Case Report

Classical Hodgkin Lymphoma Presented with Endotracheal and Endobronchial Mass - A Rare Case Report

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A 20-year-old male presented with two months history of progressive dyspnea, night sweat, and dry cough. He was admitted due to fever and productive cough for two weeks. Initial investigation showed only marked leukocytosis. Chest radiograph showed right middle lobe opacity and borderline widening mediastinum. He was diagnosed as community acquired pneumonia and treated with board spectrum antibiotics for 10 days. His symptoms improved but he still had orthopnea. Physical examination revealed "central rhonchi" after force inspiration. CT scan of chest demonstrated that a huge anterior mediastinal mass invaded into the tracheal lumen and extended to the right main bronchus. Bronchoscopic findings were intraluminal lobulated endotracheal mass at right anterolateral portion of lower trachea causing near-total obstruction. The cryoablation with tumor removal was performed. The pathology revealed nodular sclerosis and positive immunohistochemistry for CD15, CD30, and PAX-5, compatible with classical Hodgkin's lymphoma. He was treated with standard chemotherapy with near-complete disease remission after six months follow-up. The present case was a rare presentation of classical Hodgkin lymphoma with endotracheal invasion because most cases presented with external compression rather than intraluminal airway invasion.

Keywords: Hodgkin lymphoma, Endobronchial, Endotracheal, Central rhonchi

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The endobronchial and endotracheal invasions of classical Hodgkin lymphoma (CHL) is very rare. Because of the upper part of lower airway structure, especially the trachea, consists of half-ring cartilage that can be compressed, they are rarely invaded from the anterior portion. The author reported the rare case of endotracheal presentation including clinical feature "central rhonchi". Chest imaging, bronchoscopic, and pathological findings with literature review are included.

Case Report

A 20 year-old male, previously healthy, presented with high grade fever and productive cough for two weeks. He had history of progressive dyspnea, orthopnea, night sweat, and dry cough for two months. Physical examination revealed rhonchi at mid-chest and lower lung zone. He was diagnosed as community-acquired pneumonia. Chest X-ray showed patchy opacity of the right middle lobe and borderline widening mediastinum, that improved after 10 days of antibiotics (Figure 1). However, he still complained of orthopnea. Further physical examination revealed "central rhonchi" (low-pitch wheezing, not stridor) that was intensified by force inspiration and expiration, especially on supine position, suspected of intrathoracic upper airway obstruction. The CT scan of chest revealed a huge anterior mediastinal mass, 14×11×12 cm in size, that invaded the right anterolateral portion of the lower trachea and extended to the right main bronchus. The subcarinal lymphadenopathy compressed the bronchus intermedius causing obstructive pneumonia of the right middle lobe (Figure 2). Laboratory tests showed white blood cell (WBC) 39,430 cell/mm^3 (and became normal after antibiotics), negative sputum acid-fast bacilli (AFB) for three days, serum lactate dehydrogenase (LDH) of 420 U/L (125 to 220), normal beta-human chorionic gonadotropin (B-HCG), and alpha-fetoprotein (AFP). Flexible bronchoscopy was performed and demonstrated

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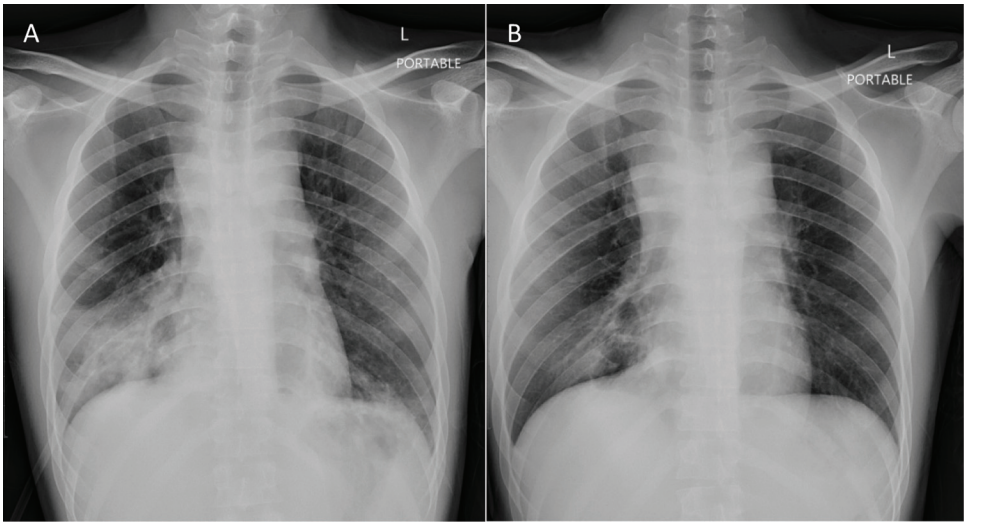

Figure 1. (A) Chest X-ray at admission showed patchy opacity at right middle lobe and (B) after 10 days of board spectrum antibiotics showed substantial improvement. Both also showed borderline widening mediastinum.

