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Case report

Endobronchial involvement of mantle cell lymphoma: A case report

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ABSTRACT

Mantle cell lymphoma (MCL) is a subtype of B-cell non-Hodgkin's lymphoma. Most cases of MCL have extranodal involvement at the time of the initial diagnosis; however, endobronchial involvement is rare. An 87-year-old man was referred to our hospital because of dyspnea on exertion. A chest CT revealed diffuse irregular wall thickening of the trachea and bilateral bronchi. A bronchoscopy revealed a diffuse irregular surface of the tracheal and bilateral bronchial mucosa and polyposis-like lesions. He was diagnosed as having MCL based on an endobronchial biopsy, and the diagnosis was confirmed using immunohistochemical staining.

1. Introduction

Mantle cell lymphoma (MCL) is a subtype of B-cell non-Hodgkin's lymphoma (NHL). MCL represents about 4%–6% of all lymphomas [1,2]. The diagnosis of MCL is based on biopsy results that show a distinctive morphology, and immunohistochemical staining reveals positivity for CD5 and CD20 as B-cell markers. Furthermore, the overexpression of cyclin D1 protein because of the chromosomal translocation t (11; 14) (q13:q32) of the *CCND1* gene is characteristic of MCL [3]. Most patients are in their 60 s at the time of diagnosis, with a male predominance. Almost all cases are already advanced at the time of diagnosis with stage III or IV diseases and have extranodal involvement, such as the gastrointestinal tract, spleen with splenomegaly, bone marrow, liver, Waldeyer's ring, skin, lacrimal glands and central nervous system [4,5]. However, endobronchial involvement of MCL is rare, and the bronchoscopic findings for MCL are uncertain. We report a case of endobronchial MCL in which a bronchoscopy was performed resulting in a diagnosis based on endobronchial biopsy.

2. Case report

An 87-year-old man was referred to our hospital for dyspnea on exertion. He was a never smoker and had no complications. His ECOG performance status was 1. Upon physical examination, an enlarged left supraclavicular lymph node was detected. A chest CT examination revealed diffuse irregular wall thickening at the trachea and bilateral bronchi (Fig. 1), atelectasis of the right middle lobe because of right middle bronchial wall thickening, an enlarged lymph node in the left supraclavicular and right axillary area, and splenomegaly. Complete blood counts revealed a white blood cell count of 4800/mm^3, a hemoglobin level of 10.8 g/dL, a platelet count of 132,000/mm^3, an absolute neutrophil count of 2822/μL (58.8%), and a lymphocyte count of 989/μL (20.6%). Moreover, large atypical lymphocytes were detected (8%). A bronchoscopy revealed diffuse irregular surface of the tracheal and bilateral bronchial mucosa and polyposis-like lesions (Fig. 2). An endobronchial biopsy was performed for the mucosa of trachea and the inlet of the right upper lobe and middle lobe. All biopsy specimens revealed atypical cells with a high nuclear cytoplasmic ratio and irregular nuclei forming sheet-like lesions. These morphologic findings were compatible with a low grade lymphoma, such as MCL (Fig. 3A and B). On immunohistochemical staining, these atypical cells were found to be positive for CD5, CD20, CD79a, and Cyclin D1 but negative for CD3, CD10, CD21, and CD138 (Fig. 3C and D). Therefore, the diagnosis of MCL was confirmed based on the morphology and immunohistochemical findings especially positivity for CD5 and Cyclin D1. After the diagnosis of MCL, best-supportive care was selected as the treatment of choice, and he was transferred to another hospital.

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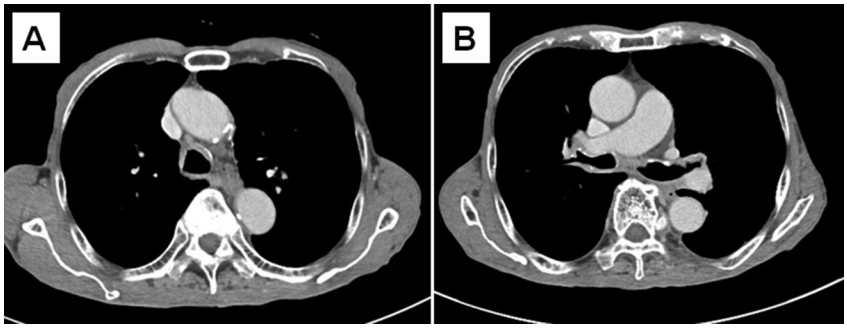

Fig. 1. Chest CT shows diffuse irregular wall thickening at the trachea (A) and bilateral bronchi (B).

