Case Report

A rare case of laryngotracheal chondrosarcoma in a patient with past history of radioiodine therapy for thyroid cancer

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Abstract

Tracheal chondrosarcoma is a rare malignant mesenchymal tumor and there are less than 15 reports in the literature. We report a rare case of laryngotracheal chondrosarcoma in a 74-year-old man. He gave a history of radioiodine therapy for thyroid papillary carcinoma about 24 years ago. Diagnostic steps, histological presentation, and therapy are described in detail.

Key Words: Radioiodine therapy, thyroid cancer, tracheal chondrosarcoma

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INTRODUCTION

Primary malignant tracheal tumors are rare, accounting for only 0.2% of all malignancies of the respiratory tract.[1] Chondrosarcoma of the trachea is a rare condition and there are less than 15 reports in review of the English literature in Medline.[1-3] Here, we present a rare case of laryngotracheal chondrosarcoma in a 74-year-old man, who has a past history of radioiodine therapy for thyroid papillary carcinoma, and briefly review the literature.

CASE REPORT

A 74-year-old male patient suffered from progressive dyspnea, hoarseness, and stridor over a period of a few months prior to preoperative assessment. He had a total thyroidectomy and was treated with 3,700 MBq (100 mCi) of radioactive iodine following thyroidectomy due to thyroid papillary carcinoma. Computed tomography (CT) scan and fiberoptic bronchoscopy were performed with suspicion of recurrent papillary carcinoma in cricoid cartilage and trachea. There was a mass in the left side of the subglotic area with intact mucosa [Figure 1]. After exploration of the neck via a collar incision, a mass on the left side proximal to the trachea and cricoid cartilage with involvement of left recurrent nerve was discovered. Proximal trachea, in addition to the left half of cricoid cartilage, was resected.

In order to reconstruct the airway, the trachea was cut in the shape of the letter L. So, the left and longer side of the trachea was sutured to the thyroid cartilage and the short right side to right half of the cricoid cartilage. Fiberoptic bronchoscopy was used for airway assessment and the patient was put on orotracheal intubation for 48 h. After extubation 48 h later, good phonation was observed in the patient without any

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respiratory problems. The patient was discharged from the hospital without any complications. In gross pathologic examination, the specimen consisted of a fragment of trachea measuring 4 × 3 × 2 cm that in cut section showed a polypoid mass in the inner wall with chondroid consistency and gray color. Microscopically, the sections revealed neoplastic proliferation of chondrocytes in a pale, basophilic, hyaline chondroid matrix. The chondrocytes showed increased nuclear-to-cytoplasmic ratios, irregular nuclear membranes, and nucleoli. Mitotic activity was scant, but cellularity was markedly increased and several chondrocytes were seen on a single lacuna, indicating low-grade chondrosarcoma [Figure 2].

DISCUSSION

Primary tumors of the trachea include <0.1% of all malignant diseases, showing an incidence of 2.6 million per year. Only 8% of these tumors are seen in children.[2] Carcinoma is the predominant type of malignancy involving the trachea, accounting for 80-90% of all cases. However, it represents only 0.2% of all malignant tumors of the respiratory tract.[4] The most common histology seen is the squamous cell carcinoma (60-90%), occurring most frequently in men, followed by adenoid cystic carcinoma or cylindroma, which is more frequent in women. Chondrosarcoma of the trachea is a rare condition and there are less than 15 reports in the literature since it was first described by Jackson in 1965. It typically affects men in the age range 32-87 years, the mean age being 65 years.[1] In terms of low-grade tumors, the male-to-female ratio is 8:1, with no particular gender predilection for high-grade tumors.[2,5]

The tracheal chondrosarcoma arise from the cartilaginous rings and may cause an obstructive syndrome depending on whether the tumor grows intraluminally or extends through the tracheal wall.[3] Dyspnea is the most common presenting symptom, followed by cough, wheezing, hemoptysis, hoarseness of voice, and pneumonia.[1] Palsy or fixation of the vocal cords occurs in more than half of all cases.[2] These malignancies have a very slow growth and tend not to metastasis, in contrast to chondrosarcomas found elsewhere in the body. These malignancies present as a bulky tumor and obstruct >75% of the tracheal lumen.[2,3] Mean duration of complaining from these symptoms is over 15 months. Asthma and exacerbation of chronic obstructive pulmonary disease are the two main misdiagnosis of the condition, due to the association of dyspnea, cough, and wheezing.[1,6]

The etiology still remains unclear; however, three theories are proposed by majority of the authors: Secondary to congenital cartilaginous rest, abnormal cartilage ossification, and secondary chondroplasia from chronic inflammation.[1,4] Our patient had a history of radioiodine therapy for thyroid cancer. Some studies report an increased relative risk for certain types of cancer in organs that concentrate iodine (such as bladder, kidney, salivary glands, or colon malignancies). Most of these patients received very high radioactive iodine doses. Increased risk of secondary primary malignancy is not reported in other studies; therefore, no definite conclusion with respect to risk for certain types of malignant disease can be drawn.[7,8] However, Franklyn *et al*.[9] reported lower incidence of cancer of trachea, bronchus, pancreas, and lymphatic in patients receiving radioiodine treatment for thyrotoxicosis. Mortality from cancer of bronchus and trachea is significantly reduced.

