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Original Article

BRONCHOGENIC CYST

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Abstract

Bronchogenic cysts are congenital anomalies caused by abnormal bronchial development from the primitive ventral foregut, which arises from cells isolated from the main pulmonary branching when lung bud separates from the primitive gut. We reviewed all pediatric patients with bronchogenic cyst who underwent surgery in our thoracic surgical unit- III during 2007-2009. They comprised 1 male and 5 female patients, with an age range of 2 to 17 years (mean age 5.66 years). Symptoms were present in 5 patients (83%), cough was the most common symptom. Other symptoms included purulent sputum, chest pain, fever, dyspnea, anorexia and/or weight loss, and haemoptysis. All patients underwent chest x-ray postero-anterior and lateral view and CT scan of chest for diagnostic purpose. Two of the cysts were mediastinal and rests were intraparenchymal. Surgery performed were resection (2), lobectomy (4). There was no operative deaths & no in-hospital mortality.

Keywords: Bronchogenic cyst, Resection, Lobectomy

Introduction

Bronchogenic cysts are congenital anomalies caused by abnormal bronchial development from the primitive ventral foregut, which arises from cells isolated from the main pulmonary branching when lung bud separates from the primitive gut. Bronchogenic cysts are commonly located in the mediastinum or lung parenchyma. The location of the cyst depends on the embryological stage of development at which the anomaly occurs. When this abnormal budding occurs during early development, the cyst tends to be located along the tracheobronchial tree. Cysts that arise later are more peripheral and may be located within the lung parenchyma[1,2]. They are generally asymptomatic, unless they attain a large size and cause compressive symptoms, which is often an incidental radiological finding[3,4]. They are particularly significant because of the difficulty in making a differential diagnosis, as they can simulate multiple lesions, both benign and malignant. It is important to recognize these conditions so that appropriate treatment (usually surgical) can be instituted, thereby avoiding unnecessary treatments. With the improved safety of pediatric anesthesia and the development of non-invasive diagnostic procedures, thoracic surgery for symptomatic cystic lesions of the lung has advanced as well as the awareness of these anomalies among pediatricians[5]. The objectives of this retrospective study were to review the clinical presentations and treatment of bronchogenic cysts. We conclude that surgical resection of all suspected bronchogenic cysts in operable candidates.

Patients & Methods

All pediatric patients with bronchogenic cyst who underwent surgery in our thoracic surgical unit- III during 2007-2009 were reviewed.

Patients' records were evaluated with regard to age, sex, clinical picture, diagnostic interventions,

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associated diseases, treatment methods, pathological findings and outcome.

They comprised 1 male and 5 female patients, with an age range of 2 to 17 years (mean age 5.66 years). The initial diagnostic evaluation had included plain radiography, computed tomography and bronchoscopy.

All the patients underwent surgery in our thoracic surgery unit were reviewed in outpatients at 1 month interval for consecutive 3 months, 6 months and 1 year. After that, the patients were advised to contact our follow-up clinic whenever needed.

Results

Clinical presentations

One patient was asymptomatic at initial presentation. She was 17 yrs. The cyst was found incidentally during pre-employment health check-up.

Symptoms were present in 5 patients (83%). Cough was the most common symptom, and it occurred in 4 patients (67%). These patients had symptom for more than one year before surgery. The majority had more than one symptom. Other symptoms included purulent sputum, chest pain, fever, dyspnea, anorexia and/or weight loss, and haemoptysis. These are listed in Table I and summarized in Table II.

Table I

Clinical presentation of the patients

Pt No.	Age	Sex	Symptoms/Signs
1	17 yr	F	Asymptomatic
2	06 yr	F	Fever, Cough, Purulent Sputum, Dyspnea
3	02 yr	M	Cough, Dyspnea
4	06 yr	F	Fever, Cough, Purulent Sputum, Hemoptysis
5	05 yr	F	Fever, Cough, Dyspnea
6	2.5 yr	F	Fever, Cough, Dyspnea

Table II

Summary of symptoms/signs in patients

Symptoms/Signs	No. of patients (%)
Cough	5 (84)
Fever	4 (67)
Dyspnea	4 (67)
Purulent sputum	2 (33)
Hemoptysis	1 (17)
Asymptomatic	1 (17)

Complications of bronchogenic cysts occurred in 4 patients (67%). In 1 patient (Patient 2), a ruptured, infected, parenchymal bronchogenic cyst caused pleuritis and empyema (Fig. 1). Three patients with intrapulmonary bronchogenic cyst presented with purulent sputum. A 6-year-old patient (Patient 4), who presented with hemoptysis, had a fistula between a bronchogenic cyst and the right lower lobe bronchus.

