

leiomyoma beyond uterus

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Title :Leiomyoma beyond the uterus

Abstract

Leiomyoma's are benign smooth muscle tumors most commonly seen in the uterus occasionally in the gastrointestinal tract. Pulmonary localization is extremely rare with the incidence of less than 2% .We report a case of 22 year old nonsmoking female presented with left sided chest pain, streaky hemoptysis. Chest X ray suggestive of left lingular and lower lobe collapse. CECT showed endobronchial obstruction in left main bronchus. Debulking done with electrocautery snare. HPE showed spindle shaped cells with eosinophilic cytoplasm. Immunohistochemistry showed SMA positivity suggestive of leiomyoma. Patient is under followup without any complication and recurrence.

keywords :leiomyoma, endobronchial tumor, benign tumor

Introduction :

Leiomyoma's are benign smooth muscle tumours of mesenchymal origin. Most commonly seen in uterus occasionally in gastrointestinal tract. Pulmonary localization is extremely rare and accounts for less than 2%¹. Benign endobronchial neoplasms are classified as mesenchymal, submucosal glandular, and surface epithelial tumors based on their origin. Mesenchymal tumors forms the majority of endobronchial tumors, hamartoma being most common². Pulmonary leiomyoma can present as tracheal tumors, endobronchial lesion or as parenchymal lesion³. Parenchymal lesion are usually asymptomatic and diagnosed incidentally, whereas tracheal and endobronchial tumors can present as chronic cough, stridor, hemoptysis, recurrent atelectasis and present as obstructive pneumonia. Endobronchial leiomyoma rarely proposed to originate from the areas of cicatricial fibrosis. Usual age of presentation is 30-40 years and there is no gender preponderance. Few literature reports to show female predominance, it could be due to over reporting of benign metastasizing uterine leiomyoma in females⁴.

Case report:

A 22 year old nonsmoking female presented with high grade fever, left sided chest pain, streaky hemoptysis, loss of weight and appetite for one month. Her menstrual cycles were regular. On chest examination trachea shifted to left and decreased movements and decreased breath sounds over left hemithorax. Her blood work showed increased in total leucocyte count of $16 \times 10^3/\mu\text{L}$ with neutrophilic predominance (70%). Chest X ray suggestive of left lingular and lower lobe collapse. CECT chest showed endobronchial lesion in the left main bronchus. Sputum report for gram's stain and culture sensitivity showed no growth. Sputum AFB smear was negative and in sputum CBNAAT, MTb was not detected. On bronchoscopy there was a smooth polypoidal lesion with wide base, completely obstructing the left main bronchus. Cryobiopsy done and debulking of tumor done with electrocautery snare. On HPE, submucosal stroma showed sheets and fascicle of spindle shaped cells with elongated nucleus with eosinophilic rich cytoplasm with no mitotic activity, no nuclear

atypia suggestive of leiomyoma. Immunohistochemistry showed SMA (Smooth Muscle Actin) positivity. USG abdomen showed no features of uterine leiomyoma. Follow up FDG PET showed no residual tumor activity in lung and elsewhere in the body. Patient was under follow up with no complication and recurrence.

Discussion :

Pulmonary smooth muscle proliferation can either be primary, which includes hamartomas, lymphangioliomyomatosis, leiomyoma and leiomyosarcoma, or metastatic, including metastatic leiomyosarcoma and benign metastasizing uterine leiomyoma. Primary pulmonary leiomyoma is a rare tumor of mesodermal origin. It usually develops from smooth muscle fibers of tracheobronchial tree, blood vessel or embryonic heterotopic muscle islets⁵. It was first described by Forkel in 1909⁶. It accounts for 33-45% of all pulmonary leiomyomas⁷. In trachea (16%), bronchi (33%) and pulmonary parenchyma (51%)⁸. Kohn described fever, dyspnea and cough to be the predominant symptom⁹. Usually symptoms depend on degree of obstruction in affected bronchus and the state of lung parenchyma. Our patient had all symptoms as described by Kohn, along with loss of weight and appetite and hemoptysis. Since tuberculosis being most prevalent in India, she was suspected to have tuberculosis. Pulmonary leiomyoma, due to its low incidence and few literature studies, was least suspected in our case. Diagnosis of leiomyoma cannot be made solely based on radiology since there are no pathognomonic features¹⁰. Usually it presents as solitary mass, if it is endobronchial it can present as atelectasis, obstructive pneumonia, hyperlucency due to distal air trapping. Fiberoptic Bronchoscopy is used to visualize endobronchial lesion but the extent of extraluminal involvement and airway distal to obstruction cannot be visualized, hence virtual bronchoscopy and 3D airway reconstruction helps to overcome this limitation¹¹. Histopathologic examination shows disorganized smooth muscle with vasculature or fibrous component¹². Immunohistochemistry shows positivity to smooth muscle actin (SMA) and desmin which helps to differentiate it from fibroma, neurofibromas and schwannoma. There are no standard treatment guidelines. Usually endobronchial lesions are treated with electrocautery, laser photocoagulation¹³. If there is a parenchymal lesion depending upon the size and location segmentectomy, lobectomy and pneumonectomy is being done¹⁴. Our patient underwent electrocautery snare resection of tumor. Post procedure followup chest X-ray became normal. Patient was followed up with FDG PET with no residual tumor activity in lung and elsewhere in the body.

Conclusion :

Endobronchial leiomyoma is an unusual cause for endoluminal obstruction. Due to its rarity it was least suspected and posed a diagnostic challenge. Bronchoscopy helps both diagnostic as well as therapeutically as the tumor has excellent prognosis following resection.

Ethics statement : Ethical approval was not required for the case report as per country's guidelines.

Consent : Written informed consent was obtained from patient to publish the report.

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