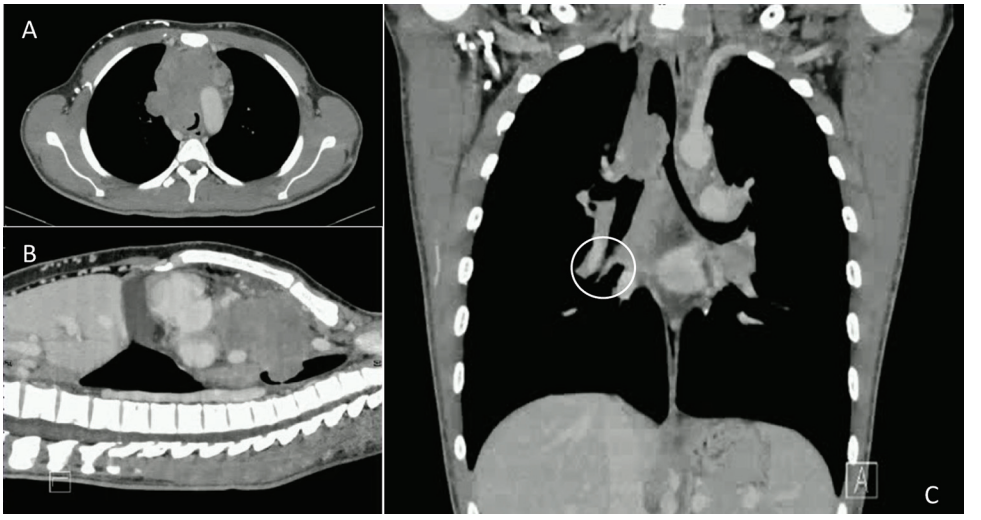

Figure 2. CT Chest X-ray axial view (A) showed huge anterior superior mediastinal mass invaded right anterolateral portion of lower trachea. Sagittal view (B) showed near total obstruction of distal trachea when patient was on supine position. Coronal view (C) showed tumor was extended to right main bronchus, multiple enlarged subcarinal lymph nodes causing narrowing of bronchus intermedius (circle) which may cause obstructive pneumonia of right middle lobe.

intraluminal lobulated endotracheal mass at the right anterolateral portion of the lower trachea that extended to the proximal right main bronchus (Figure 3A). A cryoablation with tumor removal and balloon dilatation was performed (Figure 3B). The pathology revealed nodular sclerosis (Figure 4A), and positive immunohistochemistry (IHC) for CD15, CD30, and PAX-5, compatible with CHL (Figure 4B). He was treated with Adriamycin, Bleomycin, Vinblastine, and Dacarbazine (AVBD regimen) for six cycles and radiotherapy. The patient was in complete disease remission at the 6-month follow-up by repeated computed tomography (CT) scan of the chest and bronchoscopy, which demonstrated no recurrence of the tumor (Figure 3C).

Discussion

The present case is an extremely rare presentation

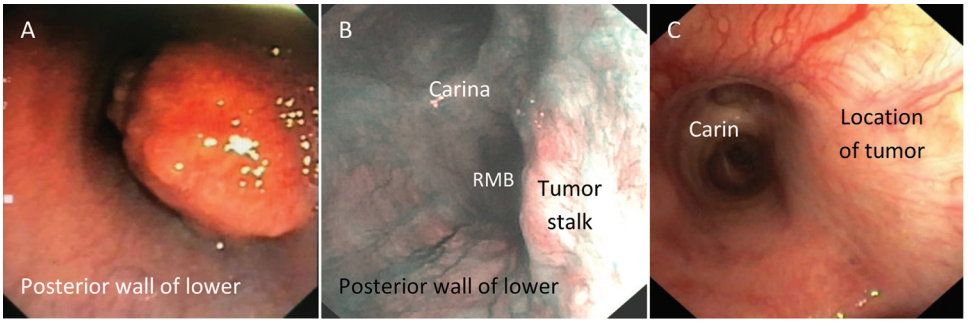

Figure 3. Flexible bronchoscope showed endotracheal mass at right anterolateral portion of lower trachea causing near-total obstruction (A). After cryotherapy with tumor removal in same setting showed a tumor stalk (B) and after complete course of chemotherapy for 6 months showed no recurrent tumor (C).

