in four patients, showing extrinsic compression of the esophagus in one patient with dysphagia. Extrinsic bronchial compression was seen in 3 out of 13 bronchoscopies.

Histology

The pathology reports of all patients operated on were reviewed. The mean cyst diameter was 4.4 cm (range 2.0–12.0). A bronchogenic cyst was diagnosed when the cyst wall was lined by a pseudostratified, ciliated, columnar epithelium (18 cases) and in the presence of at least one of the elements of a normal bronchial wall: bronchial glands ($n=6$), smooth muscle ($n=9$) or nests of cartilage ($n=10$). An example is shown in Fig. 2. Focal squamous metaplasia was seen in two cysts.

Except for two cases, all cysts could easily be recognised as being bronchogenic cysts. One cyst, located near the right upper lobe bronchus, was only lined by a fibrous wall without visible epithelium. The other one was a 3.5 cm mediastinal (hilar), unilocular cyst (Fig. 3) containing the typical yellow-grey gelatinous fluid of a bronchogenic cyst. Histological examination revealed that its wall was invaded by a squamous cell carcinoma with focal areas of carcinoma in situ (Fig. 4). No normal epithelium or any other bronchial elements were recognised. Examination of the pneumonectomy specimen, on the other hand, showed no evidence of bronchogenic carcinoma.

Table 1 Symptomatic patients

Symptoms	No. patients
Infection	
– pneumonia	3
– abscess	1
Compression	
– dysphagia	1
– coughing	1

Discussion

Since Meyer [9] described a case of bronchogenic cyst in 1859, many reports have appeared in the surgical literature. Bronchogenic cysts account for 40–50% of all congenital mediastinal cysts. They are slightly more common in men than in women (1.5 : 1) and are mainly discovered in the 3rd or 4th decade of life [8]. The majority of our patients (18/20) was between 20 and 50 years old (Fig. 1). Unlike early reports [8, 17], the recent literature suggests that the majority of adult patients, ranging from 50 to 72%



Fig. 2 Wall of a regular bronchogenic cyst with respiratory epithelium, cartilage plate and seromucous glands (HE. $\times 87.5$)



Fig. 3 CAT-scan showing cyst in close relationship with the right pulmonary artery

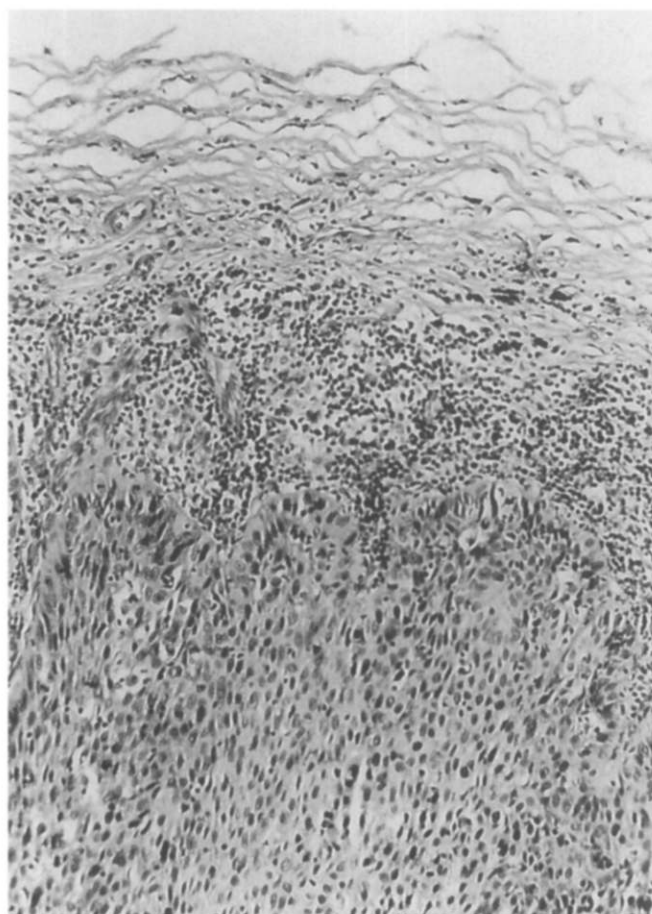


Fig. 4 Cyst wall with areas of invasive squamous cell carcinoma (HE. $\times 350$)

[14–16], is symptomatic. In our own series, which only comprises adults, only 30% of the patients presented with symptoms (Table 1). This contrasts with the clinical picture in early infancy, where bronchogenic cysts can cause severe respiratory distress. A communication between the cyst and the respiratory tree can act as a check-valve mechanism producing a rapid increase in volume and thus compressing the mediastinum [5, 8].

It is interesting that in 5 out of 17 CTs performed, a solid intrathoracic tumor was diagnosed. The apparent confusion is the result of the often high CT density of the cyst fluid, which can be misleading in differentiating a bronchogenic cyst from a solid tumor. High CT density is attributed to an increased calcium content of the cyst fluid [11].

Once a bronchogenic cyst has been diagnosed, various kinds of treatment are available. Swanson's technique of aspirating the cyst fluid, described in 1928, still inspires some surgeons [3, 6, 12]; although reports about recurrent bronchogenic cysts are scarce [13, 16], recurrence can be expected because the cyst wall is not removed. In selected cases, when the cyst is located in the paratracheal region, removal by mediastinoscopy, as reported by Ginsberg in 1972 [4], seems an elegant method of treatment. Until recently thoracotomy was the standard surgical approach; most of our patients (19 out of 20) were treated this way. Recently a mediastinal cyst, located in the paravertebral region, has been removed by thoracoscopy. We think thoracoscopy is a valid alternative for this apparent benign disorder.

It is important, however, to emphasize that malignant degeneration may occur, as was the case in a 70-year-old patient in this series, whose CT scan and photomicrograph of the histology are illustrated in Figs. 3 and 4, respectively. Carcinoma developing within a bronchogenic cyst was reported for the first time by Moersch and Clagett [10] in 1947. Bauer described a squamous cell carcinoma within a bronchogenic cyst in 1961 [1], while Bernheim reported a leiomyosarcoma arising from a cyst wall [2]. Suen and

colleagues described an adenocarcinoma in the wall of a bronchogenic cyst in an 8-year-old girl [16].

We think that the minimal risk of malignant degeneration and the much greater risk of cyst-related complica-

tions justify operative treatment in all cases. Thoracotomy or, in experienced hands, thoracoscopy seem the safest methods of achieving this.

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