Location of the cyst

Two of the cysts were mediastinal and rests were intraparenchymal. Mediastinal cysts were located in right & left hilar, one in each location. The location of the intraparenchymal cyst were right middle lobe (1), right lower lobe (2) and left upper lobe (1).

Diagnostic studies

All patients underwent chest x-ray postero-anterior and lateral view and CT scan of chest for diagnostic purpose. In one patient cyst was not seen as infected cyst was ruptured into the pleural cavity. This patient presented as a case of empyema thoracis. One patient showed air fluid level and had bronchial communication with the cyst.

The radiographic characteristics of the bronchogenic cyst were those of a round, well circumscribed, unilocular or multilocular mass, with density ranging from that of water to high density. Intrapulmonary cysts also presented as an air-filled cyst or showed an air-fluid level.

Surgery and histopathology

In all patients, operative approach was a postero-lateral thoracotomy. Complete surgical excision was performed in two cases of mediastinal cysts. In addition one patient needed decortication .Intrapulmonary cysts necessitated lobectomy (4). Right upper bilobectomy and right lower bilobectomy were done in two separate patients who had the cyst in right middle lobe and right lower lobe respectively. Marked adhesion and compression on respective adjacent lobe made dissection difficult and non expansion of the lobe. These were summarized in table III.

Table III

Location of cyst and operation performed

Pt No.	Location of Cyst	Surgical Rx
4	Right Middle Lobe	Right Upper Bilobectomy
7	Right Hilar	Resection,Decortication
8	Right Lower Lobe	Right Lower Lobectomy
9	Right Lower Lobe	Right Lower Bilobectomy
10	Left Hilar	Resection
11	Left Upper Lobe	Left Upper Lobectomy

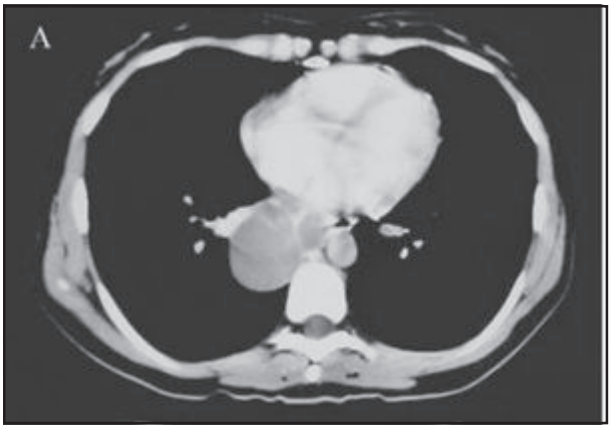

Fig.-1: *CT scan of Bronchogenic cyst*

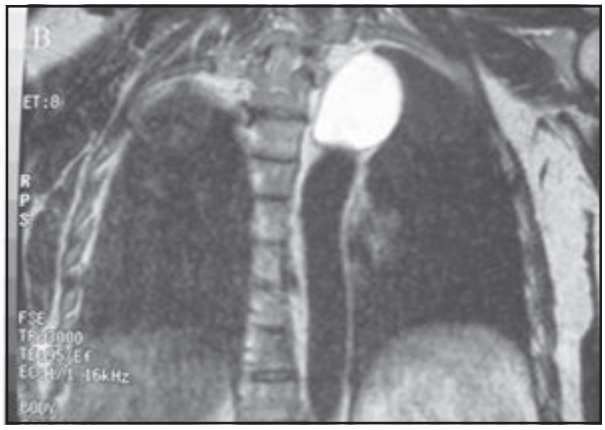

Fig.-2: *CT scan of Bronchogenic cyst*

All cysts were lined by ciliated columnar epithelium. The mucosa contained chronic inflammatory infiltrates. Vascular proliferation was seen in one cyst and other histological findings included bronchial cartilage (2), connective tissue (3), bronchial gland (1) and smooth muscle (3).

