CASE REPORT

Tracheobronchopathia Osteochondroplastica Associated with Anosmia

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ABSTRACT:

Tracheobronchopathia osteochondroplastica is a rare disorder consisting of multiple hard nodules in the submucosa of trachea and main bronchi. This rare disease remains an under recognized entity due to lack of awareness. One case of such disease reported in this report

KEYWORDS:tracheopathia osteochondroplastica, trachea, tracheopathia osteoplastica, hemoptysis, chronic cough, anosmia.

INTRODUCTION:

Tracheobronchopathia osteochondroplastica is a disease characterized bysubmucosal osseous nodules overlying the cartilaginous rings of the trachea and main bronchi and -on rare occasions- the larynx[1] . In some situation, calcifiednodular densities will protrude into the tracheal lumen[2].However, majority of TPO patients were asymptomaticthroughout their lives. The recurrence or chronic resolvingpneumonia may appear with the deterioration of airway stenosisaccompanied by such symptoms as dyspnoea, hoarseness,persistent, productive cough and haemoptysis [3,4].

CASE PRESENTATION:

A 55-year-old non-smoker single woman was referred for evaluation of a persistent dry cough for more than one year's duration. These symptoms were thought to be caused by asthma and had been treated with various medications with no significant response. Her medical history was significant for persistent nasal congestion, postnasal drips and anosmia with no history of TB contact. Laryngeal polypectomy and nasal septetomy done at 1990 and 1991 respectively with no endotracheal tube complications. Her history of anosmia dated back may years before these operations. Multinodular goiter diagnosed 5 years ago, for which thyroid function was normal and fine needle nodule aspirate showed a benign process.No history of head trauma, neurological disorders, or nutritional disorders. Clinical examination revealed average body built. There was multinodular goiter.

Otonasolaryngological examination showed features of allergic rhinitis. Chest auscultations revealed scattered expiratory rhonchi. Neurological examination was within normal. Pulmonary function tests including a flow volume loop proved normal. Her sputum cytology was negative for both malignant cells and acids fast bacilli stain and culture.

Computed tomography of the chest with 3D multidetecror views showed multiple, different sizes calcified nodules projected from the tracheal wall involving the whole length of the trachea with sparing of the posterior tracheal membrane and extending to both main bronchi (Figure 2and 3). Brain CT was normal. Laboratory tests including a complete blood count, liver and kidney functions were normal. Recent thyroid function proved normal. Fibreoptic bronchoscopy showed large numbers of "rock-like" different sizes nodules extended throughout the trachea and main bronchi and lower lobe bronchi with copius frothy secretions (Figure1). Bronchial wash sample showed small number of mature lymphocytes and was negative both for infective microorganisms and malignant cells. Microscopic examination show non-specific chronic inflammation. Cardiothoracic consultation was arranged and rigid bronchoscopy performed with biopsy samples taken from areas in between these nodules in the trachea and main bronchi and were negative for amyloid stain, but no osseous tissue could be seen in these samples. Serial chest radiographs had been within normal.

Added together, these findings are compatible with the diagnosis of tracheobronchopathia osteochondroplastica. Differential diagnosis of nodular excrescences includes tracheobronchial amyloidosis, endobronchial sarcoidosis, calcificating lesions of tuberculosis, papilomatosis, tracheobronchial calcinosis and relapsing polychondritis. Our patient didn't have any of the systemic symptoms that usually point out to the

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above differential diagnoses nor did she have their histopathological finding on bronchial biopsy.

This patient treated symptomatically with inhaled bronchodilator and inhaled steroid and one session of bronchoscopic argon photocoagulation. She got infrequent acute pulmonary infections for which symptomatic treatment given with prompt results. Serial lung function tests did not show deterioration over 5 years. Her anosmia remain unchanged.

DISCUSSION:

Incidence rates of tracheobronchopathia osteochondroplastica range from 1 in 125 to 1 in 6000 patients, as judged by in vivo bronchoscopy.[5]. The disorder is more common in men and the diagnosis is usually made in the fourth through sixth decades of life. It has been reported that 54% of patients with this disease have chronic cough[6].Most cases are asymptomatic and frequently diagnosed incidentally during intubation, endoscopy or autopsy[7,8].

Chest CT may show multiple sessile submucosal nodules with or without calcification along the cartilaginous portion of trachea. However, CT is not sensitive enough to detect milder forms of the disease. Bronchoscopy is the most definitive diagnostic test. The presence of a few or numerous sessile bony or cartilaginous nodules of the tracheal walls is typical and pathognomonic of tracheobronchopathia osteochondroplastica [9].

