

CHIRURGIE THORACIQUE/ THORACIQUE SURGERY

INFECTED BRONCHOGENIC CYST SIMULATING ACUTE SEVERE EXACERBATION OF BRONCHIAL ASTHMA

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SUMMARY

Upper airway obstruction classically produces stridor. However, wheezes can be a feature of upper airway obstruction under certain conditions in which case it becomes a diagnostic challenge. A case of infected mediastinal bronchogenic cyst earlier managed as bronchial asthma is presented to highlight such feature. A seven year old African boy presented with a 3-day history of cough and fever; and difficulty in breathing of a day's duration. He has had past episodes of cough and difficulty in breathing more than 6 years with occasional fever and which responded to Franol[®] and Paracetamol[®] given *per oris*. During this acute attack, he failed to respond to standard treatment for acute severe exacerbation of bronchial asthma. Radiologic examinations revealed features compatible with posterior mediastinal mass. A right postero-lateral thoracotomy revealed a tense cystic mass in the posterior mediastinum and 100ml of pus was drained and the cyst was completely excised. Bronchogenic cyst though, a rare cause of wheezing should be considered when a mediastinal mass is suggested on chest radiography.

Key words : Bronchogenic cyst, Simulating, Bronchial asthma, Childhood

Introduction

Sounds emanating from the airway obstruction give a clue as to the possible origin, whether upper or lower airway. Upper airway obstruction classically produces stridor because of the wide diameter of the airway involved and turbulent airflow generated, whereas in the lower airway the smaller diameter causes a whistling musical sound termed wheeze¹. However, wheezes can be a feature of upper airway obstruction under certain conditions². When this happens, upper airway obstruction becomes a diagnostic challenge due to confusion with the more common cause of airflow obstruction, such as asthma and chronic obstructive pulmonary disease. It is however imperative to make the distinction between upper airway obstruction and lower airway obstruction because as an important cause of airflow limitation, it has the potential to produce acute airway compromise and respiratory failure which may be amenable to curative surgery.

The purpose of this study is to underscore the fact that not all that wheezes is asthma, although most does. We, therefore report a case of infected bronchogenic cyst earlier managed as a case of bronchial asthma to highlight such feature.

Case report

A seven-year-old black African boy presented with a 3-day history of cough and fever; and difficulty in breathing of a day's duration. Cough was paroxysmal, worse in the night and with occasional post-tussive vomiting and exhaustion. The fever was high grade but there were no chills or rigors. He developed difficulty in breathing on the second day of illness, which was associated with significant orthopnea and limitation of speech to monosyllables.

He has had past episodes of cough and difficulty in breathing with occasional fever from age 3 months, once or twice every 2 months; and later on weekly from age 3yrs. Symptoms were relieved by Frano[®] and Paracetamol[®] given *per oris*, which were obtained from a neighbourhood patent medicine store. A similar episode 2 years earlier was treated at a peripheral hospital with an intravenous drug suspected to be aminophylline with prompt relief. There were no identified precipitating factors, history suggestive of atopy or family history of asthma.

Physical examination revealed an acutely ill-looking and sweaty boy. The temperature was normal (T=36.9°C), and he was pink in room air. Hydration status was optimal. The major positive findings were in the respiratory system which revealed

the following: a loud expiratory sound which was characterized as a wheeze, a barrel shaped chest, severe respiratory distress as shown by flaring ala nasae, and intercostals, subcostal, lower chest wall, supraclavicular and suprasternal recessions. He was tachypneic with respiratory rate of 44 breaths/min. The percussion notes were resonant, and there were widespread inspiratory and expiratory wheezes with a few coarse crackles at the lung bases. The pulse rate was 120 beats/min, regular and of good volume. The blood pressure was 120/70-60 mmHg, and the heart sounds were normal with no murmurs. He had a non-tender hepatomegaly, 5cm below the right costal margin. The neurological examination was normal. The diagnosis was severe acute exacerbation of bronchial asthma. The treatment included supplemental oxygen through a nasal catheter at a flow rate of 2 liters/min., salbutamol nebulisation (5mg), and intravenous hydrocortisone sodium succinate (100mg every 6 hours). When he was reviewed at 4hours after commencement of the steroid, he had deteriorated as evidenced by a mild central cyanosis; he was therefore commenced on aminophylline drip at a rate of 0.9mg/kg/hour taking cognizance of the fact that he had had a dose of ephedrine[®] at home before presentation in the hospital. In addition, erythromycin estolate[®], 250 mg every 8 hours was administered. Radiographic findings before thoracotomy (Figures 1-3) were compatible with a mass in the posterior mediastinum with possibilities of paratracheal lymph nodes, thymoma, teratoma, retrosternal goiter and a mediastinal bronchogenic cyst. Review of the patient by the Cardiothoracic Surgical Unit, suggested a diagnosis of posterior mediastinal tumor most likely of neurogenic or bronchogenic origin.

