

폐암으로 오인된 기관지 골연골증

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Bronchopathia Osteochondroplastica Mimicking Lung Malignancy

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Tracheobronchopathia osteochondroplastica (TO) is an uncommon benign disease of an unknown etiology and it affects the cartilaginous walls of large airways. Most cases of TO have been reported to involve the lower two-thirds of the trachea and the proximal bronchi. Unlike the usual cases of TO, exclusive bronchial involvement and the formation of a solitary mass are very rare. We experienced an unusual case that had exclusive bronchial involvement and the formation of a solitary mass and this all mimicked lung malignancy. After surgical resection, we were finally able to diagnose the mass as bronchopathia osteochondroplastica.

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Key words: 1. Cartilage disease
2. Bronchi
3. Lung neoplasms

CASE REPORT

In October, 2009, a 61-year-old woman presented with a 3-month history of cough and intermittent blood-stained sputum. She had frequently experienced symptoms of a common cold. She was a non-smoker and house keeper. She had been medicated captopril as an anti-hypertensive drug. On physical examination, the blood pressure was 130/70 mmHg, pulse rate 89/minute and respiratory rate 20/minute. Mild de-

creased breath sounds were noted at the left lower lung fields. The chest CT showed a segmental consolidation and atelectasis containing a fluid bronchogram in the anteromedial segment of the left lower lobe with several endobronchial calcified nodules and bronchial obstruction in the proximal segmental bronchus (Fig. 1A). A fiberoptic bronchoscopy showed normal tracheal mucosa. The left lower lobar bronchus was totally occluded by a hard irregularly lobulated mass that was difficult for mucosal biopsy (Fig. 1B). The

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본 논문의 저작권 및 전자매체의 지적소유권은 대한흉부외과학회에 있다.

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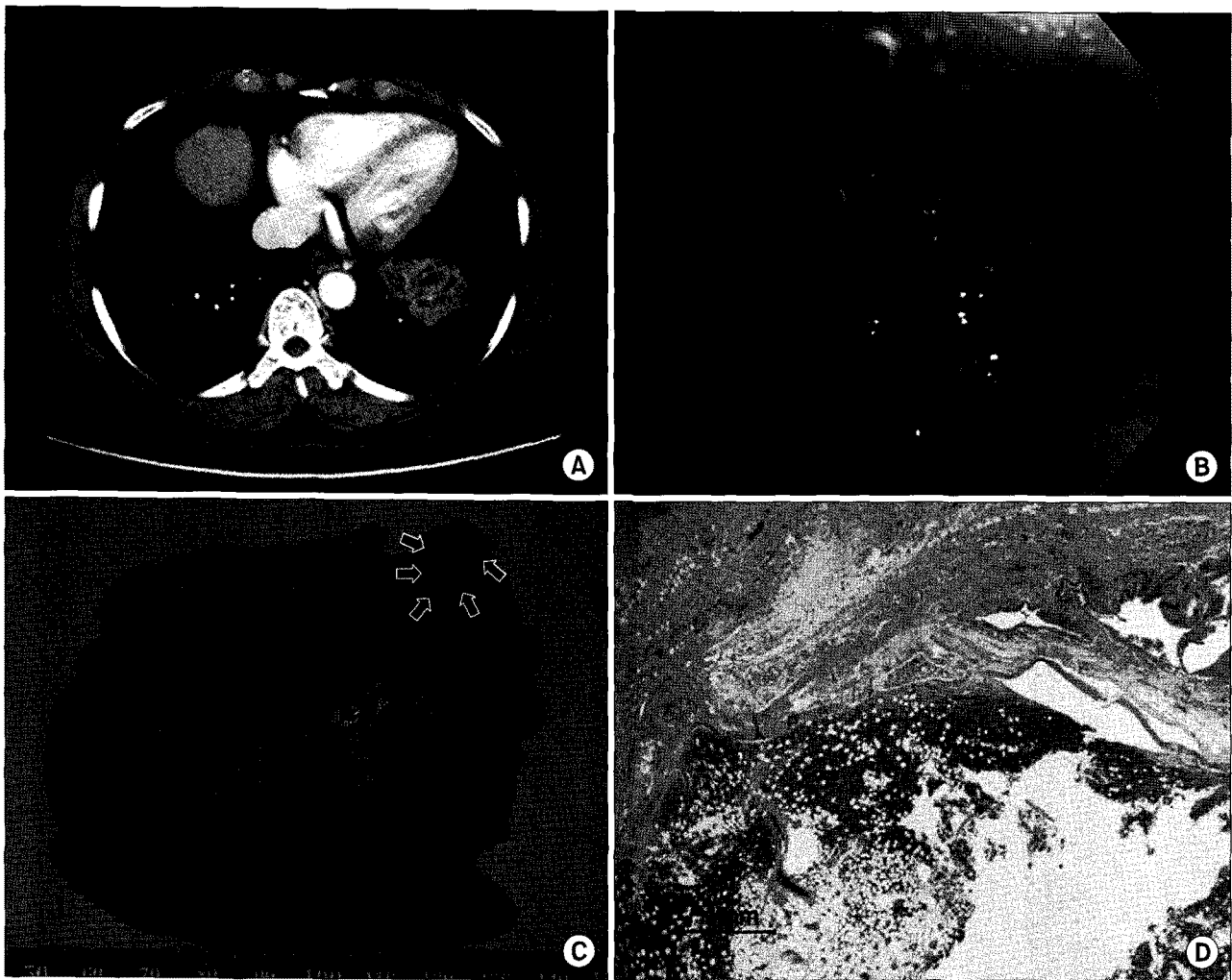


