An Unusual Presentation of Endobronchial Hodgkin's Lymphoma

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ABSTRACT
Endobronchial involvement of Hodgkin's lymphoma is a rare presentation of the disease. However, it needs to be considered in patients presenting with non-resolving pneumonia in the setting of Hodgkin's disease. In such cases, clinicians need to ensure adequate and multiple biopsies as patients may have co-existent pulmonary infection and disease involvement. A 16 years old patient reported with a history of relapsed Hodgkin's disease, with bilateral pulmonary infiltrates, that failed to resolve after empiric antibiotic therapy. Positron Emission Tomography (PET) scan was performed demonstrating equivocal uptake in left upper lobe. Bronchoscopy revealed a necrotic endobronchial mass. Initial biopsy only revealed necrotic inflammatory debris, however, since the patient continued to lose weight, repeat biopsy was performed which demonstrated CD 30 positive Hodgkin's lymphoma cells.

Key Words: Hodgkin's lymphoma. Non-resolving pneumonia. Endobronchial mass. PET scan. CD 30 positive cell.

INTRODUCTION
Hodgkin's lymphoma will commonly involve the lymph nodes with upto 85% of patients having intrathoracic lymph node involvement. Mediastinal adenopathy will commonly involve the paratracheal and subcarinal lymph nodes. Endobronchial Hodgkin's lymphoma, on the other hand is very rare and can present as non-resolving pneumonia and even be mistreated as tuberculosis in countries endemic for tuberculosis. It can also be confused clinically with small cell carcinoma and necrotizing vasculitides with pulmonary involvement.

The authors reported a case of a patient with non-resolving pneumonia and a history of Hodgkin's lymphoma who had failed multiple courses of antibiotic therapy and was referred to the pulmonary service for further management.

CASE REPORT
A 16 years old female patient first presented in February 2009 with cervical adenopathy and a mediastinal mass on chest X-ray. Cervical biopsy revealed Hodgkin's lymphoma (nodular sclerosis type). After an initial success with treatment, using radiation and chemotherapy, she subsequently relapsed (stage II) post-third line chemotherapy and was referred to the pulmonary clinic after being treated for bilateral pulmonary infiltrates with broad spectrum antibiotics for worsening cough and progressive lung infiltrates on chest X-ray (Figure 1). She had also lost 25% of her body weight in 4 months.

Positron Emission Tomography (PET) scan was done in January 2011 (Figure 2) which showed mild FDG uptake in bilateral para-tracheal soft tissue densities with maximum SUV of 2.7. An FDG avid right sided mass showed a central cold area consistent with necrosis. Distinct FDG avid right axillary lymph nodes were also noted with maximum SUV of 5.0. Additionally, heterogeneous FDG avid parenchymal changes were noted in the left upper lobe region.

Bronchoscopy in January 2011 revealed purulent secretions from the right upper lobe sub segments; therefore, bronchoalveolar lavage was performed in the right anterior and posterior segments. A whitish endobronchial mass was also noted at the orifice of the left upper lobe (Figure 3). Endobronchial biopsy specimens were taken of the mass. Histopathology results reported necrotic inflammatory debris and fibrin.

In view of failure to improve despite multiple courses of wide spectrum antibiotics, repeat bronchoscopy was performed in February 2011, which showed a new whitish plaque like mass on the distal tracheal wall. The previously seen left upper lobe endobronchial mass was also visualized again. Multiple endobronchial biopsies were performed of the original left upper lobe mass. On this occasion, histopathology report revealed that most of the tissue showed necrosis, however, a few small fragments showed large CD-30 positive tumor cells (Reed-Sternberg cells and variants) in a mixed inflammatory background making it a case of classic Hodgkin's lymphoma.

This new finding upstaged her disease from II/E to stage-IV and led to a change of chemotherapy and stoppage of antimicrobials. Subsequently, the patient was noted in follow-up to have an improvement in her symptoms as well as weight gain.
DISCUSSION

Hodgkin’s lymphoma commonly involves hilar and mediastinal lymph nodes, with Computerized Tomography (CT) revealing mediastinal involvement in up to 85% of cases.\(^1\,\(^2\)\) Although this adenopathy can commonly cause tracheobronchial compression in children, this is much less common in adults or teenagers since the lymph nodes are soft relative to the cartilaginous skeleton of the main airways. Endobronchial involvement of Hodgkin’s lymphoma has been reported in up to 14% of autopsies done,\(^2\) but symptomatic endobronchial involvement in living patients remains very rare. Kiani and colleagues reviewed the literature and found a total of 26 patients with endobronchial presentation of Hodgkin’s lymphoma in the English literature.\(^3\) These patients commonly presented with symptoms of cough, with half of cases also reporting hemoptysis. Bronchoscopically, the most common finding was one of an obstructing polypoid lesion. Another less common presentation that has been reported is in the form of a superficially ulcerated mucosal plaque like infiltrate.\(^3\) Solomonov et al.\(^4\) also looked at endobronchial involvement in non-Hodgkin’s lymphoma and noted that in a series of 441 patients, 8 cases of endobronchial involvement were observed. All patients in their series had radiological signs of lobar atelectasis. On bronchoscopy, half of these patients had complete obstruction of lobar bronchi. The authors did not mention if any of these masses were necrotic. Another case report by Arguder and colleagues reported bilateral endobronchial diffuse nodular lesions in a 65-year old man with non-Hodgkin’s lymphoma.\(^5\) In this respect, this case is unique in that bronchoscopically the present tumor was necrotic and did not have the typical polypoid appearance that is commonly reported. Another unusual aspect of this case was the presence of multiple endobronchial lesions, although Kiani et al. did note three previously reported cases of multiple endobronchial lesions in their review.\(^3\) The presence of multiple necrotic masses may be an indicator of a rapidly growing tumor, or super added infection.

The presence of a non-resolving infiltrates on radiography warrants the suspicion of a possible malignancy. While pulmonary involvement in Hodgkin’s disease can present as a non-resolving pneumonia, the clinical clues of dyspnea, stridor and wheeze point to a possible endobronchial involvement. A bronchoscopy in such a situation can be valuable for diagnosis, and can aid in staging of the disease. Endobronchial Hodgkin’s lymphoma when diagnosed early and treated appropriately, carries a good prognosis with the possibility of a complete cure.\(^6\)

Due to the dramatic implications in treatment and prognosis, endobronchial Hodgkin’s lymphoma, despite its uncommon occurrence, should be considered in the differential of endobronchial masses, especially in a relatively young patient with cough, hemoptysis, atelectasis, and hilar or mediastinal lymphadenopathy. Tumor debulking or stent placement for palliative treatment of life-threatening airway obstruction may be required prior to or as an initial adjunct to tumor-specific therapy.\(^3\) It is important to remember that tumor debulking needs to be carried out by trained interventional pulmonologists with experience in rigid bronchoscopy.

With proper multidisciplinary care such patients can expect a good response to therapy. Solomonov et al. observed that the prognosis of patients with isolated endobronchial lymphoma is not worse than other local presentations of lymphoma.\(^4\) Donlan et al. presented a similar case with endobronchial Hodgkin’s lymphoma where the patient remained in complete remission for 24 months after chemotherapy and repeat bronchoscopy showed complete resolution of the endobronchial lesions.\(^7\)

In conclusion, this case highlights the importance of early bronchoscopy and taking multiple biopsies, as these patients may have a good response to chemotherapy.

REFERENCES

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