



THE ANNALS OF THORACIC SURGERY



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Ann Thorac Surg 2003;76:940-942

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Polypoid Endobronchial Hodgkin Lymphoma With an Initial Response to Photodynamic Therapy

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Endobronchial presentation of Hodgkin lymphoma is rare and can be confused clinically and radiographically with pulmonary small cell carcinoma. We present a case of an obstructive endobronchial presentation of nodular sclerosing classic Hodgkin lymphoma, initially misdiagnosed as small cell carcinoma, with endobronchial vasculitis and associated hemoptysis. Photodynamic therapy relieved the obstruction before induction of tumor-specific therapy. This case demonstrates the successful use of photodynamic therapy in obstructive endobronchial Hodgkin lymphoma.

(Ann Thorac Surg 2003;76:940-2)

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Hodgkin lymphoma in newly diagnosed patients most commonly presents with mediastinal disease. In contrast, pulmonary involvement is uncommon, with endobronchial involvement being a particular rarity, manifesting characteristically in patients with established disease. The estimated frequency of pulmonary disease on the basis of radiographic studies is 8% to 11.6% [1]. The endobronchial lesions may take the form of a plaque-like mucosal lesion or as a polypoid mass projecting into the lumen, the latter mimicking small cell carcinoma [2-4]. We report a case of endobronchial presentation of Hodgkin lymphoma with endobronchial necrotizing vasculitis. This case demonstrates the successful use of photodynamic therapy (PDT) for endobronchial obstruction as a result of Hodgkin lymphoma.

A 44-year-old white man, a 28-year smoker, presented with a 6-week history of fatigue, dyspnea, hemoptysis, and weight loss. Chest computed tomography scan further revealed a right pleural effusion and a 9.0 cm × 6.0 cm mediastinal mass encompassing the right paratracheal, right hilar, and subcarinal region. Magnetic resonance imaging studies did not disclose any evidence of extrathoracic disease. Bronchoscopy revealed a polypoid



Fig 1. After Photofrin administration 48 hours earlier, bronchoscopy was performed, revealing an obstructing endobronchial lesion.

lesion involving the carina and the opening to the right mainstem bronchus, with extension into the left mainstem bronchus (Fig 1). A biopsy demonstrated a crushed small cell infiltrate initially interpreted as small cell carcinoma. The patient was urgently transferred to the Ohio State University's James Cancer Center to undergo PDT for presumed small cell carcinoma. The patient received 2 mg of the hematoporphyrin derivative Photofrin (Axcan Scandipharm Inc, Birmingham, AL) intravenously 48 hours before the first light therapy. Autofluorescence bronchoscopy highlighted the endobronchial mass. Light therapy was conducted using a 1.5-cm quartz diffusing fiber to deliver 250 J of light energy. Mechanical debridement was accomplished through rigid bronchoscopy. After the first debridement, 200 J of light energy was given. The final debridement was performed 96 hours after the first injection, with resultant patency of the mainstem bronchus (Fig 2).

The first biopsy specimen histologically demonstrated a vasculitis and a reactive lymphocytic infiltrate. The second biopsy specimen, although demonstrating vasculitis, showed infiltration by small lymphocytes in concert with scattered large atypical monocytoid cells exhibiting CD30 positivity with a null phenotype and coexpression of CD15 (Figs 3, 4). A diagnosis of nodular sclerosing Hodgkin lymphoma was made.

In September 2002, the patient was commenced on a six-course regimen of adriamycin (25 mg/m²), bleomycin (10 U/m²), vinblastine (6 mg/m²), and dacarbazine (375 mg/m²) on days 1 and 15. The four-drug chemotherapeutic regimen was administered every 2 weeks for a total of six treatments. After completion of chemotherapy, a repeat computed tomography and positron emission tomography scan showed no evidence of residual disease. Six months later, the patient is asymptomatic and in complete remission. The patient is currently receiving involved field radiotherapy.

Accepted for publication Nov 27, 2002.

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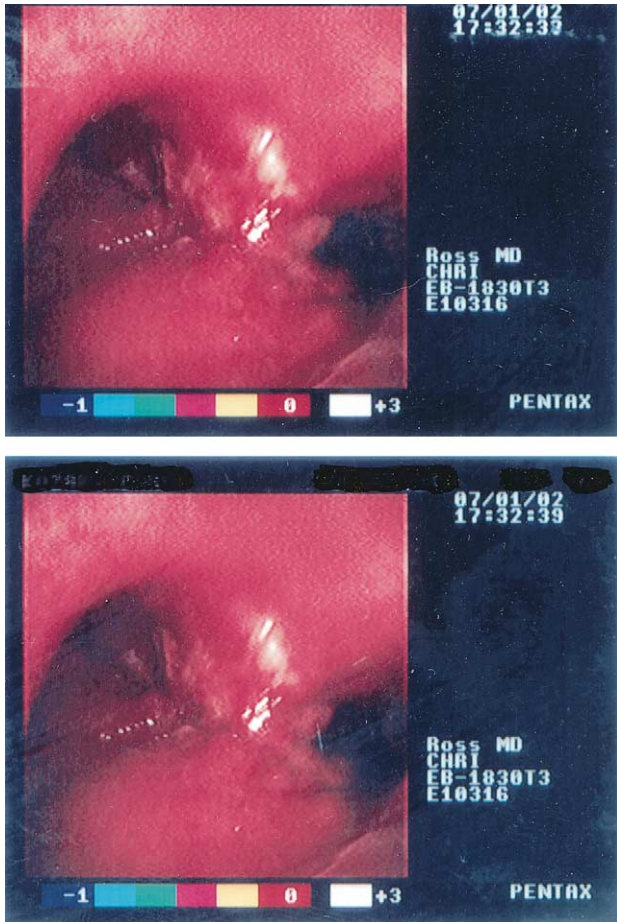