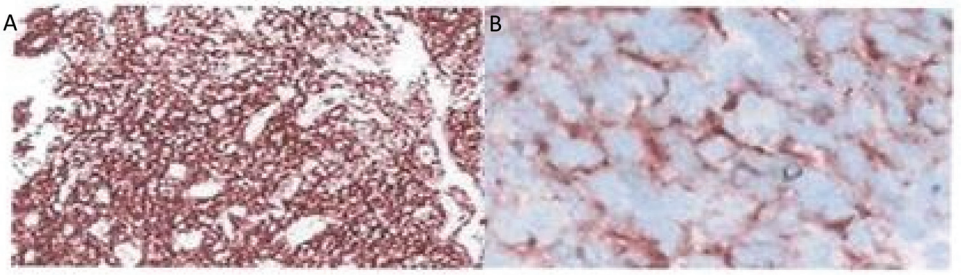

Figure 4. (A) Pathohistological of endotracheal mass showed nodular sclerosis and (B) IHC showed diffuse positive CD30 compatible with CHL.

of CHL because most cases of lymphoma-related pulmonary lesion are usually presented in non-Hodgkin's disease[1]. The largest series of clinicopathological review of 659 cases of CHL showed tracheobronchial invasion are less than 1%[2]. The most common of CHL presentation is mediastinal lymphadenopathy, which can compress airway but rarely invade to lumen[3]. Kiani et al reported in literature review, 26 cases of endobronchial CHL between 1966 and 2003. Of those, only one case involved the trachea[4]. In the past 15 year, only two cases of CHL with tracheal invasion were reported by Davidson[5] and Tanaka et al[6]. The hallmark of diagnosis of large endobronchial lesion are presented of orthopnea (mass effect) and "central rhonchi" (low-pitch wheezing, not stridor) after force inspiration or expiration or on supine position, which implies intrathoracic upper airway obstruction. The present case also had obstructive pneumonia that could blind the central rhonchus due to secretion in large airway causing normal rhonchus in both phase of breathing. There is a case report of endobronchial CHL that presented with non-resolving pneumonia, which may have been caused by obstructive pneumonitis[7]. The other symptoms including hemoptysis or chest pain were not present in our case. This may be due to lack of mucosal bleeding. The CT scan of the chest is helpful to the initial evaluation and to make decision on treatment. Castellino et al reported 203 patients with pulmonary Hodgkin's disease who were initially evaluated by a CT scan of chest. They found that treatment was altered in 9.4% of all patients, 13.8% of patient underwent radiation therapy alone, and 8.2% of patient underwent combined-modality treatment[8]. The present case showed the importance of physical examination, which could lead to further investigation, early diagnosis, and treatment decision. Histopathological finding of CHL most commonly demonstrates nodular sclerosis, which is the same as the present case. While it seems related to primary mediastinal B-cell lymphoma[9], it can be differentiated by specific IHC. Zhang and Aguilera found new IHC, Insulin-like growth factor II messenger RNA binding protein 3 (IMP3) cytoplasmic staining was 98.8% found in Hodgkin-R Reed-Sternberg cells, which was superior to CD15 (65.4%), CD30 (82.7%), PAX5 (84.0%), and MUM1 (85.2%)[10]. Other specific IHC also found in CHL is Epstein-Barr virus (EBV) and

latent membrane protein-1 (LMP-1), which found in about 82% of all case[11]. Treatments of CHL include specific and supportive therapies. Specific treatment in advanced stage CHL is standard AVBD regimen for six cycles. It showed a complete remission rate of 73% with a 63% 5-year progress-free survival and 82% overall survival[12]. The present case also received additional radiotherapy due to residual tumor size of more than 2.5 cm. after chemotherapy, which is according to the standard guideline[13]. Supportive treatment is aimed to correct airway obstruction in case of intraluminal invasion. The most preferable procedure is bronchoscope with cryoablation due to less bleeding complication[14,15].

Conclusion

CHL presenting with tracheal invasion is extremely rare. Some patients may have non-specific symptom, which are difficult to diagnose. The clinical presentations include orthopnea and "central rhonchi". This may imply intrathoracic upper airway obstruction due to anterior mediastinal mass compression to the main airway, which can lead to further investigation. Listening to the patient complaint and doing a detailed physical examination are very useful for early diagnosis and treatment in some important disease like the present case.

What is already known on this topic?

Hodgkin lymphoma can present with huge mediastinal mass that can invade the intra-luminal airway and cause airway obstruction.

What this study adds?

Presence of important physical sign call "central rhonchi" implies early presentation of intrathoracic upper airway obstruction associated with endotracheal or endobronchial lesion is very useful for early diagnosis and treatment.

Conflicts of interest

The author declares no conflict of interest.

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