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Case Report

A case of tracheal adenoid cystic carcinoma treated with carinal resection and adjuvant radiotheraphy

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ABSTRACT

Adenoid cystic carcinoma originates from the glands in the tracheobronchial mucosa and its primary treatment is complete surgical resection.

A 43-year-old male patient presented to our clinic with the complaints of cough, phlegm and haemoptysis continuing for the last 2 years. In his physical examination, a decrease in breath sounds was found in his left hemithorax during auscultation. As chest x-ray showed a left hilar enlargement, a thoracic computed tomography was taken which revealed a mass 17x21 mm in size extending from the lateral side of the lower left tracheal wall into the left main bronchus, considerably constricting the lumen. A biopsy was taken from the lesion during fiberoptic bronchoscopy and its histopathologic examination led to the diagnosis of Adenoid cystic carcinoma.

The patient was operated under general anaesthesia using right posterolateral thoracotomy which involved carinal resection and anastomosis of the distal side of the trachea with the right main bronchus end-to-end, and of the right intermediary bronchus with the left main bronchus end-to-side. Since surgical margin positivity was found in histopathologic examination, he was prescribed adjuvant radiotherapy. The patient is still asymptomatic after a 2-year follow-up.

Although negative surgical margins may not be achieved in locally advanced adenoid cystic carcinoma, resection of the tumor through complex surgical procedures with the addition of adjuvant radiotherapy enables long term survival.

Keywords: adenoid cystic carcinoma, trachea, surgery, radiotherapy

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Introduction

Adenoid cystic carcinoma (ACC) is a rarely seen salivary gland-like primary malignant neoplasm of the respiratory system and comprises approximately 0.2-0.04% of all primary lung cancers [1,2]. The primary treatment in ACC is surgical resection. Radiotherapy is used for adjuvant, definitive or palliative treatment purposes [3].

Case Report

A 43-year-old male patient presented to our clinic with the complaints of cough, phlegm and haemoptysis continuing for the last 2 years. There were no peculiarities in the patient's anamnesis other than having received medical treatment for bronchial asthma for 1.5 years. In his physical examination, a decrease in breath sounds was found in his left hemithorax during auscultation. As his posteroanterior chest x-ray showed a left hilar enlargement, a thoracic computed tomography was taken which revealed a lesion 17x21 mm in size extending from the lateral side of the lower left tracheal wall into the left main bronchus, considerably constricting the lumen (Figure 1). Thoracic magnetic resonance imaging revealed a soft tissue lesion approximately 13x16x25 mm in size with a slightly rough surface, which is localized at the level of the carina and extending to the trachea and left main bronchus in soft tissue intensity at T1 and hyperintense at T2, causing considerable constriction in the left main bronchus, and contrasted homogeneously after contrast (Figure 2). In fiberoptic bronchoscopy, an endobronchial mass was seen and a biopsy was taken. The patient was diagnosed with ACC after histopathologic examination and his PET-CT scan was taken for staging. In PET-CT, the lesion had SUVmax: 3.3 and no involvement was found in any other region (Figure 3).

The patient was operated using right posterolateral thoracotomy under general anaesthesia. After exploration, carinal resection and anastomosis of the distal end of the trachea with the right main bronchus end-to-end using 2.0 polyglactin, and of the right intermediary bronchus with the left main bronchus end-to-side using 3.0 polyglactin were performed (Figure 4). The patient was discharged at day 4 without any complications, but a histopathological examination of his surgical specimen showed surgical margin positivity. The patient was started adjuvant radiotherapy (RT) at postoperative week 4 and was administered 40 Gy RT at fractions of 2.0 Gy daily, 5 days a week for 20 days in total. At the end of a 2-year follow up, the patient was asymptomatic clinically, radiologically and bronchoscopically with no recurrence.