Fig 2. Patent right main stem bronchus after photodynamic therapy and debridement.

Comment

We have presented a case of classic Hodgkin lymphoma in which the initial presentation was an obstructing endobronchial lesion held clinically and initially pathologically to represent small cell carcinoma. An unusual component of his clinical presentation was one of hemoptysis caused in part by an extensive endobronchial vasculitis.

Hodgkin lymphoma is unique as a lymphoma in that the neoplastic cell component is associated with an inflammatory host response that may obscure the neoplastic cell populace. Hodgkin lymphoma is divided into (1) classic Hodgkin lymphoma, the spectrum of which encompasses nodular sclerosing, mixed cellularity, and lymphocyte-depleted forms, and (2) nodular-diffuse lymphocyte-predominant Hodgkin lymphoma. The neoplastic cell is positive for the activation marker CD30 and manifests a B-cell or null cell phenotype [5].

Occlusion of a main bronchus in endobronchial Hodgkin lymphoma may evoke a variety of nonspecific respiratory symptoms, including cough, hemoptysis, dyspnea, wheezing, chest pain, and stridor. Twenty-six cases of endobronchial presentation of Hodgkin lymphoma

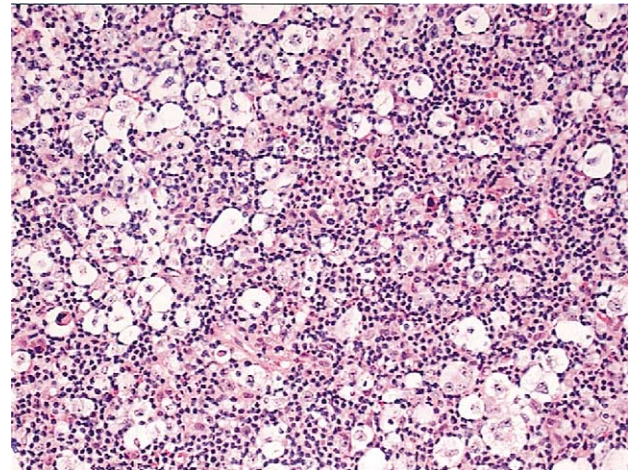


Fig 3. There is an obscuring small cell-dominant lymphocytic infiltrate within the bronchial wall. Amid this reactive cell populace are scattered typical Reed-Sternberg cells surrounded by a lacunar space, characteristic of Hodgkin lymphoma.

have now been reported in the English-language literature [2-5]. There are only two additional non-English-language citations from beyond 1966 describing endobronchial Hodgkin lymphoma. Based on the English-language review, the average age of presentation in these patients is 42.3 years, with a range of 18 to 83 years and a slight male preponderance [2-5]. Respiratory signs have been observed in most patients, with cough being the most frequent clinical feature, with or without accompanying hemoptysis. Hemoptysis is an unusual occurrence in Hodgkin lymphoma without endobronchial or endotracheal presentation, and hence its occurrence should prompt consideration of endobronchial Hodgkin lymphoma [2-5]. Radiographically, atelectasis is present in 42.3% reported cases of endobronchial Hodgkin lymphoma compared with 0.6% cases of cases of intrathoracic Hodgkin lymphoma [1]. B symptoms are present in

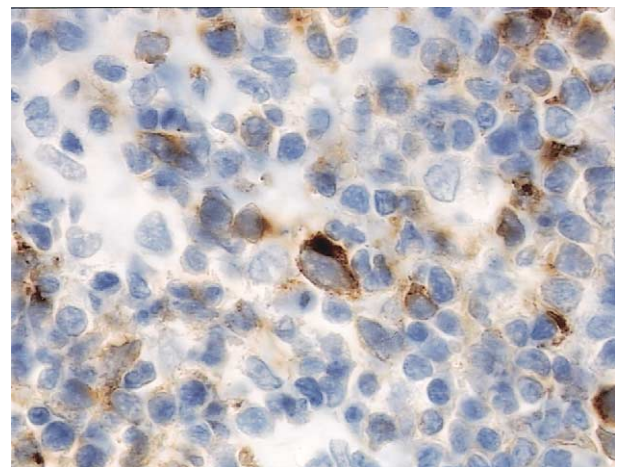


Fig 4. CD30 positivity amid the large atypical cells is consistent with Hodgkin lymphoma.

half of reported cases. Concomitant tracheal involvement occurs but is very uncommon, being described in only three cases.

