Tracheobronchopathia Osteochondroplastica: A Case Report

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Abstract

A 40-year-old male was hospitalised with palpable lymphadenopathy on the left side of the neck. Fiberoptic bronchoscopy revealed numerous hard irregular nodules projecting from the anterior and lateral walls of the trachea and extending to the right and left main stem bronchi and to the bronchus intermedius. Histopathologically, tracheal wall biopsy specimens showed the presence of osteocartiloginous tissue in the subepithelial areas. Based on the histopathological examination of the biopsy specimen excised from the left cervical lymphadenopathy, a diagnosis of nodular sclerozing subtype of the Hodgkin's disease was reached. From 1973 until November 1999, about 19 000 bronchoscopies were performed in our center and this patient is the second case of tracheobronchopathia osteochondroplastica (TO) diagnosed by bronchoscopic examination in our center during this time period.

This report summarizes the findings in this patient with TO and Hodgkin's disease.

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Key words: *Bronchoscopy, tracheobronchopathia osteochondroplastica, tracheal ossification*
Abbreviations: *TO: Tracheobronchopathia osteochondroplastica, CT: Computed tomography*

Introduction

Tracheobronchopathia osteochondroplastica (TO) is a rare disorder representing an overgrowth of tracheal cartilage and sometimes associated with ossification into the lumen of the trachea and bronchi (1,2). The etiology of the disease is unknown. Although this disorder is usually discovered as an incidental finding in later life or at autopsy, it may cause obstructive symptoms particularly in the smaller airways (1).

In the past, a diagnosis of TO was most frequently established at autopsy. With the advent of new pulmonary diagnostic techniques including bronchoscopic examination and computed tomography (CT), TO can nowadays be recognised antemortem (3).

Case Report

A 40-year-old car repairman presented with complaints of fever, night sweats, weakness, weight loss, arthralgia and bone pain. The duration of the symptoms was 2 months. The patient had a 40-pack-years-history of cigarette smoking.

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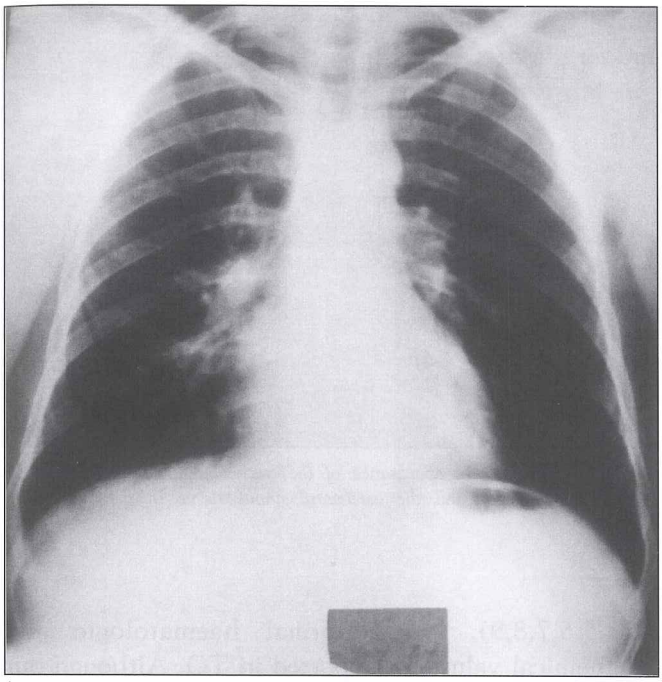

Figure 1. *The posteroanterior chest X-ray of the patient.*

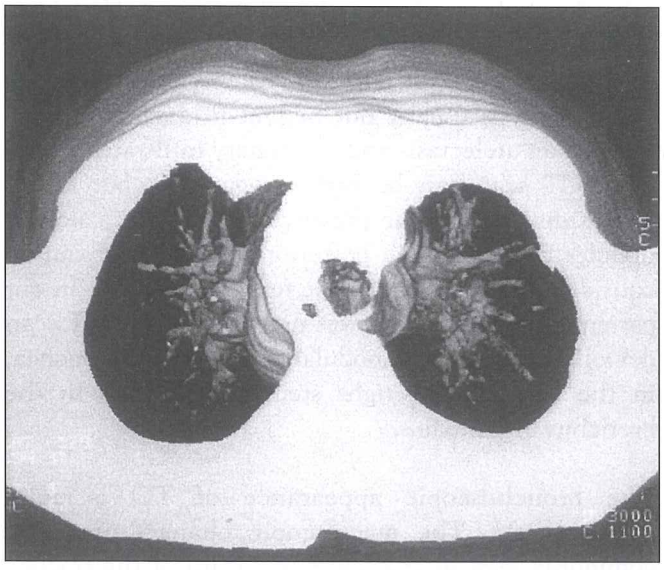

Figure 3. *The three-dimensional helical CT showing distortion and nodular infiltration in the tracheal lumen.*