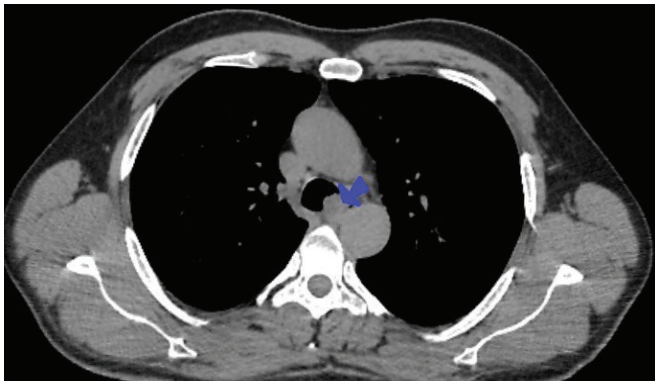

Figure 1. Lesion localized at the lower left lateral wall of the trachea, constricting the lumen as seen in the transverse section image of thoracic computed tomography (blue arrow).

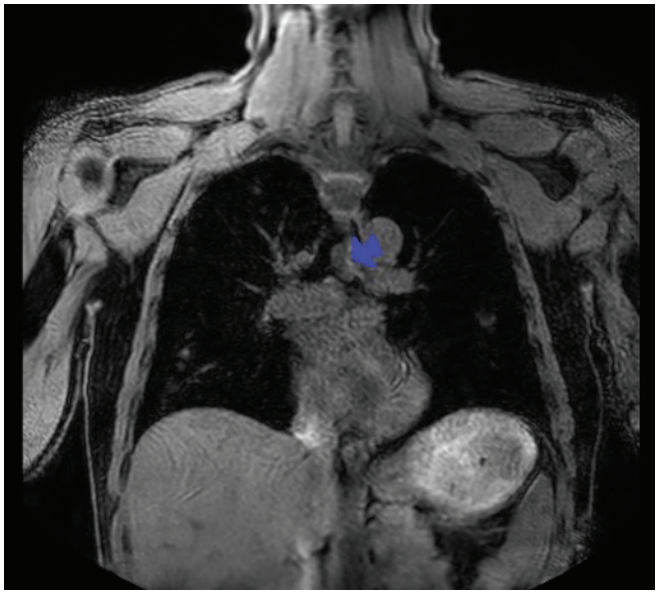

Figure 2. Soft tissue lesion with a slightly rough surface localized at the level of the carina, extending to the trachea and left main bronchus, in soft tissue intensity at T1 and hyperintense at T2 as seen in the frontal section of thoracic magnetic resonance imaging (blue arrow).

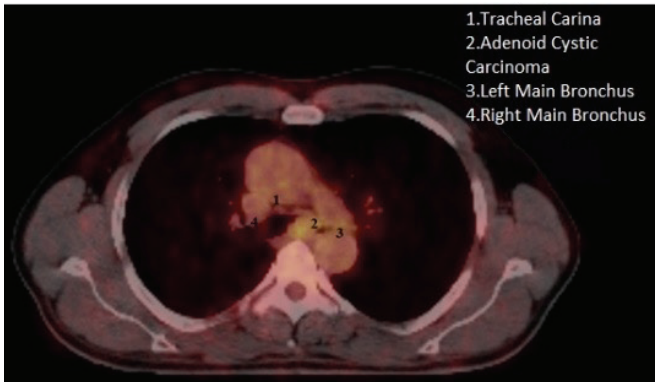

Figure 3. Lesion localized at the lower left part of the trachea and involving FDG as seen in PET-CT fusion images.

