

Refractory Wheeze: A Rare Presentation of Mediastinal Type Bronchogenic Cyst In A Child.

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ABSTRACT

A 2 year 7 month-old male child presented with recurrent wheezing which did not respond to standard inhalational bronchodilator therapy. Chest X-ray postero-anterior view revealed an opacity in right lung which after doing an MRI chest was provisionally diagnosed to be a multiloculated bronchogenic cyst with intralesional hemorrhage. Surgical excision of the cyst was done and child improved over next 6 month follow up period. Congenital lesions need to be excluded in a child with refractory wheeze.

Keywords: Bronchogenic cyst, MRI chest, Refractory wheeze.

INTRODUCTION

Bronchogenic cysts are fluid filled cysts arising as abnormality of embryological budding of tracheobronchial tree.^[1] Mediastinal bronchogenic cysts are frequently found incidentally on routine chest radiography. Usually they are like well-defined smooth masses near the carina.^[1,2] We report the case of huge multiloculated right mediastinal bronchogenic cysts presenting with recurrent wheeze.

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CASE REPORT

A 2 year 7 month old boy from rural Bengal presented to the emergency department with recurrent attacks of dry cough and wheeze for last 1 year and fever for 4 days. Fever subsided after hospital admission. Previously, he was being treated elsewhere with inhalational salbutamol and budesonide but for last 1 month exacerbations increased in frequency and hampered sleep and daily activities. So he was referred to our institute. There was no family history of asthma, atopy, allergic rhinitis, food or drug allergy or any cardiovascular disorder. There was nothing significant in the perinatal or developmental history. Child was developmentally normal and immunised as per his age.

On physical examination, blood pressure was 96/62 mmHg, pulse rate 108/min and respiratory rate 48/min with oxygen saturation of 91% on room air. Body temperature was 99.4⁰ F. His height and weight were within normal limits as per growth charts. Heart sounds were normal. Trachea was centrally placed. There was no ronchi or crackles. Breath sound was diminished in right infraclavicular, mammary and inframammary regions. On percussion these regions sounded dull.

Routine laboratory tests, electrocardiogram, 2-D echocardiogram were normal. Arterial blood gas showed compensated respiratory alkalosis. Chest X-ray postero-anterior view [Figure 1] revealed large well demarcated ovoid shaped opacity with sharp border involving right upper, middle and lower zones. A high resolution CT scan of thorax was advised but patient party refused to do it in fear of radiation hazard. Alternatively, an MRI scan of thorax was done which [Figure 2& 3] revealed a large cystic multiloculated well defined lesion measuring 10 cm X 5.3 cm X 9.2 cmin the anterior mediastinum compressing the underlying lung and pushing mediastinal vessels posteriorly and to the left. Lesion showed hyperintensity in both T1 and T2 weighted images. Provisional diagnosis of bronchogenic cyst (with probable intralesional hemorrhage) was made. Surgical exploration with removal of cyst was done. Histopathological report confirmed the provisional diagnosis of bronchogenic cyst. The child is in regular follow up and has improved significantly in last 6 months.



Figure 1: Chest X-ray (PA view) showing a large well demarcated ovoid shaped opacity with sharp border involving right upper, middle and lower zones.



Figure 2: Magnetic Resonance Imaging coronal T2W image showing hyperintense huge bronchogenic cyst in right mediastinum pushing the vessels towards left side.

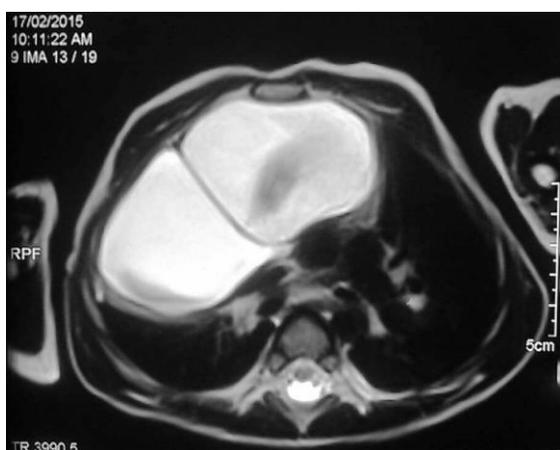


Figure 3: Magnetic Resonance Imaging axial T2W image showing hyperintense bronchogenic cyst with septation and loculation.

DISCUSSION

Bronchogenic cysts constitute 10–15% of all primary mediastinal masses and can be classified as: intrapulmonary and mediastinal. Mediastinal type bronchogenic cysts are further classified into five subtypes: paratracheal, carinal, hilar, para-

esophageal, or miscellaneous.^[3] The cysts are lined by ciliated secretory respiratory epithelium with cartilage, smooth muscle, fibrous tissue and mucous glands. These may be filled with fluid or air or both depending upon its communication with the airways.^[4]

Around 90% of mediastinal types of bronchogenic cysts are usually asymptomatic, incidentally found during obtaining chest radiograms. Like any other space occupying lesion in the mediastinum, the location and size of the cyst will determine the appearance of the symptoms. When they enlarge, they produce symptoms by pressing upon neighbouring anatomical structures. Paratracheal or carinal type can give rise to dyspnea due to compression of nearby trachea or bronchi.^[5]

Infrequently a bronchogenic cyst may get infected and can rupture into the nearest bronchus giving rise to a constellation of symptoms like fever, hemoptysis, mucopurulent sputum production etc. If size of carinal type bronchogenic cyst is huge then it may compress the nearby left atrium of the heart. The patient might complain of heaviness in chest especially on exertion and electrocardiogram can reveal left atrial overload.^[5] In our patient in spite of the large size no such overload was evidenced.

Radiological examination may show a space occupying lesion in mediastinum, a cyst with air fluid level, displacement of trachea or primary bronchus, atelectasis or emphysema secondary to secondary effects.^[6] Bronchoscopy may show compression of major bronchus or trachea. Barium swallow may show indentation of esophagus.^[7] An accurate diagnosis needs CT or MRI scan with or without the use of ultrasonogram.^[8]

CT scan identifies the cysts as well-defined, discrete, non-enhancing masses. The fluid within the cyst may appear to vary from water density to higher density according to its content. Sometimes flecks of calcium are seen within the cyst fluid and this is known as “milk of calcium”.^[9]

Magnetic resonance imaging (MRI) appearance may also vary according to the nature of the cyst-fluid, low or high signal intensity in T1-weighted imaging, and bright signal intensity in T2-weighted imaging. In T1-weighted images, fat, proteinaceous and hemorrhagic fluids appear in white and water appears in low signal intensity. In T2-weighted images water with or without proteins appears in high signal intensity.^[9]

A symptomatic bronchogenic cyst is an indication for surgical removal by either cyst excision or lobectomy by open thoracotomy or video assisted thoracic surgery (VATS).^[10] There are controversies regarding whether an incidentally found bronchogenic cyst warrants operation or not, but as the course of these pathologies are unpredictable, several surgeons prefer preventive surgery.^[10]

CONCLUSION

This case highlights that bronchogenic cysts should be suspected in early childhood having recurrent cough or wheeze with or without dysphagia. A timely CT/MRI scan can detect and appropriate surgical procedure can alleviate the symptoms and even prevent long term complications.

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