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Case Report

Congenital Lung Cyst- A Case Report

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Abstract

Background: Bronchogenic cysts are lesions of congenital origin derived from the primitive foregut and are the most prevailing primary cysts of the mediastinum. Most commonly unilocular, they contain clear fluid or mucinous secretions or, less commonly, haemorrhagic secretions or air. They are lined by columnar ciliated epithelium, and their walls often contain cartilage and bronchial mucous glands. 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. The first successful surgical excision of a bronchogenic cyst was reported by Maier in mid twentieth century leading to its classification based on Mailer postulation. No such reports have been described in South/South Nigeria.

Case summaries

Case 1: We present CH N, a 16 year-old student who presented to us on account of recurrent foul-smelling productive cough and recurrent shortness of breath of 14 years. She received several treatments for bronchial asthma and chest infection from a private hospital with no remarkable improvement. She also had occasional high-grade fevers which also subside with this treatment. On general physical examination she was found to be in mild respiratory distress and not cyanosed with SPO2 of 96% to 98% on room air. Examination of the chest revealed a left apical flattening with the trachea deviation to the left, percussion note was dull and with bronchial breath sounds and widespread crepitation. Spirometric measurement revealed restrictive abnormality. Imaging studies by way of chest X-ray showed a homogenous opacity of left lower lung zone. A diagnosis of left congenital bronchogenic cyst was made. She was started on chest physiotherapy with a postural drainage, antibiotics, bronchodilators and exercise. She had left pneumonectomy.

Case 2: We present B F, a 5day-old neonate who presented to us on account of progressive difficulty in breathing and low peripheral oxygen saturation of less than 60% on room air 12 hour after birth which improved to 98% on 1 L/min of oxygen. She was delivered at a referral hospital in town and was subsequently referred to and admitted into the SBU of the University of Calabar Teaching Hospital. General physical examination revealed a post termed neonate on oxygen by nasal prong, not in any form of distress, she was not cyanosed with SPO2 of 98% on 1 L/min. She was mildly dehydrated. Examination of the chest revealed a bulging right chest wall with trachea deviation to the left. Percussion note was dull with absent air entry over the right lower lung zone. Radiological examination by way of chest X-ray showed an air-filled mass occupying the middle and the lower lung zone causing mediastinal shift with compressive collapsed of left contralateral lung. A diagnosis of congenital bronchogenic cyst was made. She was started on prophylaxis vitamin K, antibiotics, IVF and nil per oral. She had blood workup and she had thoracotomy and cyst excision.

Conclusion: The diagnosis of congenital bronchogenic cyst should be made in adolescent or in neonates who presents with lung compression or recurrent infection symptoms.

Keywords: Congenital; Lung cyst; Hypoplasia; Pneumonectomy; Cystectomy; Lung function test; Continuous positive airway pressure

Introduction

Bronchogenic cysts are lesions of congenital origin derived from the primitive foregut and are the most prevailing primary cysts of the mediastinum as in these two reported cases. Most often unilocular, they may contain clear fluid or mucoid secretions or, less commonly, haemorrhagic secretions or air. They are lined by columnar ciliated epithelium, and their walls often contain cartilage and bronchial mucous glands. It is not common 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. The first successful surgical excision of a bronchogenic cyst was reported by Maier in mid twentieth century leading to its classification called Maier's classification1. No such reports have been described in South/South Nigeria.

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Case Presentation

Case 1:

We report a 16 year-old student who presented to us with a history of recurrent cough productive of copious foul-smelling sputum of 14 year with associated recurrent difficulty in breathing at rest and on exertion, but no history of orthopnoea, there is a history of recurrent high-grade fever with chills and rigors at onset subsequently low grade and continuous fever. Not a known asthmatic but was placed on occasional bronchodilators with no relieve of symptoms. She was seen in outpatient clinic and admitted, investigated for a congenital bronchogenic cyst and to exclude pulmonary tuberculosis. Results of investigations: AFB X3 was no MTB detected, sputum microscopy, culture and sensitivity did not grow anything, lung function tests at presentation, following weeks of chest physiotherapy and post

operation see Table 1. She had pneumonectomy with operative findings of left lung cyst occupying the upper part of the lower lobe extending downward and upward compressing the left lung with hypoplasia of the upper lobe which was non inflatable even with Continuous Positive Airway Pressure ventilation (CPAP) and adhesion of the lung to the pleurae, pericardium and mediastinum.