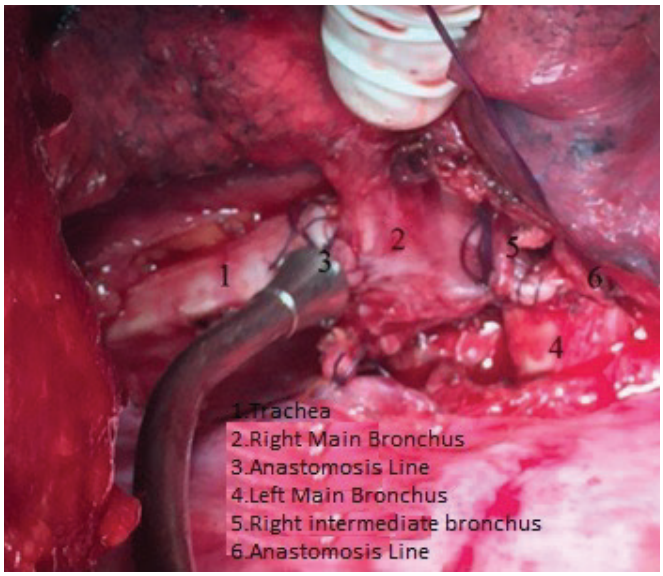

Figure 4. Anastomoses in completed form of the distal trachea and right main bronchus, and the right intermediary bronchus and left main bronchus after carinal resection as seen in the peroperative images of the patient.

Discussion

ACC is one of the rare lung tumors mostly seen around the age of 50 at equal proportions in both genders [4]. There were 2 (0.4%) ACC cases in the 515 lung cancer patients surgically treated in our clinic in the last 5 years and this data is consistent with the literature data.

In the respiratory system, 2/3 of ACC cases occur in the trachea and 1/3 in the main bronchi and distal airways. ACC is a slowly growing tumor, and depending on the mass covering the respiratory tract, symptoms such as cough, dyspnea, stridor and hemoptysis may be seen [4]. Most of these patients usually receive long-term medical treatment for the misdiagnoses of bronchial asthma or Chronic obstructive pulmonary disease. The growth in the size of the mass during this period results in worsening of the symptoms and clinical signs and leads to a misperception in the patient and physicians involved in the treatment that the disease does not respond to medical treatment. Our patient has also received medical treatment for bronchial asthma for a long time. Since his complaints persisted with the addition of hemoptysis to them in the last few months, a thoracic computed tomography was taken from the patient, which revealed a mass considerably constricting the tracheal lumen.

As ACC is a centrally localized, locally aggressive tumor and exhibits submucosal growth and perineural infiltration, the possibility of its effective treatment is quite low. A complete surgical resection whenever possible is the gold standard of treatment in these patients [3]. However, it is often difficult to achieve negative surgical margins due to the submucosal growth tendency of the tumor. Positive resection margins after surgery are seen at a rate ranging between 8-82% in the literature [3]. Since the tumor extended from the lower left lateral wall of the trachea into the left main bronchus in our case, a complex surgery was performed during the same session, which involved carinal resection, anastomosis of the distal end of the trachea with the right main bronchus, and anastomosis of the right intermediary bronchus with the left main bronchus. Despite such a wide resection, tumor was found at the surgical margin and the patient was given RT. Surgical margin positivity is an expected problem in ACC and postoperative adjuvant RT is currently recommended to sterilize the microscopic tumors at the region. With RT, local control can be achieved and long disease-free survival and overall survival can be accomplished [2,4]. No recurrence after RT was seen in our patient during the 2-year follow-up. Although ACC is known to be a slow growing tumor with low malignancy and rare regional lymph node involvement, local recurrence and distant metastasis to the lungs, bones and brain at even very late periods may occur as part of unpredictable clinical course in these patients [5]. Therefore, long-term careful follow-up is a must in all such patients including ours.

In conclusion, although negative surgical margins may not be achieved in ACC cases, resection of the tumor through complex surgical procedures and the addition of adjuvant RT will enable obtaining pleasing outcomes with long term survival rates.

Declaration of conflicting interests

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Authors' contributions

ME,OT,SK,AA,CT: conceived and designed the current case report, co-wrote the paper, collected the clinical data. The authors discussed the case under the literature data together and constituted the final manuscript.

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