Case Report

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A case of primary B-cell thyroid lymphoma with extensive tracheal invasion

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Abstract:

Primary thyroid lymphoma (PTL) is a rare malignancy originating in the thyroid gland. It can manifest as a rapidly growing mass within the mediastinum, potentially compressing the trachea and causing respiratory difficulties. Diagnosing PTL can be challenging due to its rarity, impacting the available treatment options. The choice of surgical intervention depends on the location, depth, and extent of the invasion. If the tumor has penetrated the tracheal mucosa, then complete removal is required, but this may result in serious complications. Here, we report a distinctive case of PTL involving the trachea, and further complicated by severe airway obstruction and perforation.

Keywords:

Bronchoscopy, chest surgery, imaging

Introduction

Primary thyroid lymphoma (PTL) is an uncommon entity defined as lymphoma involving only the thyroid gland or the thyroid gland and surrounding lymph nodes, without contiguity or metastasis of other organs at the time of diagnosis.[1] Involvement of the trachea by PTL is a rare issue that can mimic asthma or chronic obstructive pulmonary disease. Earlier case reports have described a tracheal or endobronchial mass as a clinical symptom.[2,3] Because it is rare, diagnosis can be difficult and may impact treatment.[4] We describe a rare case of PTL involving the trachea complicated by significant airway obstruction.

Case Report

A 74-year-old man was admitted with a 4-week history of dyspnea and stridor. Past medical history revealed hypertension and diabetes mellitus. He was an active smoker with a 60-pack-year smoking history. Chest X-ray revealed upper mediastinal enlargement. A thyroid mass measuring 6×3 cm and milimetric mediastinal

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and hilar lymph nodes were seen on computed tomography (CT) of thorax [Fig. 1a]. The tumor invaded the trachea at 13 mm length, 5 mm deep, narrowing the tracheal lumen and displacing the trachea to the right. Initially, thyroid carcinoma or lung cancer was suspected due to a left paratracheal lesion. Rigid dilatation was not considered as it was deemed disadvantageous, as it would apply external pressure. The patient was offered a tracheostomy but declined the procedure. A tracheal stent was not implanted due to the rapid change in tracheal diameter with chemotherapy and corticoteroid. A bronchoscopy was performed, showing a white ulcerous bleeding plaque over the tracheal mucosa [Fig. 1b]. Several biopsies were taken from the tumor. Histopathology revealed a diffuse large B-cell non-Hodgkin's lymphoma (DLBCL). For this patient, chest CT discovered a homogeneous tumor that had moved the thyroid gland laterally and could not be separated from the thyroid gland. These results are compatible with nodal lymphoma. On immunohistochemistry, the cells were positive for CD20 (×200). BCL6 (×400) was suboptimal and MUM1 (×400) was negative. Thus, PTL was considered with these clinical and radiological findings and supporting immunohistochemical results. These results were consistent with nodal lymphoma. Thus, the R-CHOP protocol (rituximab, cyclophosphamide, doxorubicin, vincristine, and prednisone) was initiated. The tumor decreased rapidly following the intended six cycles of the R-CHOP protocol, and stridor improved. Intrathecal methotrexate was also used to stop lymphoma cells from entering the cerebrospinal fluid around the spine and brain. The patient's condition was excellent, with a rapid and effective reduction in tumor size observed, leading to a large defect in the anterior tracheal wall due to tumor necrosis, as seen in the thorax CT after 1 month [Fig. 2a]. There was no significant pneumomediastinum or paratracheal air observed. Additionally, a tracheal stent was not implanted to avoid stent migration. The patient stabilized without respiratory complaints or interventions. After 6 months, a positron emission tomography/computed tomography scan (PET-CT) revealed increased fluorodeoxyglucose uptake in the milimetric right paratracheal, hilar lymph nodes with SUV_{max} 1.8–2.2, and bilateral ground-glass opacity in the lungs, but the mass in the left thyroid lobe had disappeared completely, and there was no involvement shown in the PET-CT. The tracheal defect closed spontaneously with the help of surrounding tissues and fibrosis, and the trachea was patent [Fig. 2b]. There was no respiratory impairment during this time. The follow-up at 18 months was uneventful and the patient was healthy.

