Clinical Brief

Bronchogenic Cyst – Cause of Refractory Wheezing in Infancy

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Abstract. A 1½-year-old male child with past history of recurrent wheezing presented with a severe and refractory episode following a peanut aspiration. A bronchoscopy was done and the peanut removed. However, as the wheezing persisted a detailed evaluation was done which revealed a bronchogenic cyst. Surgical excision of the cyst was done and the child improved symptomatically. Congenital lesions need to be ruled out in infants and children with persistent and refractory wheezing. **[Indian J Pediatr 2005; 72 (4) : 363-364]**

Key words : *Refractory wheezing; Bronchogenic cyst; Bronchoscopy*

Bronchogenic cysts are fluid filled cysts of tracheobronchogenous origin, usually manifesting in early infancy. Surgical excision is the treatment of choice.[1] The case described refers to a child having recurrent episodes of wheezing, refractory to medical therapy, who on detailed evaluation was found to have a bronchogenic cyst.

CASE REPORT

A 1-1/2 year old boy was referred to our hospital with complaints of cough, wheezing and breathlessness since 3 days, not responding to the standard anti-asthma therapy. A history suggestive of foreign body aspiration prior to onset of present illness was obtained. The child had past history of multiple wheezing episodes since 6 months of age.

On examination, child was normothermic but dehydrated, cyanosed and in severe respiratory distress with RR-82/min and HR-180/min. Child was drowsy, irritable and in air hunger. Air entry was markedly reduced bilaterally with severe bronchospasm. Arterial blood gas revealed severe respiratory acidosis with hypoxaemia (pH 7.25, PCO_2 64, PO_2-48.5, HCO_3 26.7). X-ray film of chest showed bilateral hyperinflated lung fields. Child was intubated immediately, started on mechanical ventilation and stabilized. An emergency bronchoscopy done revealed a peanut impacted in the trachea, which was removed. Air entry improved and child was extubated after 12 hours. But, despite good bronchodilator therapy, wheezing and respiratory distress persisted with a diminished air entry on left side. A repeat bronchoscopy done revealed no peanut remnants, but showed bulging of the posterior wall of left bronchus into the lumen, which was filled with granulation tissue. An indented left lateral wall of oesophagus with shift to right was noted on barium swallow (Fig. 1). 2-D Echo done did not reveal any vascular ring or tumour. Spiral CT scan of the chest showed a cystic lesion 2.5 x 2 cm in the posterior mediastinum, to the left of oesophagus at the subcarinal level compressing the left main bronchus with collapsed left lung (Fig. 2). Surgical excision of the cyst was done and the child improved symptomatically. Biopsy report was consistent with bronchogenic cyst.

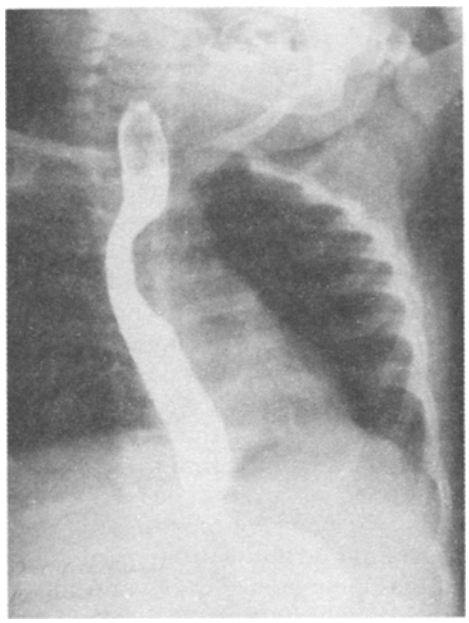

Fig. 1. Left lateral wall of oesophagus with shift to right

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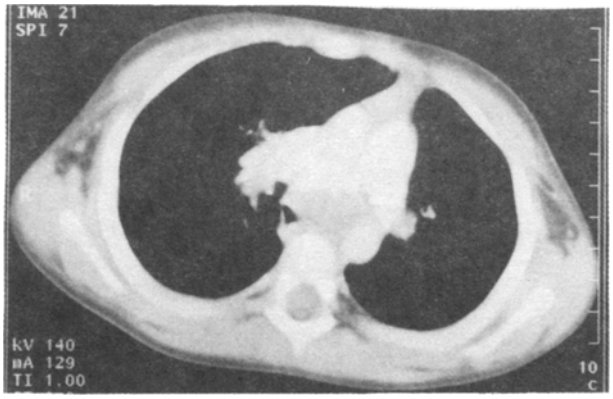

Fig. 2. Collapsed left lung

DISCUSSION

This child presented with severe wheezing refractory to all standard modalities of treatment. Usual conditions leading to wheezing in the differential diagnosis were considered and the child was investigated with bronchoscopy, echocardiogram, barium swallow and CT scan-chest. A diagnosis of a posterior mediastinal mass was made from these investigations.

Posterior mediastinal masses seen in the infants and children are neurogenic tumors, bronchogenic cyst and gastroenterogenous cysts, the last being the most common.[2] Bronchogenic cyst arises from abnormal budding of the tracheal diverticulum of the primitive foregut.[3] These fluid filled cysts are lined by ciliated epithelium and the walls contain smooth muscle and cartilage. They may or may not communicate with the tracheo-bronchial tree.[2] The common sites in the mediastinum are para-tracheal, carinal, hilar or para-oesophageal.[4] In our case the cyst was subcarinal.[5]

Literature reports the incidence of bronchogenic cysts to be 13-15% of the congenital cystic lung diseases in infants and children.[6] The cases usually manifest due to complications either as infection in the cyst or following cyst enlargement with airway compression. The former presenting as fever, chest pain, cough, haemoptysis while the latter as respiratory distress and inspiratory or expiratory wheeze.[4] Though rarely symptomatic at birth, there are case reports of neonates presenting with stridor and severe respiratory distress due to a mediastinal bronchogenic cyst.[7,8] Physical examination reveals only the secondary effects of the tumor like atelectasis or emphysema rather than the tumur itself.[2] One study reports intrapulmonary cysts to be more symptomatic than mediastinal one. A bronchogenic cyst while uncommon should be in the differential diagnosis of pneumo-mediastinum or medial pneumothorax even in premature babies on ventilators.

Radiological examination of the chest may reveal a space occupying lesion, a cyst with air fluid levels, tracheal displacement and compression or secondary effects like atelectasis or emphysema.[3] Barium swallow may show an indentation of the oesophagus while bronchoscopy reveals compression of trachea and often of one major bronchus.[2]

An accurate diagnosis is possible with CT or MRI scan of the chest aided by selective use of ultrasound.[6] In this case, both barium swallow and bronchoscopy were suggestive of a mediastinal mass and the diagnosis confirmed by CT scan. Antenatal diagnosis is possible with ultrasonography.[4]

A symptomatic bronchogenic cyst is an indication for immediate resection. Cyst excision or a lobectomy is the standard treatment while drainage is a temporary, palliative and risky procedure in cases of life threatening compression. As the long-term prognosis of an asymptomatic cyst is unpredictable, studies advocate a role for preventive surgery.[9] Apart from open thorocotomy, successful cyst resection has also been done by video assisted thoracic surgery (VATS).[10]

Acknowledgements

We thank Mr. R.V. Shenoy for typing the manuscript

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Contributors: TM, AS and RK were involved in the management of the patient and drafting of the manuscript. TM will acts as the guarantor for the paper.