CT Findings of Laryngotracheobronchial Involvement in Tracheopathia Osteoplastica: A Case Report

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We report a case of tracheopathia osteoplastica (TO) that involved both larynx and tracheobronchus. On CT scans, the laryngeal and tracheobronchial cartilages were thickened with irregular calcification. Multiple nodules with or without calcification were seen protruding into the lumen from the anterior and lateral walls.

Index Words: Trachea, CT
Larynx, CT

INTRODUCTION

Tracheopathia osteoplastica (TO) is a rare, benign condition involving the trachea and major bronchi, in which islands of osseous tissue form within the submucosa of the anterior and lateral walls. The posterior walls are spared, as they contain no cartilage. Lesions involving the larynx have only occasionally been reported (1-6).

CASE REPORT

A 62-year-old man was admitted because of severe dyspnea and hoarseness for 45 days, which was aggravated progressively with a recent upper respiratory tract infection. He had experienced less degree of dyspnea and hoarseness 2 years previously, which had not been worsened until admission. Difficulties were present during a tracheostomy with a large amount of emphysema along the neck and upper mediastinum. Endoscopy revealed almost fused vocal cords except a 3 mm opening at the posterior commissural region. CT scans showed irregular enlargement of both arytenoid and cricoid cartilages bilaterally. Glottic and infraglottic larynx was narrowed and elongated, with nodular calcification in its walls (Fig. 1a). The trachea and right main bronchus were deformed and triangular in shape. Thick, horseshoe-shaped, irregular calcific deposits were spread continuously along the anterior and lateral walls (Fig. 1b, c). With protrusion into the tracheal and bronchial lumen, tracheal and bronchial lumina were distorted with crescentic configuration. No calcific deposits were found in the posterior wall of the trachea and right main bronchus. There was no evidence of an extratracheal or extralaryngeal mass to explain the cause of these deformity. Biopsy under a ventilating bronchoscope was performed and histopathology of specimen revealed respiratory mucosa with focal squamous metaplasia and compact bone trabeculae with fatty or fibrotic marrow tissues in the underlying stroma. Chronic inflammatory cell infiltration was associated (Fig. 1d).

DISCUSSION

Tracheopathia Osteoplastica was first described macroscopically by Rokitansky in 1855 and microscopically by Wilks in 1857. Pathologically, the lesions are composed of submucosal islands of hyaline cartilage with areas of lamellar bone and occasional marrow elements. The mucosal surface is intact. A connection to the the perichondrium is often evident, suggesting that the lesions arise from native cartilage (2, 5).

When the larynx is affected, symptoms arise at an earlier stage as in this and other cases; the symptoms may include hoarseness and dyspnea and, because of the close relation to the uppermost part of the esophagus, a sensation of foreign body, salivation, pain and dysphagia with weight loss may also occur (6). There has been only a few reports of TO involving the larynx since the first description of the disease more...
than 100 years ago(6-9).

The histopathologic diagnosis has been made after endotracheal biopsy, segmental resection of the trachea, or lobectomy. Conventional tomography and computed tomography have been reportedly useful in making the diagnosis(1-6, 9). On CT scans(1-3, 5, 9), the tracheal cartilages are thickened with irregular calcification. Multiple nodules with or without calcification may be seen protruding into the lumen from the anterior and lateral walls. This is considered to be pathognomonic for TO. Typically, a long segment of the trachea is involved with possible extension to the main stem bronchi. Demonstration of calcification is essential for the radiologic diagnosis of cartilagenous tumors in general and for TO in particular. Conventional tomography and CT are the only modalities for demonstration of the calcification. However the CT has been superior to conventional tomography because CT can show that not all of the calcifications are located within the endotracheal nodules, but are also in submucosal plaques, and it reveals nodules devoid of calcification (10). There are several diffuse diseases of the trachea and main stem bronchi that decrease the airway diameter: relapsing polychondritis, amyloidosis, sarcoidosis, Wegener's granulomatosis, TO, tracheobronchitis associated with ulcerative colitis, saber sheath trachea, tracheomalacia and bronchomalacia, and infectious disorders. Of these calcification may be seen in relapsing polychondritis, amyloidosis, TO, and saber sheath trachea. Relapsing polychondritis and amyloidosis are systemic disorders in which the tracheobronchial tree is usually involved as a part of characteristic systemic manifestations. Episodic inflammation of the ears, nose, upper airways, and joints with cauliflower ear, soft tissue changes with calcification in the cartilage, and saddle nose deformity are specific findings of relapsing polychondritis. The nodules in amyloidosis may be circumferential, but those of TO typically spare the posterior membranous wall. Saber sheath trachea affects only the intrathoracic trachea.
Jong Deok Kim, et al: CT Findings of Laryngotracheobronchial Involvement in Tracheopathia Osteoplastica

and the coronal diameter is markedly reduced, resulting in a saber-sheath configuration(1, 5, 10).

In summary, if, on CT scans, small patchy calcific deposits were spread along the inner aspect of the thickened laryngeal and tracheobronchial cartilages, protruding into the lumen and sparing the posterior wall, TO should be considered.

REFERENCES


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후두를침범한기관골신생증의CT소견:1예보고

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기관골신생증은기관과주기관지의점막하를따라골·연골조직이형성되어비후되어서기도의변형과협착을초래하는양성질환이며다물로써 발생한다.기관과주기관지의후벽에는연골이없기때문에침범되지 않고전벽과후벽을침범하는것이다른질환과구별되는특정적인소견이므로이질환의진단에는CT가가장정다.또한후두가침범되는경우는매우 드물어서문헌상에단지몇예의보고만되어있었을뿐이다.62세의남자환자에서후두와기관 및주기관지를연속적으로침
범한1예의기관골신생증을문헌고찰과함께보고한다.
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