

CASE REPORT

Management of Adenoid Cystic Carcinoma of Distal Trachea

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Summary

Tracheal neoplasms occur infrequently, the majority of primary tracheal tumours in adults are malignant, and in 40% of cases it is adenoid cystic carcinoma, which can be asymptomatic for a long period of time. Surgical treatment can be justified even in advanced disease, because progression of tracheal tumour usually causes obstruction of airways. Improved long-term outcome seems to follow combined surgical resection and full-dose radiotherapy. Patients after resection of adenoid cystic carcinoma need careful and long-term observations to detect a relapse.

Key words: Adenoid cystic carcinoma, tracheal resection, tracheal obstruction

AIM OF THE DEMONSTRATION

The aim of this demonstration is to show the management of patient with delayed diagnosis of adenoid cystic carcinoma of the distal trachea which caused near-total obstruction of the lumen of trachea.

CASE REPORT

A 38-year-old female presented to the Emergency Department of Regional Hospital for fever, shortness of breath and non-productive cough in May 2011. A chest X-ray revealed normal findings and CT scan findings were also interpreted as normal, therefore she was diagnosed with chronic obstructive pulmonary disease and sent to local pulmonologist. After prolonged treatment with inhaled corticosteroids and bronchodilators, she still had shortness of breath on exertion and mild wheezing. In September 2011 she was referred to the Department of Pulmonology of our institution. Fiberoptic bronchoscopy revealed approximately 4 cm long circular tumour in the distal part of trachea with partial obstruction of tracheal lumen (Fig.1). Straightforward rigid bronchoscopy was performed for partial resection of tumour tissues, and silicone Dumont stent was inserted to avoid compromising airways (Fig.2). Pathology was consistent with adenoid cystic carcinoma. Following CT scan showed potential resectability and no distant spread of the tumour (Fig.3). Decision to proceed with surgery was made. Standard right lateral thoracotomy approach was chosen. Transthoracic approach provided excellent access to the lower trachea and carina, including the left main bronchus. Intraoperative fiberoptic bronchoscopy was used for accurate tumour localization. Resection of distal part of trachea was performed by using combined cross-field intubation and high-frequency jet ventilation techniques (Fig.4). Anastomosis was made by running non-absorbable monofilament 3-0 suture. Macroscopically complete resection of tumour was

achieved (Fig.5). The patient had uneventful recovery after surgery. Unfortunately final pathology results revealed tumour cells in the resection margins, therefore patient was referred for radiotherapy. Patient completed full-dose (60 Gy) radiotherapy course on January 2012. The last follow-up was made one year after surgery in September 2012 and showed no signs of relapse (Fig.6). Long-term follow-up is scheduled for this patient.

DISCUSSION

The incidence of primary tracheal tumours in the general population is not precisely known, tracheal tumours are accounting for less than 1% of all malignancies¹. Primary tracheal neoplasms are still often diagnosed long after the onset of symptoms. Tracheal tumours often present with signs of upper airway obstruction. Delay in diagnosis occurs because the pulmonary fields remain normal on a chest radiograph². Adenoid cystic carcinoma of the trachea occurs over a wide age range, from the twenties through the seventies with slight female predominance. No relationship has been discerned with cigarette smoking or other known carcinogenic factors. Adenoid cystic carcinoma seems to be more prevalent in the lower trachea and carina. Stage for stage, adenoid cystic carcinoma has the better prognosis, with 5-year survival rates cited to be 66-100% and 10-year survival rates of 51-62%³. Adenoid cystic carcinoma has been reported to have sensitivity to radiation⁴.

There is a higher risk for local recurrence and positive surgical margins with distal tracheal location⁵, as we described in our case. Late local recurrence of tracheal adenoid cystic carcinoma after apparent cure by surgery and irradiation is a discouraging characteristic, as adenoid cystic carcinoma can invade microscopically submucosally and perineurally for long distances beyond grossly visible disease. Frequently, adjacent to

the main mass of tumour, which clearly projects from the mucosa, there is evidence of tumour infiltration beneath the mucosa or in the tracheal wall, the margins are often indistinct.

