Abstract: Mediastinal and lung cyst are developmental in origin. A mixed lung lesion consisting of a combination of bronchogenic cyst, bronchopulmonary sequestration and congenital cystic adenomatoid malformation suggest a embryologic link for these malformation. The true incidence of congenital lung lesion is unknown. The huge fetal lung lesion have reproducible pathophysiologic effects on the fetus. We are presenting a very rare presentation of this hybrid lesion. 3 months old male child presented with history of recurrent episodes of fever and respiratory distress. Child was evaluated with CT chest with pulmonary angiogram and diagnosed as a case of CCAM with BPS. Hence we proceeded with thoracotomy and lower lobectomy. Histopathology report came as hybrid lesion (CCAM with BPS). Post-op period was uneventful.

Keyword: congenital cystic Adenomatoid malformation (CCAM), Bronchopulmonary sequestration (BPS), Hybrid lesion, Lobectomy.

History: 3 months old, 1st born, term, male child delivered by naturalis with birth weight of 3.2 kg -presented with h/o respiratory distress at birth. Bowel and bladder habits normal. At that time child was managed conservatively. At 45 days of life - post vaccination, again child had fever, respiratory distress and refusal of feeds. Child was investigated and diagnosed as bronchopneumonia with X-ray chest (fig.1). Admitted in NICU & treated. Again at 75 days of life one more episode of respiratory distress and investigated & suspecting diaphragmatic hernia on right side in X-ray chest(fig.2). Hence ,the child was evaluated thoroughly with barium swallow (fig.3) and Contrast enhanced CT chest with pulmonary angiogram (fig.4) and diagnosed as a case of intralobar sequestration with feeding artery from abdominal aorta(fig.5). Hence we proceeded with right posterolateral thoracotomy with the findings of cystic lesion in the right lower lobe with anomalous blood supply from descending aorta (fig.6). Feeding vessels were identified, ligated and divided. The entire cyst along with lower lobectomy was done.
Histopathology: Report came as (fig.7) hybrid lesion of both CCAM & BPS with the findings of polypoid projection of mucosa, increased smooth muscle in the cyst wall, absence of cartilage & inflammation and presence of mucus secreting cells.

Post-op period was uneventful.

Discussion:
The true incidence of cystic lung lesion is unknown. Congenital cystic adenomatoid malformation, Bronchopulmonary sequestration and congenital lobar emphysema are types of cystic lung lesions. Within this CCAM is a benign multicystic mass of nonfunctioning pulmonary tissue. The size of the cyst varies from 1mm to 10cm. The blood supply is from pulmonary circulation. But unfortunately it won't participate in normal gas exchange. Embryologically it arises from pseudoglandular period.

Two types of classification

By stocker: Type 0 - Bronchial dysplasia Type 1 - Bronchial / bronchiolar
Type 2 - Bronchiolar Type 3 - Bronchiolar / Alveolar Type 4 - Peripheral

By Adzick - Macrocytic & Microcytic types.

Lower lobe is the most common site of origin and and right & left sides are equal incidence.

BPS: Bronchopulmonary sequestration is also a nonfunctioning lung mass. Embryologically it develops from developing foregut with systemic arterial blood supply. There are two types - intra lobar & extra lobar variety. Lung lesion closure to paravertebral region always suspect BPS.

Management: For prenatally diagnosed cases and neonates with symptomatic / asymptomatic cases needs further evaluation with contrast CT at 4-6 weeks of life. Lobectomy is the treatment of choice. Those cases of suspecting BPS, CT angio is must to find out anomalous blood supply.

Conclusion: Lower lobe CCAM may associate with BPS. In these conditions beware of anomalous blood supply and better to do CT angiogram to rule out hybrid lesions.

References
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