

A Case of Hodgkin's Disease Presenting with Recurrent Laryngeal Nerve Palsy and Tracheoesophageal Fistula

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Summary

Malignancy is the most common cause of tracheoesophageal fistulas. The malignancies commonly implicated in the development of tracheoesophageal fistulas are primary bronchial or esophageal carcinomas. Hodgkins disease rarely leads to such a fistula. We report a case of Hodgkin's disease with a tracheoesophageal fistula as well as a left recurrent nerve palsy at presentation. This presentation has no precedent in literature. The patient was treated with systemic chemotherapy and involved field radiotherapy. On follow up 1 year after the completion of treatment, he was clinically and radiologically disease free.

Key Words: Hodgkin's disease, Tracheoesophageal fistula, Recurrent laryngeal nerve palsy

Introduction

Neoplastic disease is the most common cause of acquired fistulas between the esophagus and the tracheobronchial tree. Almost all of these are due to either primary bronchial or esophageal carcinoma¹. The occurrence of tracheoesophageal fistula (TEF) in cases of Hodgkin's disease is a rare event. Rarer is the occurrence of fistula before the start of treatment of the disease. We report a case of Hodgkin's disease with TEF as well as a left recurrent nerve palsy at presentation.

Case Report

In September 2002, a 27 year old male presented to the Lymphoma clinic of the cancer hospital of our institute with history of hoarseness and loss of weight of 3 months duration. He also complained of dry cough for the last 2 months, which was aggravated with oral intake. On examination, there was a 2 x 2cm lymph

node in left supraclavicular region. Systemic examination was normal. Biopsy from nodal mass was performed which revealed Hodgkin's disease depicting Nodular sclerosis (Fig 1) On ENT evaluation left vocal cord palsy was recorded. Contrast enhanced computed tomography (CECT) chest revealed mediastinal adenopathy involving the paratracheal, retrotracheal and subcarinal lymph nodes. The nodes were also causing displacement of trachea anteriorly, indenting the posterior wall of trachea and causing displacement of esophagus towards the right. There was a clear communication between the tracheal and esophageal wall in the CT scan (Fig 2).

Barium swallow revealed a TEF with the contrast trickling into the tracheo-bronchial tree. Bone marrow aspiration and biopsy were normal. The patient was diagnosed as a case of Hodgkin's disease stage IIB with TEF and left vocal cord palsy. He received four cycles of chemotherapy with ABVD (Adriamycin, Bleomycin, Vinblastine and Dactinomycin) from October 2002 to

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CASE REPORT

January 2003. After 2 cycles of chemotherapy he could swallow without any cough and his hoarseness had improved. Subsequently, in February 2003, he received involved field radiotherapy to the mediastinum to a dose of 25 Gray in 15 fractions over 3 weeks. On his last follow up in June 2004, he was clinically and radiologically disease free without any evidence of fistula.

Discussion

The development of TEF in a malignancy is a serious event. A vast majority of TEF's are due to primary bronchial or esophageal carcinoma¹. Hodgkin's disease with any sort of fistula formation is extremely rare. The first case of TEF caused by lymphoma in 1930 and subsequently there have been few other sporadic reports in literature.

TEF in lymphoma can develop in two possible ways. First and the more common etiology is the necrosis and breakdown of the lymphomatous diseased area between the tracheobronchial tree and the esophagus. This can occur subsequent to radiotherapy or systemic chemotherapy. The second possibility is the occurrence of fistula due to disease itself in which case the TEF manifests at the time of presentation as has happened in our case².

In his review of tracheoesophageal fistulas in lymphomas, Perry mentioned that of a total of 38 patients reported in literature 87% had Hodgkin's

disease and 13% had non - Hodgkin's lymphoma². Only 8 patients had TEF before any disease directed therapy. Kuliszkiwicz analysed 330 patients with Hodgkin's disease and reported the development of fistula between the respiratory and alimentary tract in 5 patients (tracheoesophageal in three and bronchoesophageal in two)³. In our patient, the fistula was between the trachea and the esophagus (3cm above the carina).

The cause of recurrent laryngeal nerve palsy and the TEF in our case is most likely the soft tissue extension from the nodes in the mediastinum. After an extensive literature search we could find only one case of recurrent laryngeal nerve palsy in Hodgkin's disease. However, this patient developed the tracheoesophageal fistula and recurrent laryngeal nerve palsy only after radiotherapy for Hodgkin's disease⁴. In view of paucity of literature, it can be postulated that recurrent laryngeal nerve palsy occurs due to either direct infiltration of the recurrent laryngeal nerve or due to the compression by the nodal mass. For obvious anatomical reasons, only the left recurrent laryngeal nerve can get involved.

The management of TEF in patients with lymphoma has to be individualized. Often a conservative approach is warranted in small fistulas with the expectation that many of these fistulas will close after radiation therapy or chemotherapy. In the review by Greven and Evans most fistulas (7/10) closed promptly with RT to the site⁵. In patients in whom the fistula fails to close or enlarges with therapy, direct surgical repair

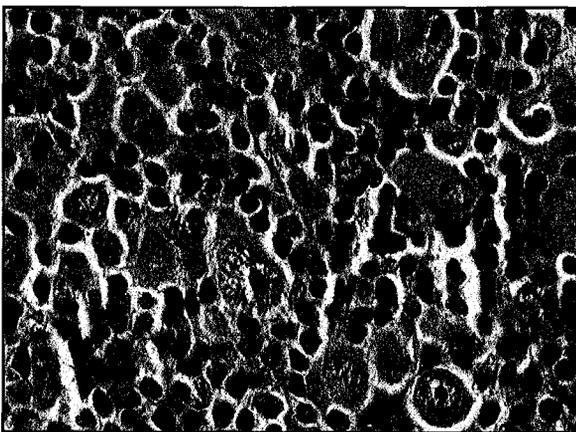


Fig 1: Photomicrograph of the nodal biopsy showing features of nodular sclerositis variant of Hodgkin's disease.



Fig 2: CT scan showing the frank fistulous tract between the trachea and the esophagus.

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may then be attempted. Unlike TEF caused by carcinoma, many of the fistulas that develop in lymphoma patients are obliterated after chemotherapy or radiation therapy without a direct surgical intervention on the fistula¹.

All the literature we reviewed was consistent in designating a favorable prognosis of TEF in Hodgkin's disease in contrast to the dismal outcome in aggressive malignancies such as carcinoma esophagus or carcinoma bronchus. In the Memorial Hospital series of 111 patients with TEF caused by carcinoma of the

lung or the esophagus, 79.2% of patients died within 3 months. Even the longest survivor lived only for 23 months. In contrast to this it was noteworthy that there was 75% survival rate at a mean of 16 months in patients with lymphoma and TEF. This compared favorably with the survival rate of patients with lymphoma who never had TEF¹.

To summarise, patients with lymphoma-related TEF have a much better prognosis than do those with TEF caused by carcinoma of the lung or esophagus. Also, they require far less active intervention.

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