Fig. 1. (A) Chest CT represents a segmental atelectasis of anteromedio-basal segment of left lower lobe with fluid bronchogram. (B) Bronchoscopy shows total obstructing hard endobronchial mass at the orifice of left lower lobe. (C) The section of the specimen revealed an endobronchial solid mass (arrow) without hemorrhage and necrosis. (D) An endobronchial lesion was predominantly composed of bone tissue with bone marrow components (H&E stain, $\times 20$).

bronchoscopic biopsy report demonstrated non-specific inflammation. Although there was no hypermetabolism in ^{18}F FDG PET/CT scan, we could not rule out for lung malignancy. Therefore, we performed left lower lobectomy by way of thoracotomy. The resected tumor was 1.8 \times 1.9 cm-sized solitary calcified mass attached at the orifice of basal segment of the left lower lobe (Fig. 1C). On microscopic examination revealed an endobronchial polypoid mass, mainly composed of mature bone having abundant marrow component and mature cartilage. These features were suggestive for bronchopathia osteochondroplastica (BO) (Fig. 1D). The patient has been

followed up in stable status with controlling postthoracotomy pain without recurrence.

DISCUSSION

Tracheobronchopathia osteochondroplastica (TO) is a rare disorder of the large airways characterized by the presence of multiple, very small osseous nodules localized to the sub-mucosa of the cartilaginous tracheal or bronchial wall[1]. The lesion has primarily been reported to occur in the lower two-thirds of the trachea, and occasionally has been found to

involve the proximal bronchi as well[1,2]. Unlike the usual case of TO, it is very rare that exclusive bronchial involvement and solitary mass formation[3,4].

This is the first published Korean case of (BO) that is exclusive bronchial involvement and solitary mass formation mimicking lung malignancy. The incidence of typical cases of (TO) at autopsy has been estimated at approximately 3/1,000, while data from bronchoscopy have varied widely from 1/125 to 1/6,000[5]. The majority of patients have mild respiratory symptoms, such as a cough, small amount hemoptysis, exertional dyspnea, and wheezing or recurrent tracheobronchial infections[2]. The diagnosis of a (TO) or (BO) can be made by fiberoptic bronchoscopy; the findings of this disorder are unique and include multiple osteocartilaginous calcified nodules within the submucosa of the anterior and lateral aspects of the tracheobronchial tree, and the membranous portions of the trachea are characteristically spared in contrast to tracheobronchial amyloidosis. The chest CT, MRI or virtual bronchoscopy can all detect these submucosal nodules in the trachea and main bronchi; in one-half of the cases the nodules are calcified[6]. In our case, there was no typical multiple tracheobronchial calcified nodules. Because we could not exclude lung malignancy in imaging studies, surgical resection was performed. The histopathology showed calcifications, chondrifications or ossifications of the upper layer of the mucous membranes. The cartilage of the TO nodules could be distinguished from normal cartilaginous rings; the bone tissues may show bone marrow foci with active areas of hema-

topoiesis[3].

There are no specific treatments for prevention of nodular formation or progression available to date. The majority of patients do well with conservative management such as antibiotic treatment, mucolytics or medical hemostasis. Surgical resection would be considered as a treatment in patients with parenchymal lung disease resulting from long-standing occlusion of the airway as in our case.

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=국문 초록=

기관골연골증이나 기관지골연골증은 큰 기도의 연골에 생기는 잘 알려지지 않는 병리를 가진 매우 드문 양성질환으로 보고되고 있다. 대부분 하부기관이나 근위부 기관지에서 발생하는 것으로 보고되고 있다. 대부분의 기관골연골증과는 달리, 기관지를 벗어나서 종괴를 형성하는 기관골연골증은 매우 드물다. 그러나 저자들은 기존의 보고와는 다르게 엽기관지에 종괴를 형성하여 육안적으로 폐암과 유사한 형태를 지닌 기관골연골증을 치험하였으며, 수술적 절제 후 기관지골연골증임을 진단할 수 있었다.

- 중심 단어 : 1. 연골질환
2. 기관지
3. 폐종양