

Case report:

Pleomorphic Adenoma of Trachea

Padmavathy R, Ravindran C., Rajagopal T.P, Anandan P.T., Sunny George, Sanjeev Kumar

Department of Pulmonary Medicine, Institute of Chest Diseases,

Calicut Medical College, Kozhikode, Kerala, India

Abstract

A 28 year old female presented with gradually progressive dyspnea and wheezing of three weeks duration. Physical examination revealed stridor. Fibre optic bronchoscopy showed an intraluminal polypoidal growth almost completely occluding trachea. Histopathology of bronchial biopsy revealed a Pleomorphic adenoma, a rare tumor arising from tracheobronchial mucus glands.

Case History

A 28 year old female presented with complaints of progressive breathlessness and wheezing of 3 weeks duration. There was no past history of dyspnea, sore throat, wheezing, chest pain, foreign body aspiration or weight loss. She had no significant past medical or surgical histories nor any addictions. On physical examination, she was dyspnoeic and had stridor. Other systems were normal.

Investigations

ESR 50mm/1st hour. Sputum negative for acid fast bacilli. ECG was normal. Chest X-ray showed a suspicious radio dense lesion in the region of trachea.

Fibre optic video bronchoscopy showed an intra tracheal growth almost completely occluding the trachea.



FIG 1: Fibre optic bronchoscopy showing intraluminal polypoidal growth almost completely occluding the trachea

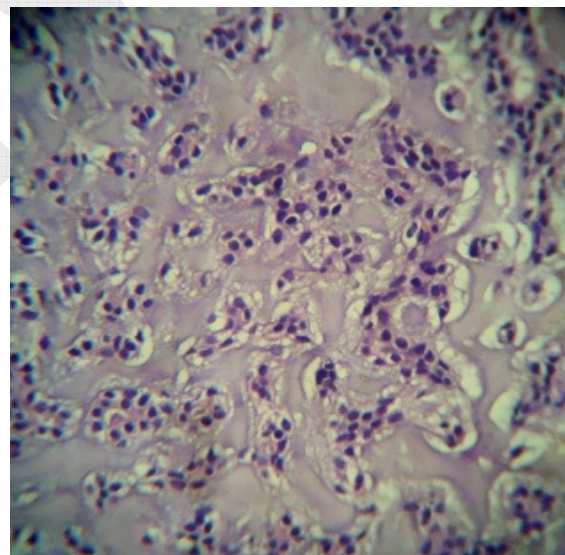


FIG 2 : Bronchial biopsy, high power microscopy showing cells with eosinophilic cytoplasm, arranged in an acinar pattern. Observe the abundant, intervening myxoid stroma.

Bronchial biopsy was suggestive of Pleomorphic adenoma arising from bronchial mucus glands



FIG 3 CT thorax showing polypoidal growth extending into the tracheal lumen.

CT thorax showed moderately enhancing endoluminal polypoidal growth of size 12 x 10 x 18 mm, arising from left lateral wall of trachea with luminal compromise. Intra luminal extension into esophagus on right side was noted. There was no significant mediastinal adenopathy. Upper GI endoscopy revealed monilial esophagitis with no intra luminal growth. The patient was managed symptomatically with bronchodilators, steroids and antifungals. Her general condition improved and she was referred for surgery.

Discussion

Primary tracheal tumors represent only 2% of upper airway tumors (1). Pleomorphic adenoma, which is the most common primary mixed tumor of the parotid gland, rarely develops in the trachea (2). To our knowledge, only 34 cases have been reported in literature, from 1922 to 2008.

The average age of these patients was 48 years (range, 26–71 years) and the sex distribution was equal.(3) Almost half of these tumors were localized in the upper one third of the trachea and 12% were localized in the lower one-third (4) .

Most intra tracheal lesions are sub glottic or intra thoracic and may present with expiratory or

biphasic stridor which may mimic non-neoplastic syndromes, like asthma and bronchitis. Pleomorphic adenoma of the trachea has an indolent course and poses a diagnostic challenge. Most patients present with a history of slowly progressive dyspnea and less commonly, hemoptysis and chronic cough which may not manifest until the lumen becomes obstructed at least by 75%. The widely diverse morphology of the tumour makes the diagnosis extremely difficult. However, fine needle aspiration biopsy maybe useful in identifying benign epithelial cells in a blue myxoid matrix background.

Grossly, the tumour is seen as gray-white, round or ovoid masses, well demarcated and encapsulated. Microscopically, it shows epithelial and mesenchymal differentiation. Epithelial component consists of ductal and non ductal cells. Mesenchymal component consists of varying degrees of myxoid, hyaline, cartilaginous, and osseous differentiation. Immunohistochemically, ductal component may stain positive for keratin, epithelial membrane antigen and carcino embryonic antigen. Spindle cell component is positive for keratin, smooth-muscle actin, and myosin. Cartilaginous areas are positive for S-100 protein (5)

There is a lack of literature regarding the investigation; management and long term follow up of tracheal pleomorphic adenomas. Most surgeons prefer to treat it similar to tumors of major salivary glands. Primary therapeutic modality is the complete tumor excision via tracheal resection. As in all major salivary gland surgery, keeping the capsule intact and resecting a margin of normal tissue reduces 10-year recurrence rate to ~2%. (6). Neutron radiotherapy, a less effective alternative for surgery, has been found superior to conventional RT in treating salivary gland tumors, but is reserved for patients with post-surgical residual disease and for recurrent pleomorphic adenoma (7). There seems to be no therapeutic role for chemotherapy in pleomorphic adenoma of trachea (8).

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Corresponding author

Padmavathy R.
Dept. of Pulmonary Medicine, Institute of Chest Diseases,
Calicut Medical College, Kozhikode 673008, Kerala, India
Ph: 09895346953

Email: dr.padmaramadoss@gmail.com