On physical examination his appearance was that of a cachectic individual. He had palpable lymphadenopathia in the neck. The cervical lymph glands were round shaped, mobile, nontender, and were approximately 10 cm in diameter. The blood pressure was 110/70 mmHg; pulse 90 beats-min^{-1}; temperature 37.2°C; and respiratory rate 16 breaths-min^{-1} with normal breath sounds over both lung fields.

The laboratory findings revealed an erythrocyte sedimentation rate of 85 mm-h^{-1}, a peripheral blood red

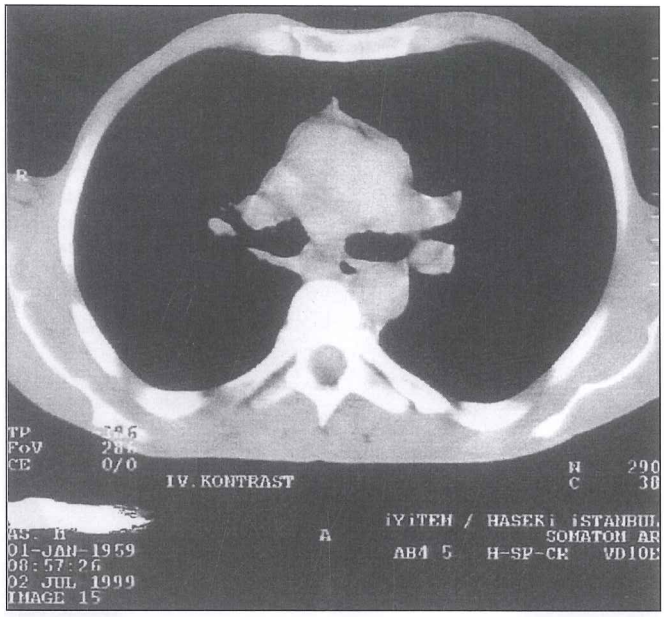

Figure 2. *The chest CT scan showing distortion and nodular densities in the left and right main stem bronchi.*

cell count of 4 530 000/mm^3, a haemoglobin level of 8.5 g/dl, a hematocrit ratio of 28.7, a peripheral blood white cell count of 8 400/mm^3, a platelet count of 442 000, a serum alkaline phosphatase level of 328 U/L, a serum aspartate aminotransferase level of 82 U/L, a serum alanine aminotransferase level of 37 U/L, a total protein level of 7.8 g/dl, a serum albumin level of 2.4 g/dl and a serum globulin level of 4.7 gr/dl. Other biochemical results were within the normal limits.

The tuberculin skin test was negative. The posteroanterior and lateral chest X-ray were normal (Figure 1).

The chest CT scan and a three-dimensional helical CT scan demonstrated multiple nodular densities in the trachea, in the left and right main stem bronchi and in the bronchus intermedius (Figures 2,3). To evaluate the nature of the tracheal lesions, fiberoptic bronchoscopy was performed. At bronchoscopy, the epiglottis, larynx and vocal cords were of normal appearance. Numerous hard, irregular nodules, projecting from the anterior and lateral walls of the trachea, extending through the right and left main stem bronchi and bronchus intermedius were seen (Figure 4). Multiple biopsy specimens were obtained by forceps from the 1/3 distal part of the trachea. Histopathologically, tracheal wall biopsy specimens showed subepithelial osteocartilaginous tissue consistent with TO (Figure 5).

