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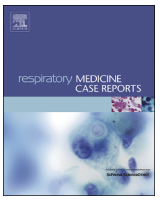

Case Report

A rare case of rapidly progressive Tracheobronchopathia Osteochondroplastica

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ABSTRACT

Tracheobronchopathia Osteochondroplastica (TO) is a rare benign disorder that is seldom progressive. Here, we report a case diagnosed with TO in our hospital. Bronchoscopy revealed multiple cartilaginous and ossifying nodules that are diagnostic for TO. Nodules protruding into the airways were observed as widespread and extended by the repeat bronchoscopy after 2 months of the diagnosis. TO was confirmed with the histopathology of the biopsies from nodules. Then he was referred to an interventional pulmonologist for laser ablation.

1. Introduction

Tracheobronchopathia Osteochondroplastica (TO) is a rare benign disorder of unspecified etiology, characterized by multiple, sessile, submucosal osteocartilagenous nodules protruding into the tracheobronchial lumen [1]. The prevelance of TO is underestimated because it can be incidentally diagnosed due to the lack of symptomatic patients. The clinical manifestations are nonspecific respiratory tract symptoms such as cough, hemoptysis, dyspnea on exertion, and wheeze [2].

The prognosis is usually good but depends on the location and extention of the nodular lesions [3]. In contrast, we present a very rare patient with rapidly progressive TO incidentally detected by bronchoscopy.

2. Case report

A 61-year-old male patient was referred to our reference chest diseases center for further examination after the narrowing of the tracheobronchial tree by multiple nodular calcifications protruding into the lumen was detected in the thoracic CT (Fig. 1) performed at another center. The current smoker (20 cigarettes per day for 10 years) patient had been suffering from a dry chronic cough for three years. The patient's comorbidities were coronary artery disease, hypertension, benign prostatic hyperplasia and umbilical hernia. The medications that the patient used chronically were 4 mg doxazosin, 100 mg acetylsalicylate, and 160 mg valsartan-12.5 mg hydrochlorothiazide daily. The operations he underwent in the past were coronary artery bypass graft and cholecystectomy. Physical examination of the respiratory system was unremarkable. The pulmonary function test revealed FEV1/FVC (Forced Expiratory Volume in 1 s/Forced Vital Capacity) = 72%, that showed non-obstruction.

Fiberoptic bronchoscopy (FOB) demonstrated bright white-colored and hard nodular lesions initiating from the proximal trachea and continuing through the left and right distal main bronchi, but segment bronchi and posterior wall of the tracheobronchial tree were clean (shown in Video). A 30-years experienced bronchoscopist completed the procedure without the requirement for a biopsy

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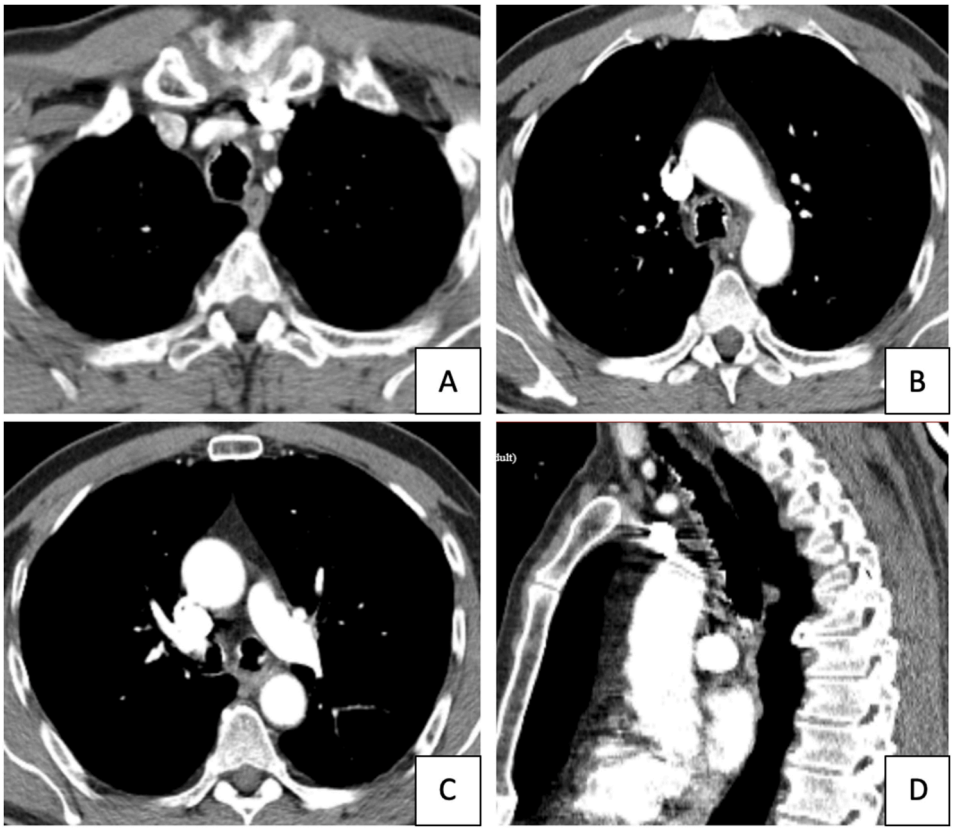

