Case Report

Large endotracheal tumor obstructing the central airway The role of Interventional Pulmonology

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INTRODUCTION

Central airway (trachea and main bronchi) obstruction either produced by benign or malignant disease is potentially life threatening and requires immediate medical attention. Management of such cases usually warrants multidisciplinary team approach. However, therapeutic interventional pulmonology with multiple flexible and rigid bronchoscopic modalities has established its central role in this entity. In this report we are presenting a challenging case of central airway obstruction caused by a massive endotracheal tumour highlighting the immediate and dramatic symptomatic relief and the long term management of the patient achieved by applying algorithmic interventional therapeutic approach.

CASE HISTORY

We are reporting on a 48 years-old male patient, ex-smoker of 40 pack/ years, who was referred to our unit complaining for cough and dyspnea on exertion during the last year. His past medical history was unremarkable for any chronic medical illness. An audible inspiratory stridor was heard over his trachea, otherwise respiratory and general examinations were unremarkable. Work up for his condition revealed a significant endotracheal mass which obstructed 80% of his tracheal diameter. Spirometric values were: FVC= 4.03L (87% pred), FEV1= 1.06L (28% pred), FEV1/FVC= (26% pred), PEF= 5.34L (59% pred), FEF_{25-75%}= 0.30L (7% pred).

Following the initial assessment, bronchoscopy was performed (Figure 1).

Thoracic surgeons declined immediate surgical resection of the tracheal tumor due to its length of 3.5 cm and the patient's critically compromised ventilation.

At our unit, complete debulking of the tumor was decided and scheduled in order to diagnostically fully assess the disease, and therapeutically re-establish airway patency as well as to provide local bronchoscopic treatment. Through the rigid bronchoscope, electrocautery of the tumor base was performed in order to decrease the tumor bleeding during debulking. Thereafter, the tumor was mechanically removed by the beveled tip of the



FIGURE 1. CT of mid-trachea, Flow volume curve and bronchoscopy before treatment.

rigid scope and the forceps. During the procedure, it was necessary to break the tumor in to pieces as the diameter of the tumor was larger than the diameter of the rigid scope. Following complete tumor removal, the tumor base was treated using cryotherapy. Cryotherapy freezes the tissue to below -40 C, which causes an immediate dehydration and cellular crystallization. A delayed effect due to local ischemia caused by microthrombi formation is also expected.

Our patient tolerated the procedure well with no immediate complications.

Follow up flexible bronchoscopy documented the full re-establishement of tracheal patency with a completely normal pulmonary physiology. Spirometric values were: $FVC= 5.12L (111\% \text{ pred}), FEV_1= 4.37L (116\% \text{ pred}), FEV_1/$ $FVC= (85\% \text{ pred}), PEF= 9.09L (101\% \text{ pred}), FEF_{25-75\%}= 5.27L$ (128% pred) (Figure 2).

Histopathology result revealed atypical small sized cells with scant cytoplasm and hyperchromatic nuclei,



FIGURE 2. Endoscopic view of the trachea after removing the tumor and F/V curve the following day.

with infrequent mitosis. Characteristic patterns included tubular and solid patterns. Immunohistological staining was positive for cytokeratin, vimentin and actin indicating the diagnosis of adenoid cystic carcinoma (Figure 3).

After discharge, the patient was further examined by his treating physicians and remained without any symptoms for 2 months. Following oncologic consultation, he was advised to further undergo surgery with complete resection of the base of the tumor with end-to-end anastomosis of the trachea. The surgery was performed successfully and safety margins were achieved. No further completion radiotherapy or oncological therapy was indicated.

DISCUSSION

Adenoid cystic carcinoma (ACC) of the trachea is rare representing almost 1% of all respiratory tract cancers. It's generally considered as a slow-growing tumor, with prolonged clinical course. Most patients present with dyspnea, and the symptoms often mimic those of asthma or chronic bronchitis. ACC arise from mucous secreting cells, usually of the salivary glands and upper respiratory tract. The most commonly affected sites are the salivary glands but ACC is known to occur in the oesophagus, nasopharynx and trachea, other sites outside the head and neck have also been reported.¹ There is no sex predilection and no association to smoking habit.

Differential diagnosis includes carcinoid tumors, basaloid squamous cell carcinoma and small cell carcinoma all of which can be typically distinguished by immunohistochemical staining. Surgical resection is the mainstay of treatment often combined to radiotherapy because of close surgical margins. When surgery isn't possible, most



FIGURE 3. A. H/E stain showing subepithelial infiltration of adenoid cystic carcinoma. **B.** Smooth muscle actin [SMA(+)] stained section of the tumor. Immunoreactivity of myoepithelial cells in the areas of adenoid cystic carcinoma.

tumors respond to interventional management with endoscopic removal of the tumor followed by radiotherapy often resulting in very long periods of remission.²⁻⁶ In other cases bronchoscopic treatment may act as a temporary means of re-establishing airway patency and "bridge" before definite surgical treatment.

Treating central airway obstruction (CAO) requires multidisciplinary team approach including thoracic surgeons, interventional pulmonologists and oncologists. Interventional bronchoscopy using both flexible and rigid scope has long established its role as a therapeutic option in early as well as in advanced-stage airway tumors.⁷ Novel bronchoscopic techniques include electrocautery, laser therapy, argon plasma coagulation, brachytherapy, photodynamic therapy and cryotherapy. Mechanical dilation of the airways (Bronchoplasty) and airway stenting are bronchoscopic possibilities which can be also used when indicated.⁸⁻¹⁰

In our case the simple debulking of the tumor was sufficient to re-establish the airway patency, and no stent placement was indicated.

Interventional techniques should ideally be available at least in all tertiary care regional hospitals in order to efficiently treat such cases of CAO which can be life threatening¹⁰. Pulmonologists should be aware of such available possibilities, and training of pulmonology fellows should include the basic techniques during their fellowship.

CONFLICT OF INTEREST

The authors have declared that no conflict of interest exists.

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