Surgery is usually indicated once diagnosis of a primary tracheal neoplasm is made. This is because these patients tend to progress rapidly once symptomatic because of the near-total tracheal luminal obstruction that is frequently present.

Tumours of the lower trachea and carina are best approached through a high right posterolateral thoracotomy. Some surgeons (Pearson) prefer a median sternotomy. Sometimes it may become necessary to accept microscopic tumour at the margins of resection, in order not to compromise the possibility of healing by excessive anastomotic tension following an extended resection⁶. Long-term survival more than 10 years, however, is also observed after tracheal resection of locally advanced adenoid cystic carcinoma⁷.

Conflict of interest: None

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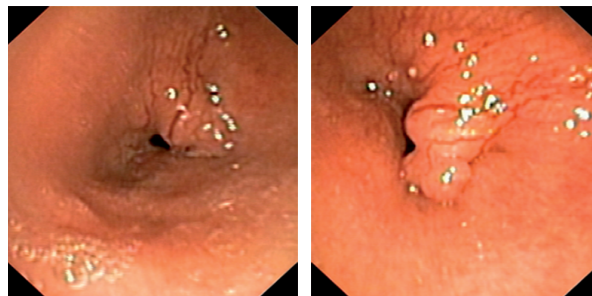


Fig. 1. Initial endoscopic (fiberbronchoscopy) view.

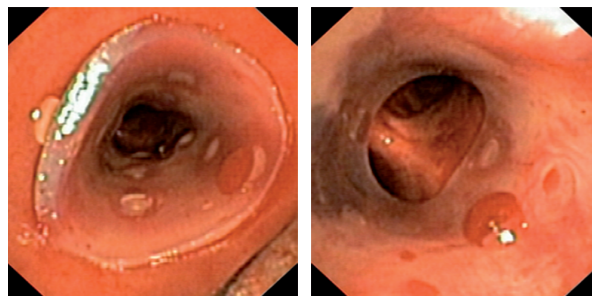


Fig. 2. Endoscopic view (fiberbronchoscopy) after insertion of silicone Dumont stent.

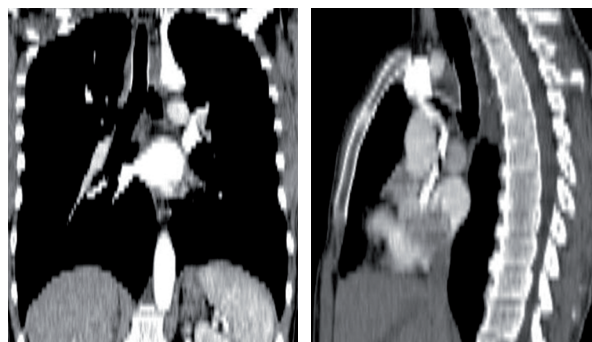
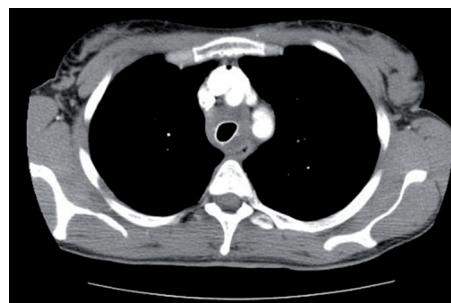


Fig. 3. CT scan (above) and reconstructions in frontal and sagittal view (below) showing stent in the distal trachea and no signs of distant spread of the tumour.

