Bronchopulmonary Foregut Malformation: Continuity of Pulmonary Sequestration and Distal Esophagus

Sir,

Bronchopulmonary foregut malformations (BPFMs) are very rare developmental pathologies. A few case reports are published in the literature. These lesions are defined as bronchogenic cyst (BC) or pulmonary sequestration (PS) that are connected to the esophagus. Proper surgical treatment mainly depends on clarifying the abnormal anatomy by using preoperative imaging studies.

A 5-month male was admitted with fever and cough. Chest X-ray showed right lower zone consolidation (Figure 1A). He received antibiotics with no improvement. Computed tomography revealed a lesion compatible with PS and aberrant vascular branches from the celiac artery and portal vein (Figure 1B). It also showed that a connection between PS and distal esophagus (Figure 1C). Esophagography showed contrast leakage from distal esophagus to the lung lesion (Figure 1D). Bronchoscopy revealed normal tracheobronchial tree. Esophagoscopy revealed a fistula at right wall of the distal esophagus. Thoracotomy revealed an extralobar PS which was connected to esophagus. PS and its connection were removed. Esophagus was repaired. The histopathology revealed PS associated with an esophageal duplication cyst which contained esophageal and gastric mucosa (Figures 1E and 1F).

There are some reports about pulmonary airway malformation communicating with digestive tract in the literature. As a result of close relationship between the respiratory and digestive system in embryonic period, PS and BC can be connected with foregut. Since 1968, Gerle et al. used new terminology as BPFM for such pathologies. In the literature, relation of BC and esophageal duplication cyst is reported. The esophageal lung malformation is another similar lesion in which ipsilateral main bronchus is absent and there is a bronchus connected with only esophagus. In contrast to this pathology, in our case tracheobronchial tree has entirely normal anatomy compatible with BPFM.

Patients diagnosed with BPFMs have variable symptoms to be managed as soon as possible. As a result of connection with the digestive system, fever, cough, pleural effusion and respiratory infection can be seen. Surgical treatment is necessary to maintain esophageal integrity. Isolated PS or PS associated BC are reportedly excised by video-assisted thoracic surgery. Because of esophageal connection, BPFMs may require to be removed by thoracotomy to restore anatomy of the distal esophagus.

BPFMs should be kept in mind before and during surgery when any type of congenital pulmonary malformation is evaluated. Anatomical locations and vascular anatomy of suspected lesions and their relationship with tracheobronchial tree and esophagus should be meticulously analysed for successful surgical treatment.

REFERENCES


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