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Bronchogenic cysts

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Summary

The medical records of all patients with bronchogenic cysts (during the period from 1967 to 1987) were reviewed. Our series consists of 26 patients who underwent surgery for excision of bronchogenic cyst.

The average age was 6 years 4 months. Among them, only 4 patients were one year old or younger at the time of surgery. 27% of the patients presented with asymptomatic lesions.The remaining 73% all presented with respiratory symptoms.

Surgery in these patients consisted of excision of the lesion for mediastinal cysts and segmentectomy or lobectomy for intraparenchymal ones. Postoperative complications included 1 pneumonia which responded to antibiotics and 1 transient paresis of the right phrenic nerve. Martality rate was zero percent. The mean follow-up period was 17 months (range: 1 month-17 years).

Introduction

Bronchogenic cysts are congenital bening masses commonly located in the mediastinium or lung parenchyma arising from anomalous budding of the primitive tracheo-bronchial tube [1], which can occur at any stage of airway development. Consequently, cysts can be mediastinal or subcarinal in location when abnormal budding occurs at the level of the carina or first order bronchi. Abnormal budding in the distal tracheobronchial tree results in intraparenchymal bronchogenic cysts. Cysts can also migrate to subpleural, pericardial, paravertebral and cervical locations if embryological connections with their parent bronchus are lost [2]. Bronchogenic cysts contain mucoid material and are surrounded by a wall containing bronchial cartilage, smooth muscle, elastic tissue and mucus glands and are lined with ciliated columnar or cuboidal epithelium [3].

Bronchogenic cysts are of clinical significance in that the symptomatology in children varies according to age at presentation. In infants, dyspnea and/or cyanosis or, more rarely, dysphagia are seen, whereas in older children pulmonary infections are the more usual mode of presentation[4]. Less frequently, they present as asymptomatic lesions. We review in this paper our experience with bronchogenic cysts during the past 20 years.

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Material and methods

The medical records of all patients who underwent surgical excision of histologically confirmed bronchogenic cysts in our institution during the period from 1967 to 1987 were reviewed. Data were collected with regard to sex, age, mode of presentation, duration of symptoms, type of investigations performed, location of the mass, preoperative diagnosis, surgical findings, per and postoperative complications, histology of the lesion and follow-up.

Results

Our series consists of 26 patients who underwent surgery for excision of bronchogenic cyst. There were 15 females and 11 males ranging in age from 2 months to 14 years. The average age was 6 years 4 months. Among them, only 4 patients (15-%) were one year old or younger at the time of surgery.

Table 1. Comparison of asymptomatic and symptomatic groups

	No (%)	Average % Age	Mediastinal %	Intraparenchymal %	Others
Asyptomatic	7/26 (27)	8.5 yrs	70	30	0
Syptomatic	19/26 (73)	8.6 yrs	65	26	11

Seven patients (27%), presented with asymptomatic lesions discovered on routine chest films done for other non-related conditions. Their ages ranged from 3 to 12 years (average 8.5 years). There were 4 females and 3 males. Seventy percent (5/7) of patients had cysts located in the mediastinum. The remaining patients had intraparenchymal lesions. There were 4 right-sided and 3 left-sided lesions (table 1).

The remaining 19 patients (73%), all presented with respiratory symptoms. One 2 month-old in this group presented with cough and dysphagia. The interval between the onset of symptoms and diagnosis ranged from no delay to 9 years (average 12.2 months). In 47% (9/19), the respiratory symptoms were infectious, and in 55.5% (5/9) of these patients the lesions were intraparenchymal. All patients aged 1 year (4/26) or less in our series, were symptomatic and presented with dyspnea and/or cyanotic spells. The average age in this group was 6 months (range 2 to 12 months). other symptoms in the older age group included chest pain, fever, upper respiratory tract infections, cough and pneumonia, the most common of these being cough in 47% (9/19) (table 2). Of note is the fact that in this series only one patient presented with reccurent bouts of pneumonia secondary to an intraparenchymal cyst. In this symptomatic group, 63% (12/19) had mediastinal lesions; 26% (5/19) had intraparenchymal lesions; and 11% (2/19) were in the inferior pulmonary ligament (table 1).

Table 2. Mode of presentation of patients with bronchogenic CYST

Asymptomatic	7 (27 %)
Symptomatic	19 (73 %)
. cough	9
. dyspnea	6
. pain	5
. fever	5
. pneumonia	3
. uri	2
. dysphagia	1

On the whole, 65% of patients has mediastinal cysts, usually in close proximity to the trachea, carina, main stem bronchi, esophagus or pericardium. Twenty-seven percent of the total (7/26), were intraparenchymal and 8% (2/26) were located in the inferior pulmonary ligament. Sixty-two percent (16/26) cysts were right-sided.

All patients in the series underwent chect films and later, once the study became available, CT scan of the thorax. Chest films were diagnostic for bronchogenic cyst in 20/26 (77%) patients. Air-fluid levels were detected in 3 patients and were diagnostic for intraparenchymal bronchogenic syst. In 2 patients the lesion was obscured by pneumonia and diagnosed 3 and 6 months later; in one patient, only after repeated bouts of pneumonia. The diagnosis of bronchogenic cyst was missed on initial chest radiograph in the remaining 4 patients and was only diagnosed between 33 months and 9 years later. In all 4 patients the lesions were mediastinal.