There was no operative deaths & no in-hospital mortality. Only intraoperative complication was significant bleeding, from dissection & adhesionolysis. One patient developed persistent air leak, treated consevatively & developed.

Discussion

Bronchogenic cysts are lesions of congenital origin derived from the primitive foregut[6] and are the most common primary cysts of the mediastinum. Most frequently unilocular, they contain clear fluid or, less commonly, hemorrhagic secretions or air[7]. They are lined by columnar ciliated epithelium, and their walls often contain cartilage and bronchial mucous glands[1,2]. It is unusual for them to have a patent connection with the airway, but when present, such a communication may promote infection of the cyst by allowing bacterial entry.

Bronchogenic cysts are found most frequently along the tracheobronchial tree in the mediastinum or within the lung parenchyma[1,2,8]. The relative occurrence rates of intrapulmonary and mediastinal bronchogenic cysts are controversial. Studies by Dilorenzo *et al*.[1], ST. Georges *et al*.[2], and Sarper *et al*.[8] indicate that the frequency of mediastinal bronchogenic cysts is greater, whereas other series, including our own, reflect a higher frequency of cysts with intrapulmonary location[6,9–12]. These discrepancies may result from the difficulties of differential diagnosis between intrapulmonary bronchogenic cysts and other acquired lesions.

They can occur in many atypical locations, including neck[7], cutaneous[13] and subcutaneous tissues[14], pericardium[15], diaphragm[16], abdomen[17], and the intramedullar part of the Spine[18]. They have also been reported to extend from the mediastinum through the diaphragm into the abdomen as dumbbell cysts[19].

Although some bronchogenic cysts are asymptomatic and are incidental findings upon radiography, most cysts are symptomatic and complications are more common in symptomatic patients. The most frequent symptoms are cough, fever, pain, and dyspnea[1-8]. Tracheobronchial compression and pulmonary infections can occur in children because of the relatively soft tracheobronchial tree[9]. In our series, 83% of patients were symptomatic.

Complications of bronchogenic cyst are frequent (67% in our series). Complications may result from compression or irritation of airways, infection of the cyst and pneumonia of the adjacent parenchyma is a common complication, especially in cysts with bronchial communications. The cyst can rupture into the trachea, the pericardial cavity, or the pleural cavity, as it did in one of our patients. Pneumothorax is not a rare complication[6,20] and is usually accompanied by pleuritis. Severe hemoptysis is rarely reported[20].

Chest radiographs and CT scans are the most valuable diagnostic studies. Intrapulmonary bronchogenic cysts generally present as one of three different patterns on the chest radiograph. If no communication is present between the bronchus and parenchyma, the cyst appears as a homogeneous mass with a density of water or a nodule. Differential diagnosis includes benign and malignant tumors, hydatid cysts, granulomas, hamartomas, vascular malformations and lung sequestration. When communication is established with the bronchial tree, the cyst presents

as air filled or contains an air-fluid level. Such a cyst may be confused with lung abscesses, complicated hydatid cysts, infected bullae, and cavities resulting from tuberculosis, fungal diseases, carcinoma, or infarctions. Mediastinal bronchogenic cysts usually present as ovoid or round, well-defined masses of homogeneous water density on the chest radiograph. Occasional air fluid levels or peripheral calcification may be found. The differential diagnosis includes aneurysms, lymphadenopathy, thyroid goiter, cystic hygroma, neurogenic tumors, lymphoma, teratoma, metastatic tumor, enteric, esophageal duplication or pericardial cysts[1, 6, 8-12, 21].

Ultrasonography may be helpful in evaluating those lesions that abut the posterior chest wall and can easily distinguish cystic from solid lesions, thereby eliminating the common neurogenic tumors from consideration. The barium esophagogram is not helpful except in those rare cases of cysts communicating with the gastrointestinal tract[22].

Complete extirpation, with ligation of the point of attachment to the patent bronchus, is usually possible. The prognosis after complete excision is excellent in all patients[1,2,6,20,23]. In patients with pulmonary lesions, lobectomy may be necessary. Incomplete excision will lead to a high recurrence rate.[20]

Transtracheal and percutaneous cyst aspirations have been proposed as alternatives to operation, but these methods are not widely accepted because of possible cyst recurrence, which carries a substantial morbidity rate[2,20].

Conclusion

We conclude that one should go for surgical resection of all suspected bronchogenic cysts in operable candidates.

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