Bronchogenic Cyst in a Patient with Difficult Asthma

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ABSTRACT

Difficult to treat asthma is an asthma syndrome that brings in our mind other differentials. Mediastinal masses are not common findings, but are important variables. Bronchogenic cyst is a congenital anomaly of the foregut that is typically found in the mediastinum and diagnosed accidentally. We present a 4-year-old girl with allergic asthma that began at 8-months of age and finally a bronchogenic cyst was detected in this patient. The patient had history of asthma since she was eight months old. She had a history of several asthma attacks which had partly responded to asthma management.

During the last episodes of asthma attacks, she was hospitalized in Pediatric Intensive Care Unit. Imaging studies showed a 4×3 cm mass in the posterior part of the thoracic cavity that had led to tracheal narrowing was found for which the patient underwent thoracotomy and in surgical exploration a cyst that had compressed the thoracic trachea. Pathological examination of the cyst revealed a bronchogenic cyst. Bronchogenic cyst is an uncommon developmental abnormality but in a patient with obstructive pattern of airways it should be considered in differential diagnosis of asthma, especially if the asthma management is not successful.

Key words: Asthma; Bronchogenic cyst; Mediastinum

INTRODUCTION

Dyspnea, wheeze and cough are the most common symptoms of asthma. If symptoms do not response to asthma therapy further evaluation should be considered.

Although anatomic abnormalities are less common in asthma, but should be suspected and further diagnostic investigations done.1,2 Bronchogenic cyst is formed during 3 and 6 weeks of embryonic period from the primitive foregut.3 It is commonly located in the mediastinum.4 Approximately 50% of these cysts are present at birth and are more common (74%) in boys.5 They may lead to various symptoms and
complications. The majority of them are discovered incidentally. In differential diagnosis of children with wheezing, diagnosis of asthma is important and in addition to asthma, a mediastinal mass which compresses the airways and may present with wheezing or cough should be considered.

CASE PRESENTATION

We report a 4-year-old girl who was a known case of asthma since eight months of age with a history of 3-4 times asthmatic attacks every year along with respiratory distress, croupy cough for which she had been admitted in different hospitals and had two times admissions in PICU. The patient was a full-term infant delivered via Cesarean delivery and her birth weight was 2550 gr. Familial history showed allergic conjunctivitis in mother and allergic rhinitis in uncles. The patient's symptoms exaggerated with exposure to perfumes, spices and smoking. Due to several presentations and delayed response to asthma management, other causes were investigated. On her last admission she had severe respiratory distress, croupy cough, expiratory and inspiratory ronchi in chest exam.

![Figure 1. Chest radiography (PA): Mediastinal widening is seen (arrow) is suggested mediastinal mass.](image1)

She did not respond to medical management such as bronchodilator and systemic steroid in PICU, and no complete response occurred due to questionable abnormal bulging in the upper mediastinum in CXR (Figure 1).

A 4×3 cm mass in posterior aspect of mediastinum compressing trachea was detected in Spiral CT with contrast (Figure 3). Arterial blood gas showed \( Pao_2 = 49.8 \) mmHg and \( O_2sat = 88.6\% \). Pulmonary function tests with bronchodilator challenge showed obstructive pattern with significant increase in FEV1 post bronchodilators. A surgical consultation was done. The patient underwent right-posterolateral thoracotomy. Surgical examination revealed a cyst compressing the trachea (severe stenosis of trachea just up to carina) with large fistulous tract of upper cyst to trachea and an evidence of tracheomalacia. The cyst had been adherent to the esophagus and was gently removed. The cyst which was full of a gelatinous content was of \( 10 \times 8 \times 7 \) cm size. Pathologist demonstrated a cystic structure with milky fluid. The cyst wall was lined by ciliated pseudostratified columnar epithelial cells with underlying fibrovascular connective tissue. Mucus glands with cartilaginous tissue were also present (Figure 4). There was no evidence of malignancy in cyst fluid cytology. She had no postoperative complication.

![Figure 2. Chest CT scan (HRCT): Mediastinal emphysema.](image2)
Following two months of medial treatment, spirometry was performed which showed an improvement in the obstructive pattern. Up to now the patient has not had any asthma attack and her disease is conveniently controlled.

**DISCUSSION**

Difficult to treat asthma is asthma syndrome that brings in our mind other differentials. Mediastinal masses are not common findings, but are important variables. Congenital bronchogenic cysts are rare (an incidence of 1/42000), which are benign developmental malformation of the embryonic foregut due to aberrant budding from the ventral diverticulum. These cysts most frequently occur in the mediastinum and within the lung but their locations vary depending on the level of the abnormal budding. Gursoy, et al reported in 28 patients bronchogenic cyst (BC) at the pulmonary parenchyma (53.5%), mediastinum (43%) and intrathorasic extrapulmonary (3.5%). In our patient BC originated from the trachea in posterior mediastinum. It is a less common site for BC. It is poorly recognized by clinicians due to variable presentation.6,7,11 BC is usually asymptomatic, unless it becomes infected, larger to compress or gets perforated.7,15 The most common symptoms are cough, fever, chest pain and dyspnea.6,7,11 Mediastinal mass due to compression of the airway may present with wheezing or cough. Altyn et al. reported a 14-month-old boy with a BC in the mediastinum which had caused respiratory distress.1 Our patient with presentation of dyspnea and respiratory distress was hospitalized in PICU and further imaging studies showed a mass compressing the trachea. In almost all cases, diagnosis is based on microscopic studies which reveal ciliated and mucin-producing pseudostratified columnar epithelium of respiratory type, but may show various pathological patterns.3,11 No malignant change of BC has been reported in pediatric population.4 Microscopic studies in the patient revealed a cystic structure lined by ciliated pseudostratified columnar epithelial cells with no evidence of malignancy. Early surgical treatment is suggested both for diagnosis and prevention of complications.2,13,15 In differential diagnosis of asthma in children, physicians should be careful and in some patients, CXR or CT should be done to facilitate diagnosis. The patient had several previous attacks with poor response to bronchodilators with a pattern of obstructive airway in spirometry. Previous CXR seemed to be normal. She had a positive family history of atopy, so all findings were suggestive for allergic asthma, but severe croupy cough and delayed response to asthma management necessitated further investigations which led, to the
diagnosis of BC. Although croupy cough, respiratory distress could be noticed as presenting symptoms of asthma in children, allergic asthma in children not responding to conventional management protocols needs more exhaustive evaluation.

REFERENCE


