



Experimental Pneumatic Lithotripter in a Symptomatic Case of Tracheobronchopathia Osteochondroplastica (TO)

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Abstract

Purpose: Tracheobronchopathia Osteochondroplastica is a rare benign disease characterized by calcified sub mucosal nodules in large airways. Treatment is usually palliative with more options (laser ablation or surgical treatment) in severe symptomatic cases. No definitive treatment for this disease has been introduced yet. In current study we report a new technique of endobronchial pneumatic lithotripsy in a symptomatic case of Tracheobronchopathia Osteochondroplastica

Method: Patient was a 28-year-old woman presented with dyspnea to the referral center for pulmonary diseases in Tehran, Iran. She underwent bronchoscopy during which submucosal obstructing nodules were recognized. Pathologic evaluation of biopsies was compatible with diagnosis of Tracheobronchopathia Osteochondroplastica. As patient was reluctant to undergo surgical treatments or laser therapy, we suggested a novel procedure of applying pneumatic lithotripsy device through rigid bronchoscope in order to fragment the obstructing nodules and carefully explained the details to patient. She agreed to undergo this treatment.

Results: During the procedure pneumatic lithotripsy probe reached to obstructed segments through rigid bronchoscope channel and repeated oscillatory movements of probe by air pressure resulted in fragmentation of obstructing sub mucosal nodules. No complications were observed and patient reported symptom improvement within the first week. Spirometry demonstrated improved pattern compared to truncated flow volume loop prior to the intervention.

Conclusion: This current technique may offer a new, safe and minimally invasive treatment option for severe cases of Tracheobronchopathia Osteochondroplastica. Patients with comorbidities in whom surgery is considered high risk would benefit the most from this new method.

Keywords: Tracheobronchopathia Osteochondroplastica; Pneumatic lithotripsy; Symptomatic; Endobronchial; Treatment

Introduction

Tracheobronchopathia Osteochondroplastica (TO) as a rare idiopathic condition usually affects large airways. It was first introduced by Wilked in 1857 [1] and is characterized by multiple sub mucosal osseous or metaplastic cartilaginous nodules protruding into the lumen of trachea [2,3]. As the nodules originate from cartilage, they spare the posterior wall of trachea [3,4]. Although etiology of this disease is unknown, transforming growth factor beta (TGFB) and bone morphogenetic protein (BMP) have been hypothesized to be important factors in pathogenesis of Tracheobronchopathia Osteochondroplastica as they both affect proliferation of bone and cartilage [5]. TO is asymptomatic in most cases and symptoms are nonspecific, hence the disease is believed to be underdiagnosed [6,7]. Large nodules may cause airway obstruction in patients [4] and lead to symptoms like chronic cough, dyspnea and wheezing which are usually misdiagnosed as asthma [8,9]. Definitive diagnosis of Tracheobronchopathia Osteochondroplastica is based on fiber optic bronchoscopy findings as well as histologic features of bronchial biopsies after rule out of differential diagnosis, i.e. Amyloidosis, Sarcoidosis and post-TB airway calcification [3,6]. Treatment is not usually required as a result of the asymptomatic course of disease in most cases [4]. Yet it basically includes conservative managements such as avoiding irritants and maintaining airway humidity

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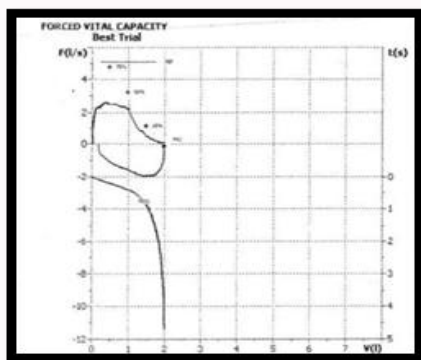


Figure 1: Flow-volume curve of patient before treatment.

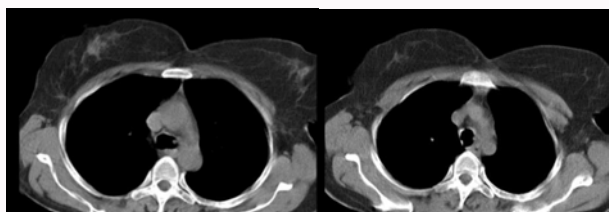


Figure 2: Calcified tracheal nodules in chest CT scan of patient.