Fig. 1. Thoracic CT with contrast revealed Fig. 1A. Narrowing of proximal trachea with multi-nodular calcifications, Fig. 1B. Narrowing of distal trachea with multi-nodular calcifications, Fig. 1C. Narrowing of proximal main bronchi with multi-nodular calcifications Fig. 1D. Multiple calcific protuberations on the anterior wall of trachea.

to diagnose TO. Antitussives and influenza vaccine were prescribed as symptomatic treatment and the patient was advised to quit smoking.

At the first follow-up after 2 months, the patient had no complaints added to the dry cough. FOB was reperformed for the control and histopathological diagnosis, and it revealed that the endotracheal and endobronchial bright white-colored and hard nodular lesions had become widespread. The middle tracheal lumen was narrowed by these lesions at the rate of 40% (Fig. 2). Histopathology of punch biopsies taken from the hard-content lesions via FOB was reported as lymphoid proliferation, edema, cartilage, osseous, and lipomatous tissues in the bronchial wall (Fig. 3). The patient histopathologically diagnosed with TO was referred to the interventional clinic for laser ablation via rigid bronchoscopy.

3. Discussion

TO was defined firstly by Wilks in an autopsy case as ossific deposits on the antero-lateral cartilaginous walls except the membranous posterior walls of the large airways in 1857 [4]. The detection incidence rate in autopsies is 0.25%–0.3%, and at bronchoscopy is 0.80%–0.01% [5,6]. Males have the dominance compared to females (3:2) [7]. TO is appeared mostly in the range of 4–7 decads [6,8] However, rare pediatric patients have also been reported [9,10] The demographics of the patient is in line with the previous literature.

TO is mostly misdiagnosed with allergy or asthma [11], due to symptom similarity such as cough and wheeze and slow progression [12,13]. Nowadays, due to the increasing number of bronchoscopists aware of the disorder, clinic-radiological misdiagnosis of TO can be reduced. Pathognomonic bronchoscopic view is nodules protruding into the tracheobronchial lumen, which resembles a “rock-garden” [6]. Zhu Y et al. have staged TO by typical bronchoscopic visualization and histopathological exam: Stage I (early stage, mild grade), Stage II (middle stage, moderate grade), and Stage III (late stage, severe grade) [14]. Bronchoscopy is useful in the diagnosis and staging of TO, as well as in the differential diagnosis. In contrast to, endobronchial sarcoidosis, papillomatosis, polychondritis, and amyloidosis involve posterior wall of the trachea [15,16]. While tracheal diffuse calcification can be observed physiologically in the elderly, it can be observed pathologically in tuberculosis, Wegener's graulomatosis, fibroma, and chondrosarcoma [1,17].

However some researchers argue that bronchoscopic visualization alone is not sufficient in the diagnosis of TO and should be verified with histopathologically in symptomatic patients undergoing endobronchial interventions [5,18]. No biopsy was taken from the