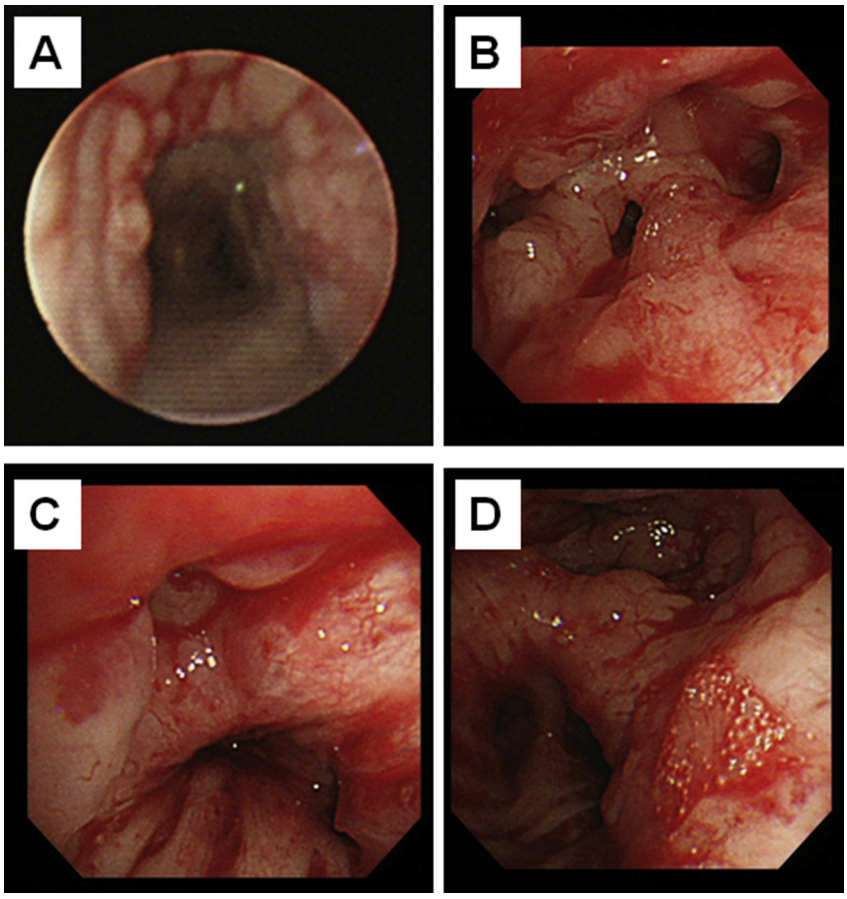

Fig. 2. Bronchoscopy shows diffuse irregular surface of the tracheal and bilateral bronchial mucosa and polyposis-like lesions. (A) Trachea. (B) Bronchus of right upper lobe. (C) Bronchus intermedius. (D) Left secondary carina.

3. Discussion

MCL has a high frequency of extranodal involvement, but endobronchial involvement is rare [4] [5]. To the best of our knowledge, only two cases of MCL with endobronchial involvement have been previously reported [6,7]. A previous report that evaluated extranodal sites of MCL based on CT examinations revealed that pulmonary involvement was present in 7% of the cases at the time of diagnosis, but endobronchial involvement was not detected [8]. A bronchoscopy was not routinely performed at the time of the initial diagnosis of NHL except in one case with abnormal findings on a chest CT examination. Therefore, the correct frequency of endobronchial involvement in patients with NHL including MCL is uncertain. ^{18}F-fluoro-2-deoxyglucose positron emission tomography (FDG-PET) is useful for the detection of extranodal sites of malignant lymphoma. Furthermore, FDG-PET has a high sensitivity in detecting both nodal and extranodal disease in patients with MCL [9]. Although, the present case did not undergo FDG-PET, the usefulness of FDG-PET for the detection of the endobronchial involvement of MCL should be evaluated in greater detail.

