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Case Report

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Tracheobronchopatia osteochondroplastica case resulting with big airway obstruction

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ABSTRACT

Trakeobronchopatica osteochondroplastica is an unusual non neoplastic abnormality with an unknown etiology. It is characterised with the improvement of one or both of osteoid or cartilaginous milimetric nodules in the submucosa of trachea and bronchial walls. It can be focal or diffuse. Trachea posterior wall conservation is characteristic. Even though most of the cases are asymptomatic, most frequent symptoms are cough, effort dyspnea, wheezing or recurrent respiratory tract infections. Diagnosis is coincidentally determined with otopsy or bronchocopy. Recently, coincidentally diagnosed patients number is increased with the frequent CT usage. A 50 year old male applied our clinic with cough and phlegm whose respiratory function test indicated big airway obstruction. Thorax CT demonstrated a totaly atelectatic right middle lobe medial and lateral segment. Fiberoptic bronchoscopy indicated endobronchial lesions in the right middle lobe's medial and lateral segments. Biopsy proved uncertainty of carcinoid tumor and small cell tumor. In the result of right middle lobectomy, patology informed TO.

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1. Introduction

Tracheobronchopatia osteochondroplastica is a very rare non neoplastic tracheobronchial disorder with an unknown etiology (Leske et al., 2001). It's characterised by bony or cartilaginous(or both) 1-8 mm. nodules in the submucosa of trachea and bronchial walls. It can be focal or diffuse (Ekinci et al., 2012). Trachea posterior wall conservation is characteristic (Martin, 1974). Even though most of the cases are asymptomatic, most

frequent symptoms are cough, effort dyspnea, wheezing or recurrent respiratory tract infections (Leske et al., 2001). Hemoptysis can be rarely seen because of a nodule ulseration or acute infection (Sevim et al., 2002). Diagnosis is coincidentally determined with otopsy or bronchoscopy. Recently, coincidentally diagnosed patients number is increased with the frequent CT usage (Mariotta et al., 1997).

2. Case

50 year old male referred to our clinic with cough and sputum continued for a long time. Nonsmoker patient was followed up with coronary artery disease, HT and asthma. In chest X-Ray, there was no pathology (Fig. 1A and 1B). In respiratory function test, obstructive pattern was detected. (FEV1 was 1970 ml (56%), FVC:2370 ml (69%), FEV1/FVC was 83%.) In Thorax CT right middle lobe lateral segment was atelectatic (Fig. 2). In fiberoptic bronchoscopy right middle lob lateral segment was obstructed with endobronchial lesion, right middle lobe medial segment was obstructed with a pulsating lesion. Biopsy was reported as carcinoid tumor? small cell tumor? (Fig. 3). In PET CT, density in the right hilar region reaching to thoracal pleura suv max was 2.8; dansity in the right hilar 1 cm region anterolateral to the right main bronchus suv max was 3.0. With these results, exploration was planned to the patient. In the exploration, right middle lobe was totally atelectatic and right middle lobectomy was performed. There was no postoperative complication. Right middle lobectomy pathology material was reported as TO. Follow up period was 6 days.

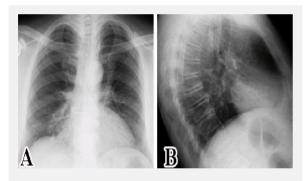


Fig. 1. A. Preoperative PA chest X-Ray; B. Preoperative lateral chest X-Ray.



Fig. 2. Preoperative Thorax CT of the patient.

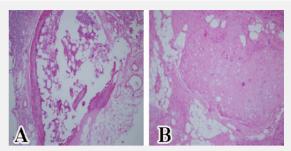


Fig. 3. A. Histopathologic view of bone nodules; B. Histopathologic view of submucosal cartilage.

3. Discussion

Tracheobronchopatia osteochondroplastica is a benign pathology submitted by Rokitansky in 1855 (Mariotta et al., 1997). Estimated prevalance is 0.7%. Typically there is a predisposition for 5. decade and males. Otopsy prevalance is 0.3%. Even though the exact pathology is unknown, endositosis or eksositosis of cartilaginous rings and bony or cartilaginous metaplasia of elastic tissue in the internal elastic fibrose membrane are blamed (Mariotta et al., 1997). Factors like chronic inflamation, infection, trauma, toxic material inhalation can have an important role. Atrophic rhinitis, cold, amiloidosis, silicosis and tbc are also suggested to be related to TO. In our case, there was no positive symptom related to these disorders (Tuncer et al., 2007).

Although patients are generally asymptomatic, dyspnea, cough, hemoptysis, recurrent airway infections and dysphagia are the probable symptoms. Our patient had cough and sputum.TO seems to be with narrowing and irregularity in the effected tracheal and bronchial segments. In Thorax CT, also, thickened tracheal cartilages with protected posterior membranous portions can be seen (Lundgren et al., 1981). Small calcific nodules aligned along trachea protrusing to tracheal lumen are present. This image is more irregular than normal cartilage calcification. In bronchoscopy, white cartilaginous or bony mucosa covered nodules like a bead set in tracheobronchial tree are seen and TO is diagnosed with the nodules seen in bronchoscopy. As in our broncoscopic view ,growing and inosculating nodules can cause obstruction (Tuncer et al., 2007). This view can interfere with benign diseases like amiloidosis, sarcoidosis. Like our case, differential diagnosis can be difficult beside malignities. Our patient's preoperative bronchoscopic biopsy result was uncertain to be small cell lung cancer or carcinoid tumor. Diagnosis was certain with pathologic diagnosis of surgical material. Prognosis is generally good and conservative approach is exhibited. (Willms et al., 2008). To decide the treatment protocol, airway obstruction has first degree importance (Simsek et al., 2006). In patients with symptomatic airway obstruction, both diagnostic and therapeutic surgical exploration and resection can be the exact solution.

Sengül et al. 83

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