Case Letters

Tracheobronchopathia osteochondroplastica following laryngeal tuberculosis

Sir,

Tracheobronchopathia osteochondroplastica (TPO) is a rare, idiopathic, benign disorder that is usually asymptomatic and mostly diagnosed incidentally. Characteristic feature is the formation of multiple nodules in the trachea, proximal bronchi, and

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rarely in the larynx with sparing of the posterior membranous trachea.^[1] We describe a patient wherein TPO developed during the course of treatment of vocal cord tuberculosis (TB).

A 66-year-old male presented with hoarseness of voice and fever for 1 month. Indirect laryngoscopy

revealed vocal cord nodularity and biopsy from vocal cord nodule demonstrated necrotizing granulomatous inflammation consistent with TB [Figure 1]. Computed tomography (CT) scan (baseline) showed normal tracheal appearance [Figure 2a]. Patient received four-drug antituberculous treatment. At 6 months despite improvement in fever, hoarseness persisted. Follow-up CT demonstrated nodular tracheal wall with few nodules showing calcification, along with vocal cord nodularity [Figure 2b]. Flexible bronchoscopy demonstrated numerous nodules involving vocal cords, anterolateral wall of the trachea, and bilateral proximal main bronchi with sparing of posterior tracheal wall [Figure 2c]. Biopsy from tracheal nodule had stony hard feeling and revealed focal calcification and ossification confirming the diagnosis of TPO [Figure 3]. The patient was counseled regarding the indolent nature of the condition and advised conservative management.

TPO is a disease of unknown etiology that is usually incidentally diagnosed in sixth and seventh decade of life.^[1] Patients are usually asymptomatic, however, symptomatic ones may present with cough, wheezing, dyspnea, or hemoptysis. The etiology of the disease is not yet identified but association with certain chronic inflammatory conditions, chemical or mechanical irritation, degenerative, or metabolic abnormalities is described. In one case report, diagnosis of TPO and amyloidosis was simultaneously made on postmortem examination.^[2] In another, TPO was diagnosed in a patient chronically infected with *Mycobacterium avium* intracellulare.^[3] Association of TPO with silicosis and IgA deficiency has also been reported.

On systematic review of literature to identify reports describing TPO and TB, two relevant publications were identified. In one patient, TB was diagnosed 21 years before the diagnosis of TPO.^[4] In two cases, TPO was identified along with active TB and persisted after the treatment of TB.^[5,6] In our patient, TPO was diagnosed following the treatment of histopathologically confirmed laryngeal TB, and the diagnoses of both TB and TPO are substantiated with histopathological confirmation of the diagnosis. This is the first report which strongly supports granulomatous etiology such as TB as possible etiology for TPO.

Differential diagnosis of TPO includes tracheobronchial amyloidosis, granulomatosis with polyangiitis, endobronchial sarcoidosis, TB, papillomatosis, and bronchial and tracheal wall tumors. Flexible bronchoscopy is considered diagnostic that reveals characteristic "rock garden" or "cobblestone" appearance due to the nodular projections in the lumen that gives gritty sensation to the passing bronchoscope.^[2] CT is a useful modality that may show calcified airway nodularity. Histopathology is seldom needed for the diagnosis and it is often difficult to obtain a biopsy owing to stony hard nature of the nodules.^[3] Treatment of this disease is usually conservative. In patients with severe airway obstruction, bronchoscopic removal or surgery may be required.

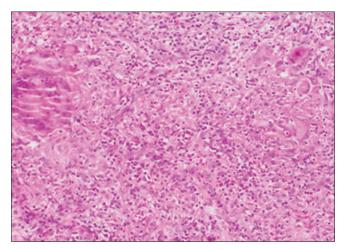


Figure 1: Vocal cord nodule biopsy section shows granulomatous inflammation with Langhans giant cells (H and E, ×40)

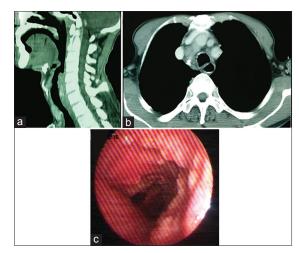


Figure 2: (a) Baseline contrast-enhanced computed tomography neck (sagital section) showing normal appearance of the trachea. (b) Follow-up computed tomography (axial section) showing multiple nodules arising from tracheal wall with focal calcification. (c) Flexible bronchoscopy demonstrating multiple nodules in anterolateral walls of trachea with sparing of posterior tracheal wall

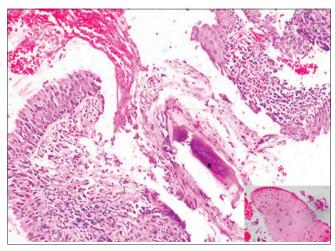


Figure 3: Biopsy from tracheal nodule revealing foci of squamous metaplasia of respiratory epithelium and nonspecific chronic inflammation with a tiny focus of calcification. In the inset, an area of ossification is seen (H and E, ×400)

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Conflicts of interest

There are no conflicts of interest.

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