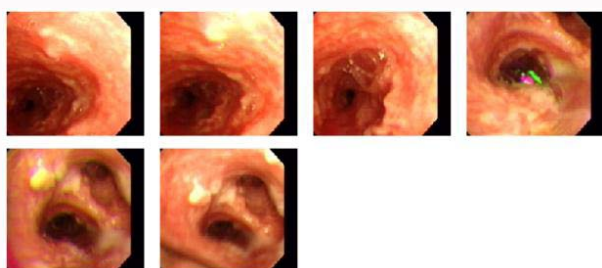


Figure 3: Bronchoscopy view of obstructing nodules in patient.



Figure 4: Pneumatic lithotripsy device and probe.

[4]. In patients with TO impaired clearance of airway secretion and obstruction caused by submucosal nodules may result in recurrent infections and airway colonization by certain microorganisms (such as *Pseudomonas aeruginosa*, *Klebsiella pneumoniae* and *E. coli*) [9,10] which may deteriorate the stenosis and obstruction [4]. Antibiotics are prescribed in case of respiratory infections. Inhaling corticosteroids may be used to suppress underlying inflammation although their role is still controversial [3] Treatment options in symptomatic cases include laser ablation, surgical tracheal segment resection, coring out the lesion, mechanical debulking, stent placement, and tracheoplastic surgeries [9,11-15]. No definite treatment

has been introduced for elimination of obstructing lesions in Tracheobronchopathia Osteochondroplastica yet [6,7]. All available treatments are symptom relieving nonspecific modalities. In current study we experimentally applied a novel interventional technique by using endoscopic pneumatic lithotripsy device (used in urologic medicine for intracorporeal fragmentation of impact calculi), during bronchoscopy for fragmentation of submucosal nodules in a young woman, who was suffering from symptomatic Tracheobronchopathia Osteochondroplastica and observed its effectiveness and possible complications.

Case Presentation and Methods

A 28-year-old Iraqi woman with history of pulmonary tuberculosis who was successfully treated for 9 months was referred to our center at Masih Daneshvari Hospital, a referral center for pulmonary diseases in Tehran, Iran with complaints of dyspnea and exercise intolerance resistant to conventional bronchodilators. Patient denied cigarette smoking and there was no history of prior intubation, allergies or other comorbidities. On physical exam no abnormalities were detected. Vital signs and oxygen saturation were normal. Sputum sample smear and culture were negative for *Mycobacterium tuberculosis*. Pulmonary function tests were performed and flow-volume curve demonstrated appearance of fixed upper airway obstruction with truncated inspiratory and expiratory loops. No changes were observed after bronchodilator administration (Figure 1). Chest CT scan revealed narrowed trachea with calcified nodules causing airway obstruction (Figure 2). Patient underwent fiber optic bronchoscopy for more detailed airway evaluation. Mucosal irregularities and nodules causing fixed airway obstruction at level of trachea and right upper bronchus were observed (Figure 3). Tracheal biopsies were taken. Histopathology finding was consistent with the diagnosis of Tracheobronchopathia Osteochondroplastica. No acid fast bacilli were identified in biopsy specimens. Treatment options were explained to the patient but she was reluctant to undergo surgical procedures or laser ablation, therefore we suggested an experimental method of bronchoscopic lithotripsy for fragmentation of nodules. The procedure details were explained to the patient by assistance of an Arabic translator. Patient agreed and voluntarily underwent the procedure. Informed consent was obtained before intervention.

The procedure

Patient underwent general anesthesia, rigid bronchoscope (Olympus; internal diameter of 8mm) was inserted. Patient's heart rate, oxygen saturation and blood pressure were monitored during the procedure. Pneumatic lithotripsy probe (Swiss LithoClast® -probe 1.0 mm × 605 mm (3.0 Fr) (Figure 4) was carefully applied via bronchoscope channel through zero degree lens and passed to the obstructing segment. Repeated jackhammer like action (maximum frequency of 12 Hz), by oscillatory movement of lithotripsy probe in direct contact with calcified nodules was used for fragmentation of submucosal nodules in obstructing segments and successfully fragmented a number of nodules in obstructed segments of trachea and right upper bronchus (Figure 5). No complications including mucosal injury or perforation due to mechanical nature of the procedure were observed during and after the procedure. Patient was monitored in recovery room for 6 hours and discharged home safely. Patient was carefully followed up for three weeks. She reported symptom improvement during the first week; she could manage everyday tasks without complaints of dyspnea. Post-intervention pulmonary function test conducted after three weeks revealed

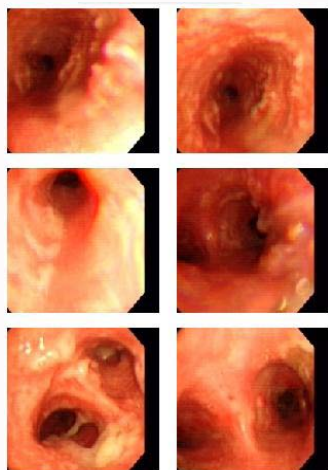


Figure 5: Bronchoscopy view of trachea and main bronchus after treatment.