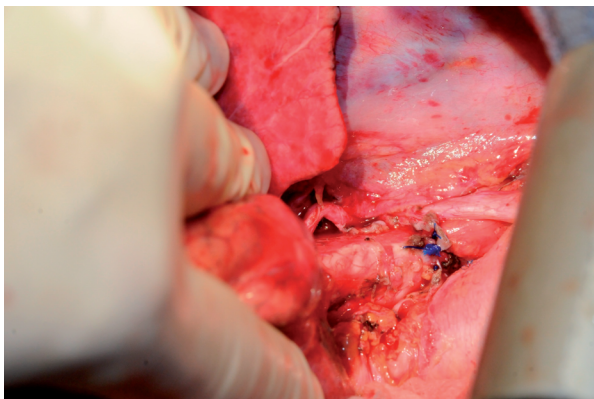
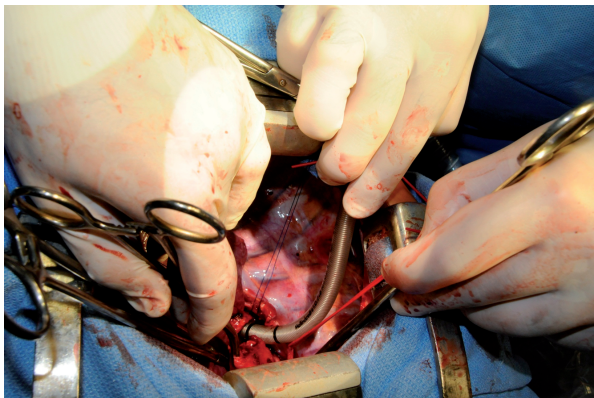
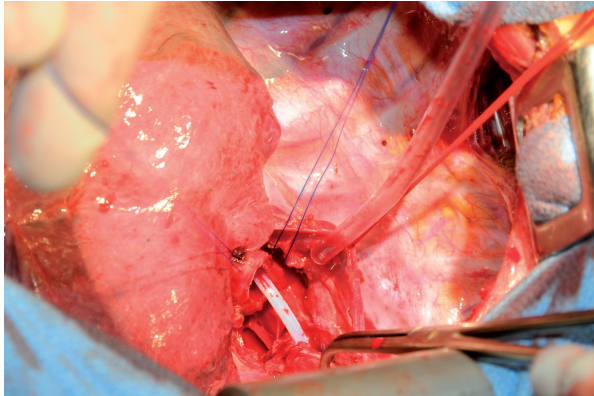


Fig. 4. Intraoperative views: resection of trachea using high-frequency jet ventilation technique (above), starting anastomosis of posterior wall of trachea using cross-field intubation technique (middle), tracheal anastomosis completed (below).

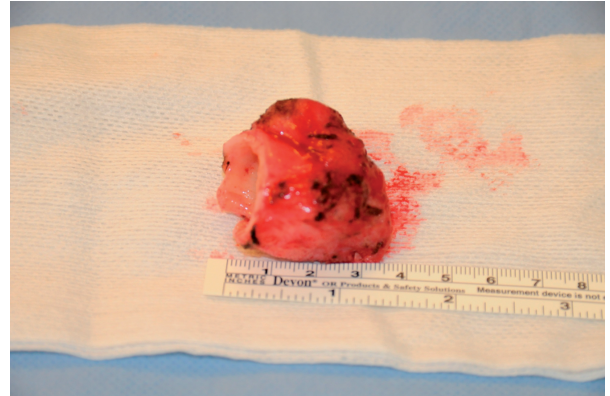


Fig. 5. The resected specimen.

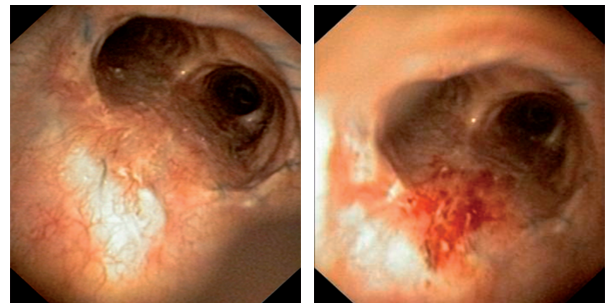


Fig. 6. Endoscopic view on follow-up one year after operation – before (left) and after (right) control biopsy.