Discussion

Thyroid lymphomas are rare tumors and should be suspected in patients with no tender, painless, rapidly expanding lumps throughout the entire gland without nodular and other pathognomonic symptoms. Most PTLs are non-Hodgkin's lymphomas. Similar to our case, 50%–80% of PTLs are DLBCLs and 20%–30% are mucosa-associated lymphoid tissue (MALT) lymphomas, primarily extranodal marginal forms.[4] Other uncommon subtypes involve follicular lymphoma (12%), Hodgkin's disease (7%), small lymphocytic lymphoma (4%), and Burkitt's lymphoma (4%).[4] Most instances of the disease are in elderly women with a rapidly expanding thyroid tumor and symptoms of airway obstruction.[1,5] However, in our patient, thyroid lymphoma invaded the trachea and caused significant stridor. Since it is an unusual entity, diagnosis can be difficult and may impact prognosis. The importance of diagnosing PTL lies in the fact that this disease is highly curable if detected early, without the need for invasive surgery. The surgical strategy depends on the location, depth, and scope of the invasion. Sleeve resection is a viable oncological procedure that can be done in one stage, but it carries a danger of serious consequences from anastomotic collapse. The diagnosis was made using fiberoptic bronchoscopy. A biopsy was performed from the tumor in the anterior tracheal wall at bronchoscopy, and the histopathologic diagnosis was B-cell NHL lymphoma. The current standard of medical therapy is R-CHOP. The cure rate exceeds 50% with this treatment.[6] In the literature, surgical access has been shown to have no influence on survival in patients with PTL, although it is crucial for retrieving tissue for diagnosis when palliation is required for large obstructing tumors.[7] The optimal management of PTL is yet unknown. Surgery and radiotherapy (RT) were the primary treatments for PTL in the past. Nevertheless, these treatments have poor success and high recurrence rates, and PTL is known to be sensitive to chemotherapy and radiation.[8] Surgery is not very helpful for PTL, except for palliative therapies to ease tumor-induced airway obstructions. Consequently, surgery is now infrequently utilized, and the best practice for PTL is to use both RT and chemotherapy.[9] The prognosis depends on the stage of the dis-

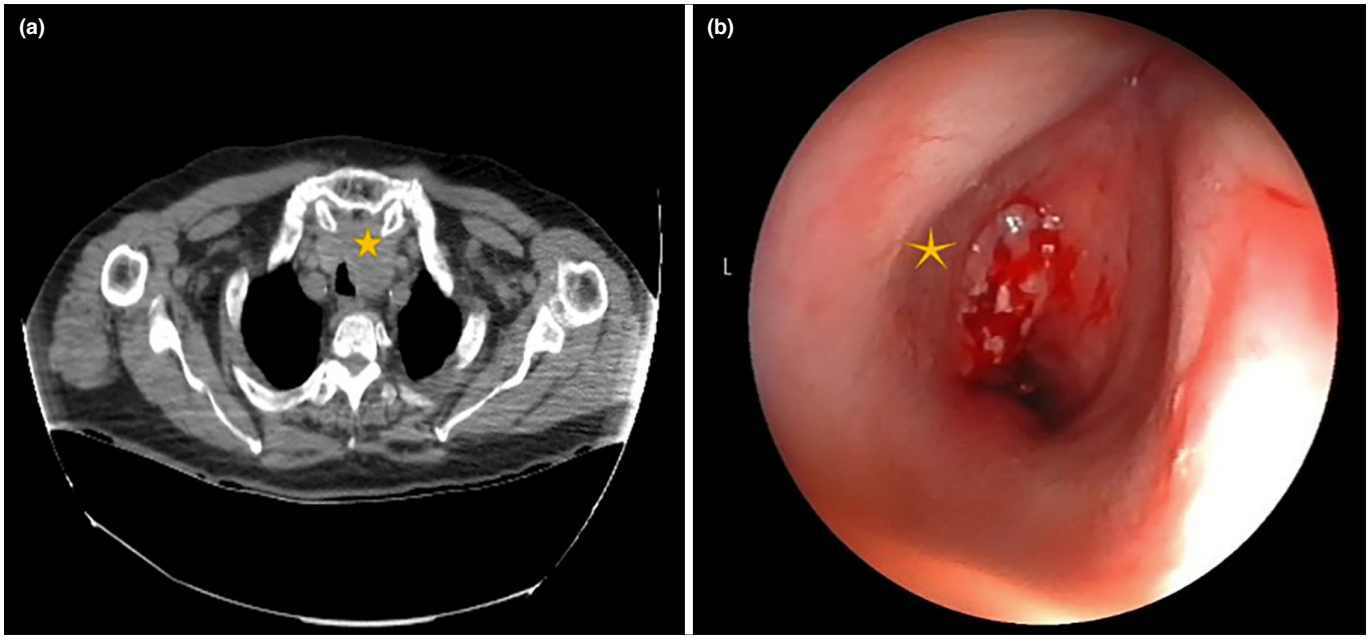

Figure 1: (a) Axial computed tomography section of the thorax showing a large anterior mediastinal mass surrounding the trachea (yellow star). (b) Fiberoptic bronchocopy showing invasion of the left tracheal wall just below the vocal cords (yellow star)