The histopathological examination of the biopsy

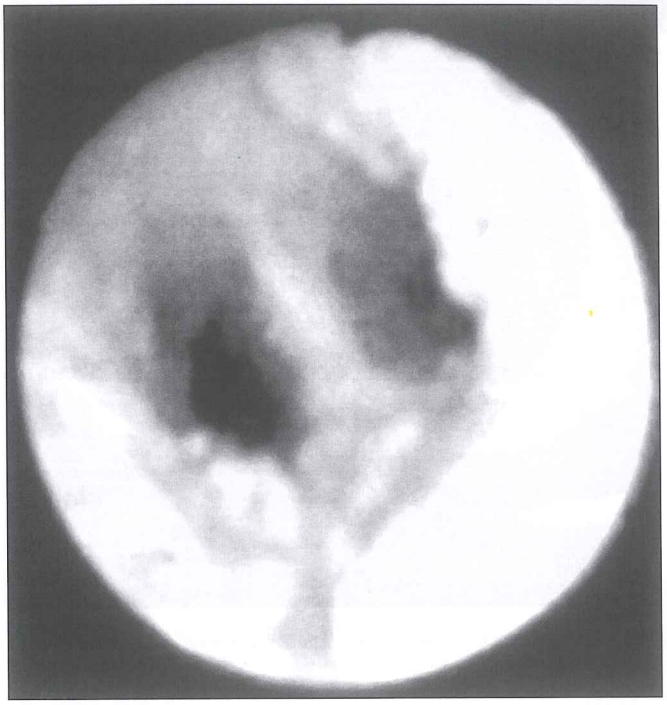

Figure 4. *Fiberoptic bronchoscopic appearance of the lesionse in the trachea, the main carena, the right and left main bronchi.*

specimen obtained by excision from the cervical lymph node led to a diagnosis of the nodular sclerozing subtype of Hodgkin's disease.

Discussion

Tracheobronchopathia osteochondroplastica (TO) is a rare disorder. Until 1974, only 245 cases of TO had been reported in the literature (4). Lundgren et al. reported 9 cases of TO among 2180 bronchoscopies (2) and Ronald et al. 2 cases among approximately 1500 bronchoscopies (5). From 1973 until November 1999, about 19 000 bronchoscopies were performed in our center. This patient is the second TO case diagnosed by bronchoscopic examination in this period. The first TO patient of our center was reported in 1990 (6).

TO is a degenerative disease of the trachea and bronchi occurring in elderly men and is characterized by nodules of cartilage and bone within the submucosa of these structures. These nodules present as polypoid masses that may narrow the airway lumen causing dyspnea and wheezing (2,3,7).

Most patients with TO are asymptomatic throughout their lives. If symptoms occur, they are usually the result of an airway obstruction (2,3,7). The most common symptoms are cough, dsypne, hemoptysis; recurrent respiratory tract infections may also develop

Figure 5. *Histological appearance of the tracheal epithelium showing the hyperplastic changes and the osteocartilaginous tissue in the submucosa (HEX40)*

(2,3,5,6,7,8,9). No abnormal haematologic and biochemical values are reported in TO. Although our patient was asymptomatic, the laboratory findings of anemia and elevated serum alkaline phosphatase and serum alanine aminotransferase levels led us to investigate the patient and a diagnosis of Hodgkin's lymphoma was reached.

Chest X-ray is usually not helpful in diagnosing TO. Sometimes atelectasis and pulmonary infiltrates can be seen. CT scan can be highly suggestive, but is not always diagnostic. The presence of multiple, calcified nodules that spare the posterior tracheal wall can be considered as pathognomonic for TO (3,6,7,9). In our patient, the chest X-ray was normal, but the CT scan demonstrated multiple nodular densities in the trachea, in the left and the right stem bronchi and in the bronchus intermedius.

The bronchoscopic appearance of TO is quite characteristic. The macroscopic changes are most commonly seen in the lower two third of the trachea and in the main bronchi (2,3,10). Tracheobronchial amyloidosis, endobronchial sarcoidosis, papillomatosis and bronchial tumours should be considered in the differential diagnosis (3,5,8). Our bronchoscopic findings were characteristic for TO and this diagnosis was confirmed histopathologically.

Our patient was diagnosed as TO and Hodgkin's disease. To our knowledge, Hodgkin's disease has never previously been reported in association with TO.

TO is a benign disease and there is no specific treatment. Treatment is indicated only in the presence of obstructive symptoms. Treatment includes

cryotherapy, laser excision, external beam irradiation and bronchoscopic removal of the obstructing lesions (3,7). Our patient was asymptomatic so we did not consider administration of any treatment.

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