Histology: Sections shows lung tissue displaying interstitial pneumonia, composed of lymphocytic exudates involving the interalveolar tissue. In some alveolar space there are fibrin exudates. The pleurae are involved in organizing pleurisy with large area of progressive fibrosis with occasional desquamate changes are seen in the smaller airway.

Case 2

We report a 5 day-old neonate who presented to us on account of progressive difficulty in breathing and low peripheral oxygen saturation of less than 60% on room air which improved to 98% on 1 L/min of oxygen. She was delivered at a referral hospital in town by vertex delivery and child cried immediately after birth. She was later referred to and admitted into the SBU of the University of Calabar Teaching Hospital. The pregnancy, antenatal care and delivery were uneventful. General physical examination revealed a post termed neonate on oxygen by nasal prong, not in any form of distress, she was not cyanosed with SPO_2 of 98% on 1 L/min. She looked mildly dehydrated. Examination of the chest revealed a bulging right chest wall with trachea deviation to the left. Percussion note was dull on the right lower lung zone and on the left lower zone with absent air entry over the right lower and left lower lung zones. Radiological examination by way of chest X-ray showed an air-filled mass occupying the middle and the lower lung zones causing mediastinal shift and compression collapsed of left lower lobe. A diagnosis of congenital bronchogenic cyst was made. She was started on prophylaxis vitamin K, antibiotics, IVF and nil per oral.

Discussion

This girl presented to us 14 years after the onset of recurrent cough productive of copious foul-smelling sputum with associated recurrent difficulty in breathing present at rest and on exertion and high-grade fever with chills and rigors at onset subsequently low-grade continuous fever (Table 2). These findings are similar to work done by Sarper et al. [1]. She was not a known asthmatic but has received steroids, bronchodilators and antibiotics. She was seen at the outpatient clinic, admitted, investigated for pulmonary tuberculosis. Results were all negative for tuberculosis. Chest X-ray (Figure 1-4) suggested a mass on the left lung field occupying almost the entire left hemi thorax and the CT-Scan (Figure 5) showed a huge emphysematous bulla in the left upper lung zone with the rest of the field showing extensive dilated cyst occupying almost the entire lower lung zone with multiple cystic bronchiectasis of the left lung. She had a combination of chest physiotherapy, pre-operative exercise, antibiotics and bronchodilators (Figure 6). She eventually had dry cough with an improved lung function. She had a left Pneumonectomy due to long standing compression of otherwise health lung with extensive fibrosis and non-inflation of the lung. Pathologic examination confirmed the diagnosis of bronchogenic cyst (Figure 7).

Patient presented with FEV1 of 1.6L which was inadequate for pneumonectomy, she had chest physiotherapy, antibiotics and pre-operative exercise with a marginal increase in FEV1 to 1.7L (Table 1) and considering other physiological parameters she had

Table 1: Preoperative exercise equivalent of 5 flights of steps is predictive of survival post operation.

Date	FVC	FEV1	Predicted postoperative FEV1=FEV1 [1-no of lung segment 0.0526] in L
18.11.2012	1.64 L	1.64 L	0.86L
25.11.2012	2.68	1.36	0.72L
02.12.2012	2.79	1.74	0.92
13.12.2012	1.78	1.01	
15.12.2012	1.85	1.11	

Table 2: Patient's characteristics.