Barium esophagogram and bronchoscopy were done in 6 and 4 patients respectively. All but one bronchoscopy were negative, and in only 2 cases did barium swallow show extrinsic esophageal compression. Prior to the availability of CT-scan, some patients (5/26) had chest tomograms done, but in all cases, this study was complementary to positive chest X-ray findings. Once CT-scan became available, it was diagnostic in all cases (6/6).

Surgery in these patients consisted of excision of the lesion for mediastinal cysts and segmentectomy or lobectomy for intraparenchymal ones. All lesions were approached via thoracotomy.

Among the cysts which were adherent to the trachea, 50% (2/4) required repair of the membranous posterior tracheal wall by simple suture after excision. Only two lesions presented with macroscopically visible airway communication not detected pre-operatively. Both were adherent to the right mainstem bronchus. Simple suture of the bronchial defect was sufficient once the lesion was excised. Despite the fact that peroperative rupture of the cyst occured in four cases, all specimens were histologically complete. A correct preoperative diagnosis of bronchogenic cyst was made in 19 patients (73%). Postoperative complications included 1 pneumonia which responded to antibiotics and 1 transient paresis of the right phrenic nerve.

Six patients were lost to follow-up. The remaining 20 patients were followed for an average of 17 months, ranging from 1 month to 17 years. No sequelae were noted and all patients had normal control chest films.

Discussion

Bronchogenic cysts were first described in 1948 by Maier [5]. He noted that these cysts could be situated in the mediastinum or any-where in the pulmonary parenchyma. He further described the embryological, clinical and pathological characteristics of these congenital lesions. It has been noted that these cysts tend to be asymptomatic in older children and adults [6]. In our series, the average age for both symptomatic and asymptomatic patients was essentially the same (8.6 years vs 8.5 years respectively). We did note two different types of patients, in the symptomatic group, in agreement with previously mentioned author. Infants aged 12 months or less presented with respiratory distress (4/19). Older children presented mostly with respiratory infections (9/19).

No patient in our series presented with congenital lobar emphysema due to extraluminal airway obstruction by a bronchogenic cyst. This finding has repeatedly been described [3,7,8,9]. On must there fore consider the possibility of an unrecognized bronchogenic cyst whenever congenital lobar emphysema is diagnosed [10]. Hypoplasia of either pulmonary artery[11], cardiac arrhythmias [12], superior vena cava syndrome[13], pericardial compression[14], and bronchial atresia [15] have all been described secondary to bronchogenic cysts in our series. Sirivella et al. [16] recently described non-productive cough as the most frequent symptom in his review of 20 cases of bronchogenic cyst. Our series corroborates this finding.

No difference in location of bronchogenic cysts between the symptomatic and the asymptomatic group was found. The majority of the lesions were mediastinal in both groups. Contrary to what has been published concerning the predominantly intraparenchymal location of bronchogenic cysts [2,17,18], our series demonstrated a predominantly mediastinal location (65%). Other atypical locations, such as cervical[19,20] and subcutaneous [21,22], were not encountered. Our series also failed to reveal a previously reported consistent relationship between perihilar or subcarinal location of bronchogenic cysts and the presence of related symptoms[23] in that 2 of our 7 asymptomatic patients had such lesions.

As previously mentioned, and contrary to other authors, barium swallow and bronchoscopy were not uniformly useful diagnostic aids[2,24]. Noticeably, bronchoscopy failed to demonstrate a comunication between the right main stem bronchus and the bronchogenic cyst which was evident at the time of surgery. Other diagnostic modalities available today such as magnetic resonance imaging are not as useful in evaluating mediastinal and hilar lung masses as CT scan [25]. We conclude that chest X-ray and CT scan are the two most valuable diagnostic studies (table 3).

In 1943 Adams and Thornton [26] first recommended partial excision and mucosal scarification of mediastinal bronchogenic cysts. Since then,

Table 3. Value of different diagnostic studies

Diagnostic Study	% Positive
Chest X-ray	20/26 (77 %)
Barium swallow	2/6 (33 %)
Bronchoscopy	1/4 (25 %)
CT scan	6/6 (100 %)

other authors have advocated a similar approach [27]. Arguements in favour of a more agressive approach, with total excision of these lesions, include reported cases of malignant degeneration [28,29] and symptomatic recurrence [30]. All cases in our series were approached by thoracotomy. Other authors report excellent exposure via median sternotomy for cysts located anterior to the pulmonary hilum[30,31,32]. They recommend that if posterior hilar exposure becomes necessary, a double-lumen Carlens endotracheal tube can be used to deflate the respective lung. If simultaneous bilateral pulmonary deflation should be necessary to carry out total excision of the cyst, cardio-pulmonary bypass can be used[30]. We did not encounter any problems with regards to accessibility of the lesion using thoracotomy. Complete excision was possible in all cases and all patients were cured.

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