A right postero-lateral thoracotomy was done with finding of a tense cystic mass located between the oesophagus and trachea extending across the midline to the left side; 100ml of purulent material was initially drained and the cyst was completely excised. Two drains were left, one located in the cyst bed and the other in the pleural cavity. The postoperative chest radiograph (Fig 4), showed absence of the previously described mediastinal mass. Associated atelectatic changes were present in the lung bases.

The pus aspirate was sterile on culture. He developed post operative pneumonia, which was effectively treated with ceftazidime after a failure of response to Ciprotab[®]. He was discharged well after 14 days on admission.

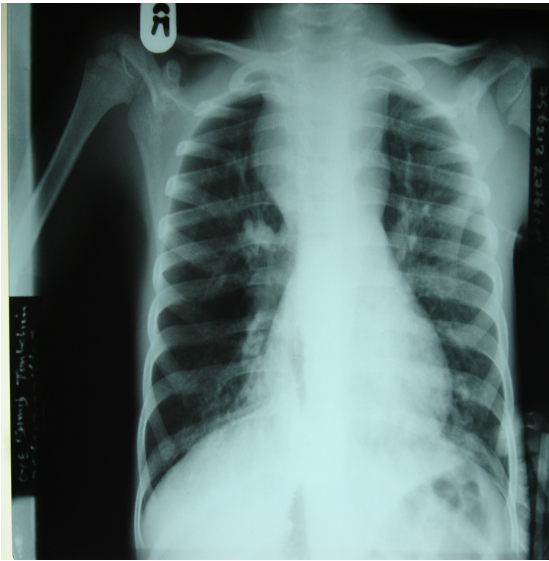


Fig. 1 : The antero-posterior radiograph of the chest showing superior mediastinal widening by a soft tissue mass which extends from the root of the neck into the superior mediastinum. This mass is lobulated and is seen to displace the upper airway to the right. No underlying calcification or bony destruction seen.

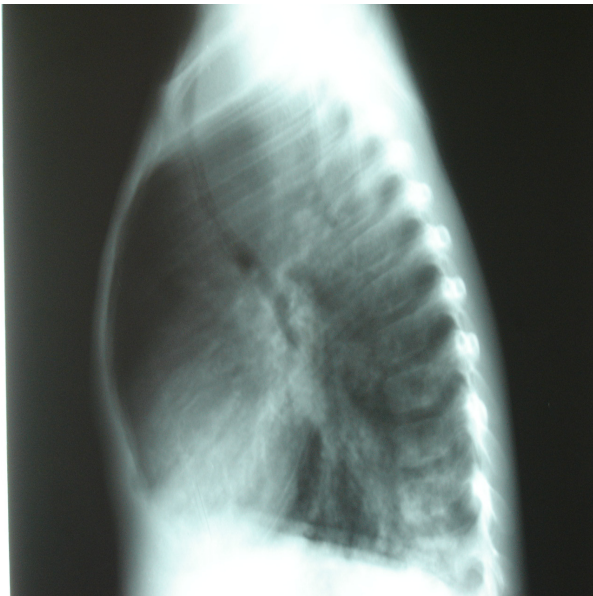


Fig. 2 : Lateral chest radiograph showing a localized mediastinal mass to the posterior compartment with associated anterior bowing and displacement of the trachea suggestive of an upper airway obstruction from a posterior mediastinal mass.