Patient's characteristics	Case 1	Case 2	Remarks
Sex	Female	Female	100%
Age at Presentation	16th Year	Second day of life	
Difficulty in breathing	Mild	Severe	
SPO^2 at presentation	97%-98%	60%	Large cyst with severe compression
Features of Infection	Recurrent chest infection requiring hospital admission	Born and referred the same day	
Side of cyst	Left sided in between lobed and compress both lobes	Right sided in between the middle and lower lobes compressing both lobes	50%
Types of Presentation	Acute or chronic	Acute	
Type of Surgery	Lt Pneumonectomy	Rt cyst excision	
Location of cyst	Lower part of Lt upper lobe	Between the Rt middle and lower lobes	Both were the intrapulmonary cysts

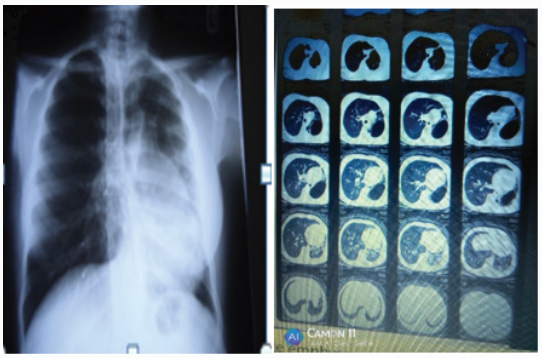

Figure 1: Chest X-ray/CT-scan showing a mass occupying almost the left hemi thorax (Case 1).

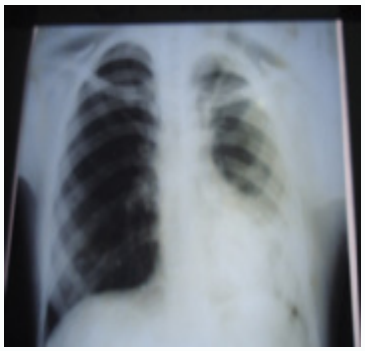

Figure 2: Chest X-ray showing the mass occupying almost the left hemi thorax following chest physiotherapy, bronchodilators and antibiotics pre operation.

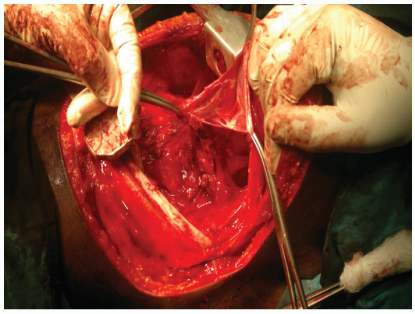

Figure 3: Case 1.

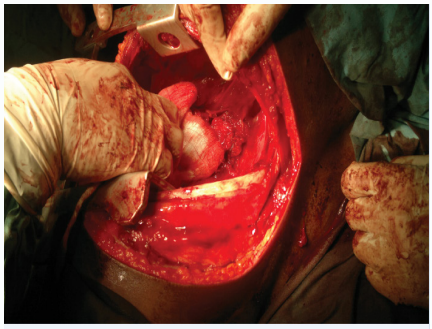

Figure 4: Case 1.

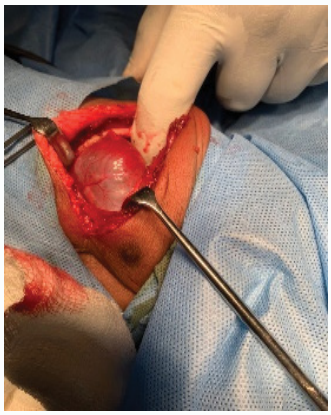

Figure 5: The bronchogenic cyst as seen at Surgery (Case 2).