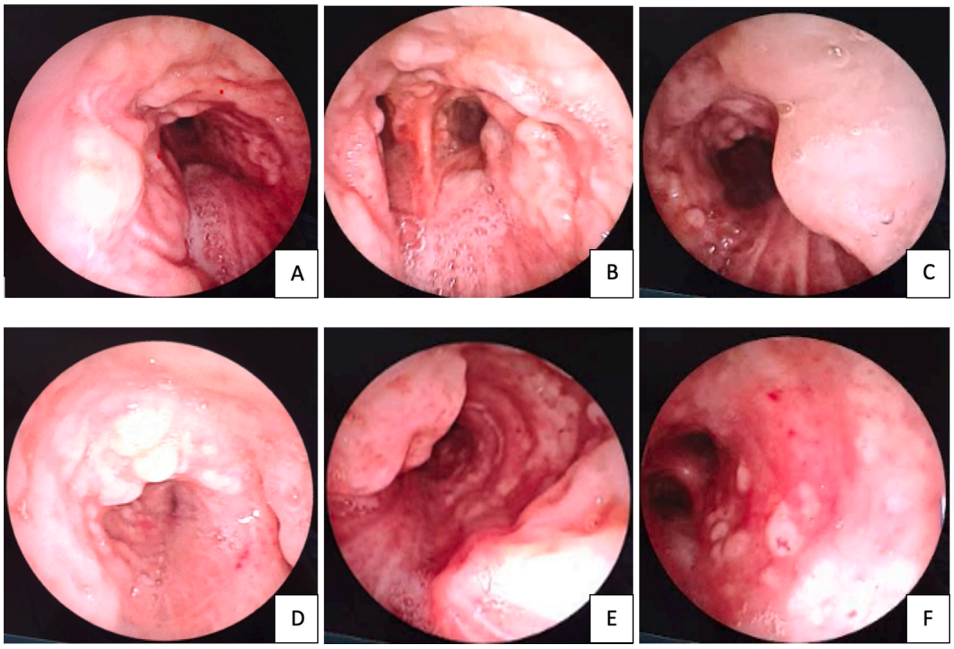

Fig. 2. Bronchoscopic view of the widespread and extended multiple cartilaginous and osseous nodules protruding into Fig. 2A Middle tracheal lumen, Fig. 2B distal tracheal lumen, Fig. 2C proximal right main bronchus lumen, Fig. 2D intermedier bronchus lumen, Fig. 2E proximal left bronchus lumen, Fig. 2F distal left bronchus lumen, called as "rock-garden".

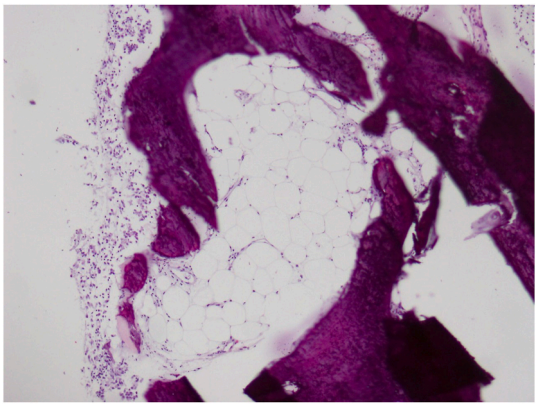

Fig. 3. Lymphoid proliferation, cartilage, osseous, and lipomatous tissues in the bronchial wall (10x10 Hematotoxylin-Eosin).

nodules in the first FOB. When the tracheobronchial lumen was seen as moderately narrowed due to the widespread and extended nodules by the repeat FOB, biopsies were taken from the nodules before referring the patient to the interventional clinic.

Thinned mucosa over the cartilaginous nodule or edematous mucosa with metaplastic epithelium may indicate chronic inflammation [19]. Similarly, in the histopathology exam of our patient, lymphoid proliferation and edema indicating chronic inflammation were observed. Currently, the pathogenesis is poorly understood and factors such as chronic infection, metabolic disorders, chemical and mechanical exposures, Klebsiella ozaenae, squamous metaplasia [9,13,20] and bone morphogenetic protein 2 [21] are considered to act a role.

There are still no guidelines for the treatment and follow-up management of the disorder. Conservative treatment including inhaler steroids, bronchodilators, antibiotics, glucocorticoids [22] and avoidance of the airway irritants, is usually sufficient. Patients with severe airway obstruction require treatment such as bronchoscopic excision of the nodule, laser ablation, surgical resection and radiotherapy [6,23]. Treatment of this patient with advanced airway stenosis was planned as laser ablation.

TO is not associated with the development of malignancy, and any such association is a coincidence [12]. In a previous study, bronchoscopic follow-up in 18 patients disclosed stability in 10 (55%), mild progression in 5 (28%), and major progression in 3 (17%) following the diagnosis [12]. The prognosis of a 39-year-old woman with advanced TO has been reported as stable after 20 years [13]. TO is commonly known as slow progressive. However, two patients with rapidly progressing TO have been reported, one 6 weeks [24] and the other 8 months [25] after diagnosis. TO caused severe airway stenosis in these patients, even resulted in death for the first mentioned patient. Similarly, our case had a rapidly progressive TO that caused moderate airway stenosis 2 months following the diagnosis.

4. Conclusion

TO is a rare benign disorder that can be diagnosed incidentally by the clinic-radiological examinations of suspicious diseases. Bronchoscopy is an effective device in the diagnosis, staging and management of the disease. Since the disorder seldom progresses rapidly, we consider that patients should be followed up closely to treat timely. Such management may be life-saving, especially in patients with dyspnea symptom and physical examination findings.

Declaration of competing interest

The author has no conflicts of interest to declare. I have no financial supports or funding to report.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2023.101853.

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