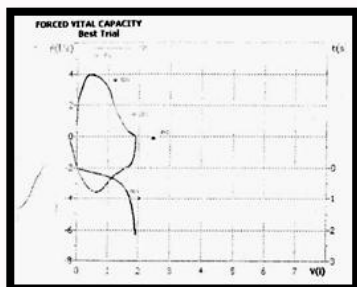


Figure 6: Flow-volume curve three weeks after treatment.

improvement in airway obstruction. Flattening of expiratory curve had also improved significantly in comparison to the pre-procedural curve (Figure 6).

Discussion

We treated a symptomatic case of Tracheobronchopathia Osteochondroplastica using pneumatic lithotripsy probe which successfully fragmented a number of obstructing nodules, improved stenosis and patient's symptom without any complications. As most TO patients are not symptomatic and disease course does not progress in 55% of patients following the diagnosis [12], conservative management is considered in most cases [3,4]. In severely symptomatic patients however, invasive management and surgical treatments are indicated [6,7]. Some of them include: resection of tracheal segment, partial laryngectomy [3] laser removal of nodules or stent placement [4,11] and mechanical debulking [7]. Jabbararjani et al. [15] studied 10 patients with Tracheobronchopathia Osteochondroplastica all whom underwent laser therapy with Nd: YAG laser photo-evaporation (LPE). Coring of lesion with rigid bronchoscope tip was conducted in 4 patients and one patient underwent endobronchial stent placement after 6 years. Six patients reported improvement, 3 had no improvements and one patient died after 6 years. Datau and Musani [11] reported 2 patients successfully treated by sculpting airway to resemble airway original anatomy by combinatory applying laser photocoagulation, rigid bronchoscopy, stent placement and debulking. Although a diversity of treatments has been reported, no specific method has been approved as definite management for TO [7]. On the other

hand, in patients with Tracheobronchopathia Osteochondroplastica, different treatment modalities such as bronchoscopic dilation or stent placement are practically impossible in severe cases due to rigidity of tracheal wall secondary to calcification. Grillo et al. [9] identified linear tracheoplasty as a definitive surgical treatment in severe cases of Tracheobronchopathia Osteochondroplastica with acceptable results. All above mentioned studies reported either palliative or invasive surgical treatments for airway dilation none of them targeted the calcified obstructing nodules as the cause of stenosis, which should be eliminated. In current study however we aimed to fragment these obstructing nodules using a particular endobronchial lithotripsy device. Bronchoscopic Broncholithotripsy was reported in 1994 by Aust et al. [16]. They fragmented and removed a large impact Broncholithiasis, by using an electro-hydraulic lithotripsy probe or a pulsed dye laser fiber attached to flexible bronchoscope in a symptomatic patient, who was too high risk to undergo thoracotomy. We applied pneumatic lithotripsy device via rigid bronchoscope channel for nodule fragmentation in a symptomatic case of TO for the first time. Pneumatic lithotripsy breaks up compact renal calculi by pneumatic jackhammering and probe swing and is widely used as a safe, inexpensive acceptable intra-corporeal endoscopic technique for urinary stones in urologic medicine. No significant difference in terms of complications (mucosal injury or perforation) was identified between pneumatic and conventional laser lithotripsy in studies [17,18]. We observed nodule fragmentation during the procedure by direct probe contact using pneumatic pressure without any attributing complications (perforation or mucosal injury). The stenosis was improved as flow volume curves demonstrate. Given the result, this method may open new prospects for definite minimally invasive non-surgical symptom relieving treatment in severe cases of TO and introduce new era in treatment of Tracheobronchopathia Osteochondroplastica. We suggest that this technique is applied in severe cases that are considered high risk for surgical procedures due to underlying conditions. Possible shortcomings of this method could be mucosal injury or perforation due to mechanical nature of pneumatic lithotripter and possible distal airway obstruction by fragments of nodules after break-up. Although none of above complications was observed during our procedure, future studies enrolling more patients are warranted to correctly evaluate the effectiveness and possible complications of this new technique and to introduce the best device setting for effective destruction of nodules with fewer complications.

Conclusion

Endobronchial pneumatic lithotripsy could effectively fragment the obstructing nodules and improve symptoms and stenosis in severe cases of TO and could be an appropriate alternative to surgery in those who are poor candidates for surgical procedures due to underlying conditions.

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