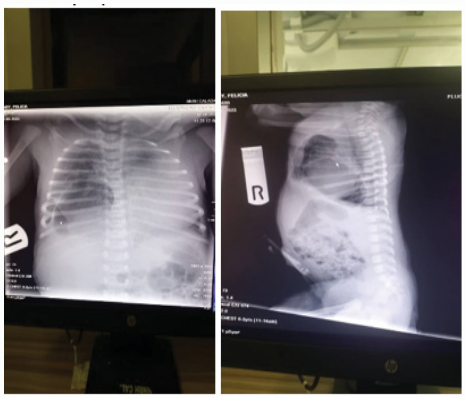

Figure 6: Right Upper lobe congenital cyst (Case 2).

Figure 7: Chest CT Scan showing cyst occupying half of the right chest (Case 2).

pneumonectomy with initial drop in FEV1 but presently patient is doing well at full capacity. The second patient was a 5 day old female neonate who presented to us twelfth hour after birth because of progressive difficulty in breathing and low peripheral oxygen saturation. She had a Chest X-ray (anteroposterior and right lateral which showed an air containing cyst). She was managed on oxygen by nasal prong, nil per oral, intravenous fluid and antibiotics, her father became her walking donor. The size of the cyst determines the time of patient's presentation. The larger the size of the cyst, the worse the compressive symptoms and the early the presentation as in this second case. She had the cyst excised through a muscle sparing right posterolateral thoracotomy and excision of the cyst.

Congenital bronchogenic cyst is a congenital pulmonary malformation from the primitive foregut with a prevalence of 1: 42,000-68,000. It is the most common primary cyst of the mediastinum [1], it constitutes 13% to 15% of the congenital lung cyst and 6% to 10% of all primary intrathoracic masses in the infancy and children. They are formed from the abnormal budding of the ventral foregut anywhere along the respiratory system and oesophagus [2]. The location of the bronchogenic cyst can vary depending on the time of their formation [3]. Up to 85% are in the mediastinum and 12% in the lung parenchyma involving the lower lobes whereas, in our patient it was located in the lower part of the left upper lobe enlarging downward and upward compressing the lower and upper lobe parenchyma. While in the second patient it arises from the junction between the right middle and right lower lobes.

Sarper A et al. [1] in their series reported 4 of their 6 cases were located in the upper lobe However, un usual locations like the neck, pericardium or abdomen have been reported [4,5]. There is a slight male preponderance in patients with bronchogenic cyst (81.8%:18.2%) [1,5]. Whereas two of our only patients were female. The age at diagnosis ranges from the neonatal period through the sixth decade. However, the majority of bronchogenic cysts are diagnosed in patients below the age of 10 years, one third diagnosed by the age 2% and 10% in the neonatal period [1,5].

Tibet et al. reported that bronchogenic were symptomatic in 70.8% of all affected children 1, these symptoms are either secondary to their compressive effect of the enlarged cyst on adjacent structures or infection in the cyst [2]. In our first patient it was a combination of both compression and infection. In the second patient it was more of compressive symptoms.

Compression is common in infants and young children presenting as intercostal retraction, cough, and wheeze while infection is more common in the adults presenting as recurrent pneumonia [1]. Considering the duration of symptoms in our first patient it is a combination of both infant and adult's symptoms. In rare conditions congenital mediastinal bronchogenic cyst can present in the neonatal periods with severe respiratory distress and stridor [1] as in our second case.

Bronchogenic cysts are lesions of congenital origin derived from the primitive foregut and are the most common primary cyst of the mediastinum. They comprise 10% of all primary intrathoracic masses in infants and childhood there is a slight male preponderance in patients with bronchogenic cysts (57%:43%). The age at diagnosis range from the new born period to the sixth decade. However, the majority of the bronchogenic cysts are diagnosed below the age of 10 years, one third diagnosed by the age two, 10% in the neonatal period.

Conclusion

Congenital bronchogenic are rare primitive foregut malformations that can present at birth and up to the 6th decade. It usually presents with either compressive or infective symptoms or a combination of the two. The diagnosis is made early in life while in adolescent and adulthood quality time and resources are wasted in investigation and treatment for other conditions.

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