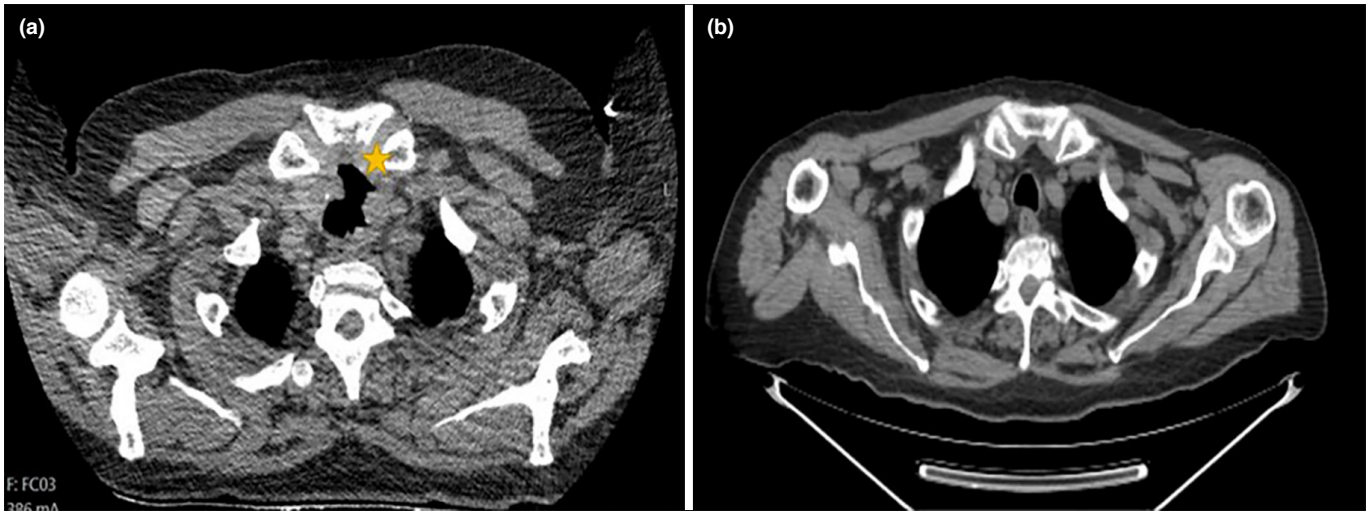

Figure 2: (a) Following adjuvant therapy, a defect was occurred on the anterior tracheal wall (yellow star). (b) Following 6 months of therapy, the defect had spontaneously closed and the trachea became patent

ease and the histological categorization of the tumor.[10] MALT lymphomas, because of their slower progress and more effective response to treatment, have a better prognosis than DLBCL. The 5-year survival rate in patients with intrathyroidal disease is 90% and declines to 35% in patients with extra thyroidal invasion.[10]

PTL with tracheal involvement is an extremely uncommon tumor. Cases of PTL are more symptomatic and have early presentation because of progressive dyspnea. The diagnosis was supported by biopsy and histopathological analysis and had a beneficial response to chemotherapy. Moreover, early detection of tracheal involvement by lymphoma should alert clinicians to consider the probability of tracheal injury during treatment of this potentially serious clinical issue. Our patient had no other significant comorbidities and responded well to multimodal treatment. Preemptive airway management is a viable option in treating invasive PTL with tracheal perforation.

Informed Consent
Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Authorship Contributions
Concept – Y.A.K.; Design – Y.A.K.; Supervision – Y.A.K., Y.Y.; Materials – Y.A.K.; Data collection &/or processing – Y.A.K., F.Y.; Analysis and/or interpretation – Y.A.K.; Literature search – Y.A.K.; Writing – Y.A.K.; Critical review – Y.A.K., Y.Y.

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There are no conflicts of interest.

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