In general, tumors of the trachea arise from the posterior and lateral wall; however, in a small

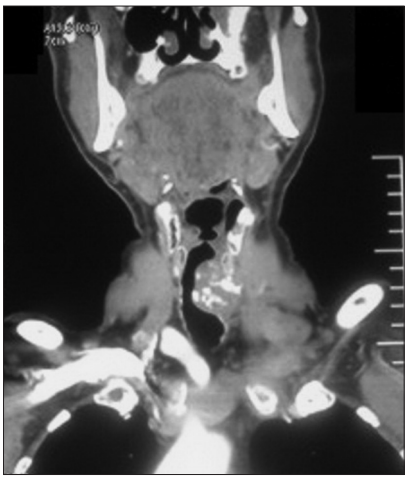

Figure 1: Coronal computed tomography scan of the neck showing a large endotracheal soft tissue mass with coarse calcification foci on the left side of the subglotic area, causing severe narrowing of the tracheal lumen

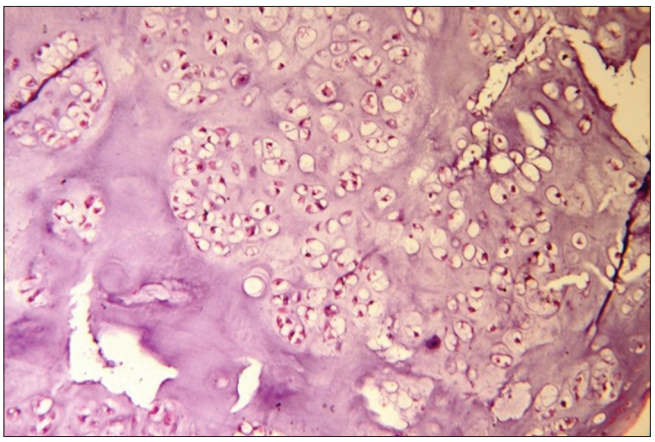

Figure 2: Representative section revealed basophile hyaline chondroid matrix, typical lobulated architecture, increased cellularity, and mild cellular atypia, consistent with low-grade chondrosarcoma (H and E, ×400)

percentage of malignant neoplasms, the involvement is circumferential. In the course of a malignant disease, there may be a spread through the wall, with extension of the mass beyond the confines of the trachea.[4]

They extend in an intraluminal direction and are macroscopically impossible to discern from enchondromas. Microscopically, tracheal chondrosarcoma is a typically gray-white, firm, well-circumscribed, multi-lobulated mass; is commonly covered with normal mucosa; and like chondrosarcomas of the larynx, appear to be slow growing respiratory epithelium. The cartilage is usually hypercellular, with occasional binucleated chondrocytes. The well-differentiated tumors may be less cellular and contain islands of cells, grouped like normal hyaline cartilage.[1,2,4]

Typically, the initial plain chest radiography is normal.[3,6] Rarely, an hourglass-shaped tracheal stripe may be noted. Also, if significant air has been trapped distal to the obstructing mass, the lung fields may be overinflated bilaterally.[1] If the mass is large enough or exophytic, a filling defect will be seen on an esophagogram, which may also demonstrate any fistulous connection.[4,6] The tumor is best diagnosed with a CT scan, which shows the lesion and provides information about tumor localization, length, intra-tumoral calcification, extra-tracheal extension, resectability, nodal involvement, and distant metastases. Calcification is seen in 75% of cartilaginous tumors and may be punctate or linear, and central or peripheral.[3,10,11] Magnetic resonance imaging has no added value compared to CT. However, bronchoscopy is the best method for diagnosis and initial treatment of tracheal obstruction. Both flexible and rigid bronchoscopy should be utilized to evaluate the movement of the vocal cords, to identify the proximal and distal extent of the lesion, and to perform biopsies.[1,2]

The optimal treatment for tracheal chondrosarcoma is tracheal resection with free margins and end-to-end anastomosis. But in this case, resection and anastomosis were difficult due to the involvement and resection of left half of the cricoid cartilage. Aggressive complete surgical resection, with or without initial endoscopic airway restoration, is the treatment of choice. Incomplete resections are complicated by high rates of local recurrence and by an increased risk of metastatic disease.[3,12] Chondrosarcoma, especially a lower-grade tumor, is not responsive to chemotherapy. Although chondrosarcoma has been considered radio resistant, there are reports of chondrosarcomas arising in bone being successfully treated with radiotherapy.[1,6]

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