The pathogenesis of endobronchial Hodgkin lymphoma is unclear. Presumably, endobronchial lesions may arise in mucosa-associated lymphoid tissue [5]. However, a more common mechanism is one of contiguous transmural spread from adjacent lymph nodes.

Our case may have caused diagnostic confusion with a primary endobronchial vasculitis, such as Wegener's granulomatosis. Although vasculitis has been reported at presentation in patients with newly diagnosed Hodgkin lymphoma, those have been paraneoplastic phenomena distant from the sites of lymphoma involvement. In contrast, this is a case of localized bronchial wall vasculitis in a patient with an endobronchial presentation of Hodgkin lymphoma. Its pathogenetic basis may be one of immune complex reaction comprising immunoglobulin and tumor antigen.

Careful staging and optimal treatment can lead to cure in 75% of patients with Hodgkin lymphoma. Although the conventional therapy for any Hodgkin lymphoma is chemotherapy (ie, adriamycin, bleomycin, vinblastine, dacarbazine) with or without radiation therapy, the overall survival for the reported cases being 76.2% to 84.2%, an interesting treatment uniquely applied in this case was the use of PDT. An alternative approach to relieve the obstruction could have been self-expanding metal stent placement, which achieves immediate relief in most patients. However, PDT was chosen over stent placement in light of its established use for palliation of extensive endobronchial small cell carcinoma, the initial presumptive diagnosis in this case. Photodynamic therapy has been proven to be an effective modality for treating endoluminal bronchogenic carcinoma, esophageal carcinoma, and certain metastatic carcinomas [6]. The premise of PDT is one of selective retention of photosensitizing agents within tumor cells whereby the agent, after wavelength-specific irradiation, generates toxic oxygen radicals evoking tumor necrosis [6]. In our experience, PDT has been effective in more than 70% of patients with endobronchial obstruction. This report represents successful application of PDT for obstructing endobronchial Hodgkin lymphoma, hence expanding the range of malignancies for which PDT therapy can be effective as palliative or definitive treatment.

References

1. Filly R, Blank N, Castellino RA. Radiographic distribution of intrathoracic disease in previously untreated patients with Hodgkin's disease and non-Hodgkin's lymphoma. *Radiology* 1976;120:277-81.
2. Kim KI, Lee JW, Lee MK, Lee CH, Park SK. Polypoid endobronchial Hodgkin's disease with pneumomediastinum. *Br J Radiol* 1999;72:392-4.
3. Tredaniel J, Peillon I, Ferme C, Brice P, Gisselbrecht C, Hirsch A. Endobronchial presentation of Hodgkin's disease: a report of nine cases and review of the literature. *Eur Respir J* 1994;7:1852-5.
4. Harper PG, Fisher C, McLennan K, Souhami RL. Presentation

of Hodgkin's disease as an endobronchial lesion. *Cancer* 1984;53:147-50.

5. Yousem SA, Weiss LM, Colby TV. Primary pulmonary Hodgkin's disease. A clinicopathologic study of 15 cases. *Cancer* 1986;57:1217-24.
6. Moghissi K, Dixon K, Stringer M, Freeman T, Thorpe A, Brown S. The place of bronchoscopic photodynamic therapy in advanced unresectable lung cancer: experience of 100 cases. *Eur J Cardiothorac Surg* 1999;15:1-6.

Traumatic Pulmonary Arteriovenous Malformation Presenting With Massive Hemoptysis 30 Years After Penetrating Chest Injury

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A 39-year-old man presented with massive hemoptysis requiring emergency double lumen endobronchial intubation, bronchial arteriography and embolization, and subsequent right lower lobectomy. He had suffered a shrapnel blast injury to the right chest as a 9-year-old boy. Pathology of the resected specimen revealed lodged metallic foreign body with traumatic arteriovenous malformation. We present this case to alert thoracic surgeons to this extremely rare clinical entity that can present itself many years after the penetrating trauma, which requires urgent surgery.

(Ann Thorac Surg 2003;76:942-4)

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Traumatic pulmonary arteriovenous malformations (AVMs) after penetrating chest trauma are rarely described. We present the following clinical case requiring urgent surgery to remind thoracic surgeons of this unusual but life-threatening entity in patients presenting with massive hemoptysis after penetrating chest injury.

A 39-year-old man presented to our institution in July 2001 with two episodes of copious hemoptysis. He had suffered hemoptysis 5 years earlier and was investigated at another institution where a conservative approach was undertaken. He was previously fit and healthy, a non-smoker, with no history of previous pulmonary infections, including tuberculosis. He had sustained a penetrating shrapnel injury to the right chest at the age of 9 years from a bomb blast in Bangladesh. Clinically there were no specific findings on examination except for a scar

Accepted for publication Jan 17, 2003.

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