Endobronchial lymphoma is categorized into two patterns: diffuse submucosal infiltration (type I) and localized solitary mass (type II) [10,11]. Interestingly, the previous two cases and the present case all exhibited a type I pattern. Several possible mechanisms of endobronchial metastasis have been reported such

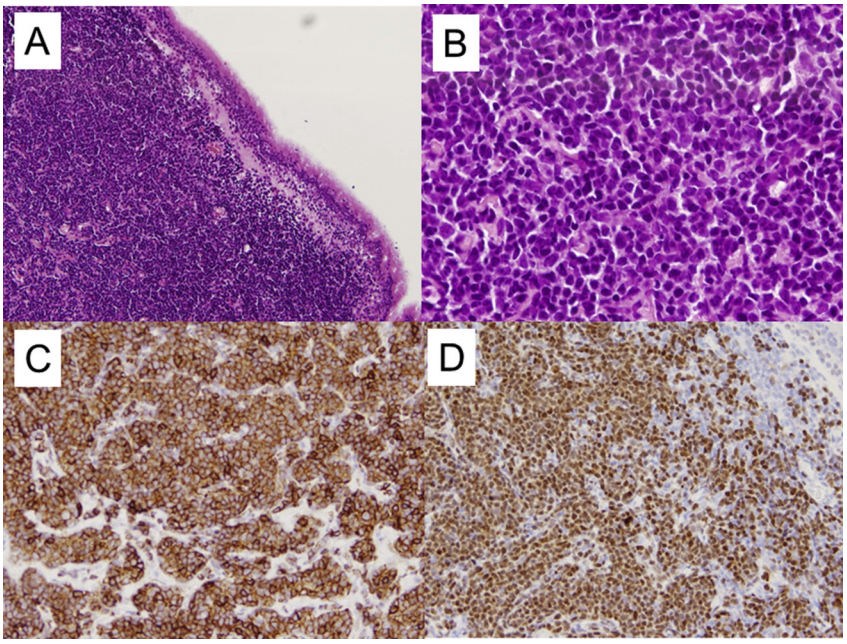

Fig. 3. Pathological findings of endobronchial biopsy. (A) Atypical cells localized at the under of bronchial mucosa (H&E staining, ×100). (B) Atypical cells with a high nuclear cytoplasmic ratio and irregular nuclei formed sheet like lesions (H&E staining, ×400). (C) Immunohistochemical staining revealed positivity for CD5 (×200). (D) Immunohistochemical staining revealed positivity for Cyclin D1 (×200).

as direct bronchial invasion from a parenchymal mass, direct bronchial invasion from a mediastinal mass, lymphatic spread to peribronchial connective tissue, transbronchial aspiration of tumor emboli, and direct hematogenous metastasis [12,13]. The present case did not have pulmonary parenchymal involvement or mediastinal involvement; on the other hand, an enlarged superficial lymph node and large atypical lymphocytes in the peripheral blood were noted. These results suggest that lymphatic and/or hematogenous metastasis may have caused the endobronchial involvement in the present case.

The diagnosis of MCL is made based on a biopsy of lymph nodes, tissue, bone marrow, or blood phenotype [4]. Most cases display a typical morphology of infiltrating small-to medium-sized cells and positive immunohistochemistry results, especially for cyclinD1 [3]. In the present study, a histopathological diagnosis based on an endobronchial biopsy was possible, indicating that endobronchial biopsy could be a useful diagnostic procedure if endobronchial involvement is suspected.

In conclusion, the endobronchial involvement of MCL is rare. Bronchoscopic findings revealed a diffuse irregular surface of the tracheal and bilateral bronchial mucosa and polyposis like lesions. A histopathological diagnosis can be made based on an endobronchial biopsy. Trachea and bronchial wall findings on chest CT should be carefully examined at NHL cases.

Conflicts of interest

The authors have stated explicitly that they do not have any conflicts of interest in connection with this article.

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