Fig. 3 : The lateral projection of the neck showing normal cervical curvature, vertebral bodies, pedicles and disc spaces. The outlined airway also appears within normal limits.

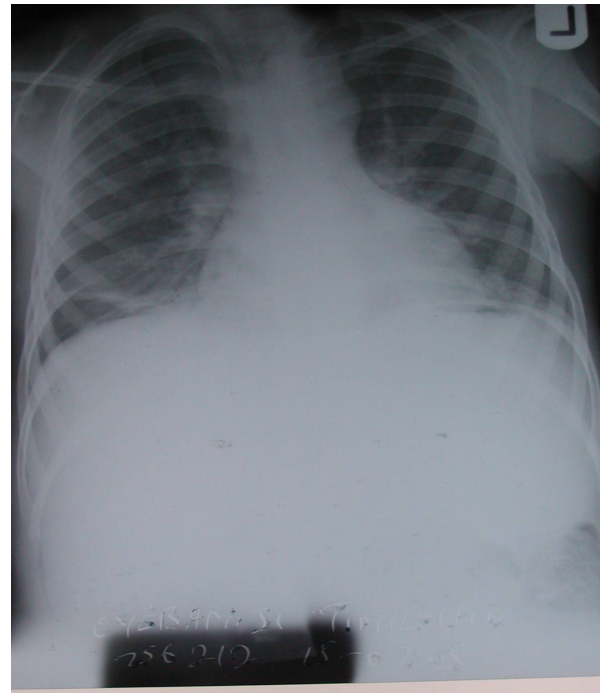


Figure 4. The post-operative chest radiograph showing absence of the previously described mediastinal mass

Discussion

Bronchogenic cyst is a congenital malformation of the respiratory system derived from the primitive foregut³, which varies in size and location. Common locations include mediastinal, paratracheal, paraesophageal and intrapulmonary, and often on the right. Cough is the predominant symptom. Complications include infection, and also increase in size which can then lead to pressure symptoms and signs on the adjacent structures.

Although pulmonary function testing can aid in the diagnosis of bronchial asthma, this has not been found to show any pathognomic features. Demonstration of bronchial hyperresponsiveness although more specific than pulmonary function testing also suffers from the same shortcoming. Hence, the diagnosis of bronchial asthma is essentially clinical as there are no biochemical markers yet for the disease. The clues to the diagnosis being repeated wheezes, recurrent or persistent cough, night time disturbance by wheeze or cough, and these symptoms being incited by viral upper respiratory infections, exercise or excitement, potential allergens such as those associated with pets, pollens, dust, or feathers and cigarette smoke. Our patient presented with many of these features, and the diagnosis of asthma was inadvertently strengthened with the history of therapeutic response to bronchodilators on previous occasions of breathlessness. During admission into our hospital, initial chest radiograph and poor response to β -2 agonist and glucocorticoid treatments heightened suspicion of a superior mediastinal mass which was confirmed by a lateral film. A CT-scan or MRI would have enabled a preoperative diagnosis; however these investigational procedures are very expensive and not within the reach of most patients in our environment. The place of radiologic evaluation is again highlighted by this case, when there is poor response to usual management lines of apparent bronchial asthma.

Bronchogenic cyst presenting with wheezes is uncommon. Diagnosis is often by radiologic evaluation following complaints as in our patient or during investigations for other cardiorespiratory conditions. Surgical excision through thoracotomy is the treatment of choice because it permits complete excision of the cyst and therefore forecloses recurrence⁴ However, there is an associated morbidity due to long muscle-cutting incision. Hence, thoracoscopic resection of mediastinal bronchogenic cyst has been advocated because it provides less operative pain, shorter

hospital stay and better cosmetic outcome than the standard thoracotomy. Pneumonia is a common complication following surgery as seen in this case, often from spillage of infected cyst contents into the tracheobronchial tree.

In general, wheezes are produced by intrathoracic (lower airway) obstructive processes. However, fixed monophonic wheezing is produced by opening and closure of one airway at a time. It is usually caused by obstruction of a large, central airway by a process such as external compression, e.g., vascular ring, adenopathy, and bronchogenic cysts among others. Sometimes these processes will also lead to inspiratory stridor¹.

Acknowledgements

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