Our patient has typical bronchoscopic and radiological findingsof tracheobronchopathia osteochondroplastica. Nonspecific histopathology result is explained by that both fiberoptic and rigid bronchoscpic biopsy pieces were taken from areas in between the nodules as it is very difficult to biopsy the“bone-like”nodules with the conventional biopsy needle.

Although the larynx could be involved in such disease[10,15], our patient did not show typical features of tracheobronchopathia osteochondroplastica in her laryngeal polyp that was excised more than 20 years before her last presentation (unfortunately biopsy result was missed) and this may refer also to the very slow progression of this disease.

Bronchoscopy is useful for ruling out other causes based on findings from CT, such as amyloidosis, sarcoidosis or infection[5]. Although tracheobronchopathia osteochondroplastica is generally a benign disease that progresses very slowly and needs no specific treatment[14], it has been reported to cause respiratory insufficiency from tracheal stenosis, which requires surgical intervention.[11,12].

Management of this rare disease also includes bronchodilators, prompt treatment of pulmonary infections, and bronchoscopic dilatation when indicated. However, in severe cases, bronchoscopic removal of obstructing lesions and surgery has been performed with therapeutic effect[13].

Treatments attempted included cryotherapy, laser excision, external beam irradiation, and bronchoscopic removal of the obstructing lesions. The prognosis is usually favorable. Clinicians should include this disease in the list of differential diagnoses when confronted with symptoms like persistent and often productive cough, haemoptysis, dyspnoea and wheeze; therefore, in patients with chronic unexplained cough, bronchoscopy should be performed [14]because unexpected findings of tracheobronchial tree may be detected like this rare disease.

Presence of anosmia was not reported as an upper respiratory condition associated with tracheobronchopathia osteochondroplastica.

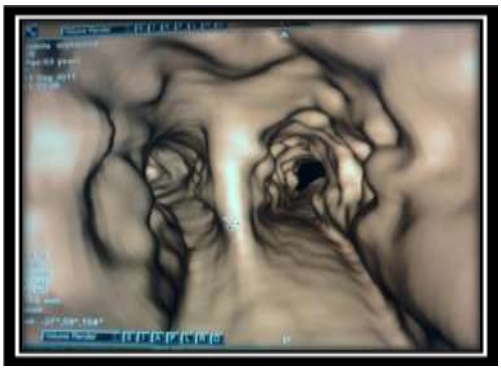

Figure 1:Bronchoscopy showing multiple tracheal nodules involving the carina and main bronchi.

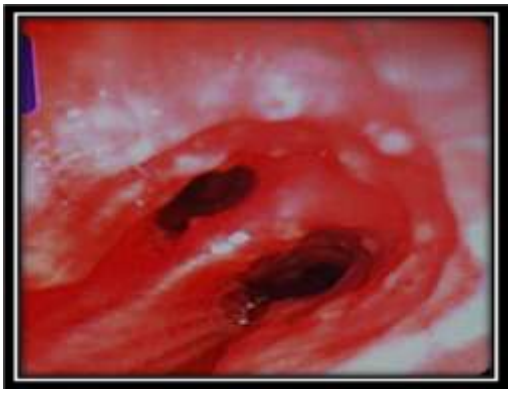

Figure 2: CT chest showing calcified tracheal wall the carina and main bronchi.

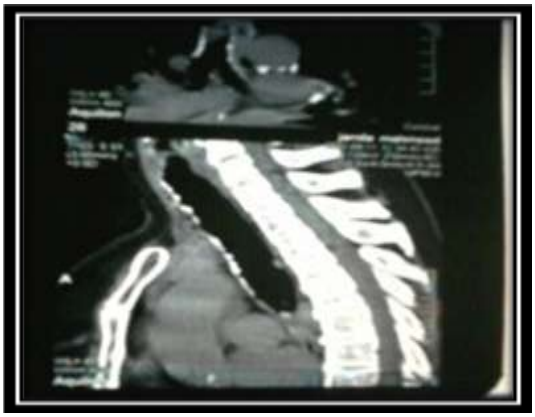

Figure 3: 3D multidedector CT showing nodular structure of the trachea and major bronchi.

CONCLUSION:

Awareness of the tracheobronchopathia osteochondroplasticaas a differential diagnosis to other causes of chronic cough and wheezes is important, to avoid unnecessary intervention or treatment. Anosmia may be one of associated condition for this rare disorder.

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