Case Report

Tracheomediastinal Fistula Caused by Non-Hodgkin’s Lymphoma

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Non-Hodgkin’s lymphoma involving the tracheobronchial tree is uncommon. A 60-year-old man presented with severe cough for two months. Bronchoscopy disclosed an ulcerative lesion at the lower trachea and carina with anterior tracheomediastinal fistula, and biopsy revealed tracheal wall B-cell lymphoma. After chemo-radiotherapy, the tracheomediastinal fistula healed, but eight months later, tracheal stenosis was diagnosed by bronchoscopy. A self-expansible metallic stent was placed, which successfully maintained the airway.

Keywords: tracheal tumor, tracheal stenosis, fistula, mediastinum

Introduction

Non-Hodgkin’s lymphoma involving the tracheobronchial tree is uncommon. Neither mediastinal lymphoma with tracheal invasion and tracheomediastinal fistula nor primary malignant tumors of the trachea are adequately described. Initial presented symptoms mostly result from tracheobronchial obstruction in the primary tracheal lymphoma. We present a patient with tracheomediastinal fistula that had resulted from non-Hodgkin’s lymphoma and describe its management and complications.

Case Report

A 60-year-old gentleman presented with severe cough and blood-tinged sputum for 2 months. Chest radiograph showed mediastinal widening and an irregular tracheal air column with a right tracheal wall defect. Chest computed tomography (CT) revealed a tracheal tumor with extended mediastinal lymphadenopathy and a suspicious tract of fistula (Fig. 1). Bronchoscopy revealed an ulcerative lesion that had invaded into the anterior mediastinum on the distal trachea, of about 3 cm in length. Histopathologic examination of the biopsy reported a tracheal wall with diffused infiltrate by polygonal cells containing vesicular nuclei, prominent nucleoli and little cytoplasm (Fig. 2). Immunohistochemical staining of leukocyte common antigen (LCA), CD20, and BCL-6 were positive. Thorough investigations, including abdominal CT, bone marrow aspiration, and Gallium scan, showed that the lesion only involved the trachea, surrounding mediastinum, and pulmonary hilum. The final diagnosis of the lesion was diffuse, large B-cell lymphoma.

After 6 cycles of chemotherapy with an R-CHOP regimen and radiation therapy focusing on the involved field, the tracheomediastinal fistula healed, and no further mediastinitis occurred. However, five months after the initial diagnosis, tracheal stenosis was diagnosed by bronchoscopy. A self-expansible metallic stent was placed, which successfully maintained the airway.
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Fig. 1  (A) Chest radiograph shows mediastinal widening and an irregular tracheal air column with a right tracheal wall defect (arrow). (B) Ulcerative lesion on the distal trachea invaded the anterior mediastinum. (C) Enhanced chest computed tomography (CT) shows a tracheomediastinal fistula.

Fig. 2  Tracheal wall with diffused infiltrate by polygonal cells containing vesicular nuclei, prominent nucleoli and little cytoplasm [Hematoxylin-eosin: (A) × 100, (B) × 400]. The immunohistochemical stain of CD20 and leucocyte common antigen (LCA) are positive [Immunostain: (C) × 400, (D) × 400].
radiography or CT.

Discussion

Primary tracheal malignant neoplasms are rare and account for 0.1% of all malignancies. Primary tracheal lymphoma is even rarer, comprising about 0.23% of all tracheal tumors.1,2) The most common presentation of bronchoscopic findings is an obstructive polypoid lesion and a superficial ulcerative mucosal lesion.3) Mediastinal lymphoma is more common, but cases with tracheobronchial involvement are little described. Tse4) and Brinchault5) reported cases with Hodgkin’s disease and tracheobronchial erosion or fistula.

The most common symptoms resulting from tracheal lymphoma are dyspnea, cough, wheezing and hemoptysis. Flexible bronchoscopic biopsy often fails to obtain an adequate amount of tissue, which is often nonspecific, even when there is gross evidence of an endobronchial tumor.3,4) In our case, we used rigid bronchoscopy to obtain an adequate specimen for tissue diagnosis and prevent airway obstruction or asphyxia from blood clots during the procedure.

Treatment strategies including surgery, chemotherapy, and radiotherapy are usually used alone or in combination.1) Surgical interventions used for releasing the critical airway obstruction include laser, photodynamic therapy, rigid bronchoscopy, and tracheal-bronchial stent placement.3)

Tracheal lymphoma complicated with a tracheomediastinal fistula is extremely rare; only three cases including our case have been presented in the literature.4,6) Mediastinal lymphoma is more common, but tracheomediastinal fistula is uncommon. Tse and colleagues used a pericardial patch and omental patch to repair the defect and a stent was used into the left main bronchus.4) Ranes and colleagues demonstrated metallic stenting in the right and left main bronchus for preventing further mediastinitis, and the tracheomediastinal fistula healed at the 13th week.6) In our case, the tracheomediastinal fistula healed during tumor-specific therapies without mediastinitis.

The outcomes of the primary tracheal lymphoma and mediastinal lymphoma with tracheobronchial involvement reported in the literature were relatively favorable after proper treatment. In our presenting case, there was no recurrent disease detected during 7 years of surveillance.

Conclusion

Non-Hodgkin’s lymphoma involving the tracheobronchial tree with tracheomediastinal fistula is rarely reported. If it is not complicated with mediastinitis, as in our presented case, the mediastinal fistula could heal after proper tumor-specific therapy. Late complications, such as stenosis, could be managed with tracheal dilatation and stenting.

Disclosure Statement

The authors have no conflicts of interest.

References