Case Report

Bronchoscopic resection of endobronchial inflammatory myofibroblastic tumor: A case report and systematic review of the literature

Animesh Ray, J. C. Suri, Dipak Bhattacharya, Ayush Gupta

Department of Pulmonary Critical Care and Sleep Medicine, VMMC and Safdarjang Hospital, New Delhi, India

ABSTRACT

Inflammatory myofibroblastic tumour (IMT) is a rare tumour affecting the tracheo-bronchial tree in the adult population. The clinical presentation of this tumour is diverse and diagnosis can be definitively clinched by histopathological examination. Treatment of this tumour usually requires surgical resection with bronchoscopic resection being described in few cases. We describe a 32 year old male presenting with hemoptysis who was diagnosed to have IMT. Resection of the tumour was done with the help of rigid bronchoscopy. Post-resection, hemoptysis stopped and no recurrence of tumour was noted on subsequent follow-up. We also present a systematic review of literature of all the cases of tracheo-bronchial IMT treated with bronchoscopic resection and conclude it to be a useful alternative to surgery in such cases.

KEY WORDS: Bronchoscopic resection, inflammatory myofibroblastic tumor, plasma cell granuloma

Address for correspondence: Dr. Animesh Ray, Department of Pulmonary Critical Care and Sleep Medicine, VMMC and Safdarjang Hospital, New Delhi, India. E-mail: doctoranimeshray@gmail.com

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare tumor involving different organs, with lung being one of them. When it involves the pulmonary system, it can have diverse manifestations. The treatment is controversial due to a paucity of data on this rare tumor. We report a case of IMT that was successfully treated with bronchoscopic resection and also review the available literature. To the best of our knowledge, this is the first case report of successful resection of endobronchial IMT from the Indian sub-continent.

A 32-year-old patient, non-smoker and without any known co-morbidity, presented with complaints of recurrent hemoptysis for the last 2-3 years. His physical examination was unremarkable and chest X-ray did not show any abnormalities. A contrast-enhanced computed tomography (CECT) of the thorax was performed [Figures 1 and 2], which revealed an eccentric, well-defined homogenous lesion arising from the right bronchus intermedius just distal to the origin of the upper lobe bronchus (dimensions of 2 cm × 1 cm × 1 cm). No parenchymal lesion or mediastinal lymphadenopathy was noticed. Fiber-optic flexible bronchoscopy showed the mass (of the size mentioned above) almost obstructing the intermediate bronchus of the right side [Figure 3]. An endobronchial biopsy was undertaken and sent for histopathological examination, which showed sheets of plasma cells admixed with few histiocytes and lymphocytes suggestive of IMT [Figure 4]. On immuno-histochemistry, the plasma cells showed positive staining for syndecan, with both kappa and lamda positive cells; which was compatible with IMT.

A review of the literature was undertaken to determine the most appropriate line of management. Options available were either surgical resection or endoscopic resection. As the lesion was endobronchial, it was decided to endoscopically resect the tumor. Under general anesthesia, a ventilating rigid bronchoscope (Wolf ventilating bronchoscope of size 8 mm × 400 mm) was inserted. A fiberoptic bronchoscope (FOB) (outer diameter of 5.9 mm) was inserted through the rigid bronchoscope and positioned in the trachea. Then, a snare forceps was inserted

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DOI: 10.4103/0970-2113.129866

through the FOB and the protruding portion of the tumor was grasped and elecrtocauterized. The remaining portion of the tumor was removed with the assistance of a toothed forceps and electrocautery. Removal of the whole tumor was achieved [Figure 5] and hemostasis was secured. The mass was sent for histopathologic evaluation, which confirmed IMT. The post-operative period was uneventful. The patient was followed-up clinically and bronchoscopy was repeated at a 2-month interval. The patient did not have any recurrence of hemoptysis and the repeat bronchoscopies were normal (at 2, 4 and 7 months) [Figure 6].

DISCUSSION

IMT is defined by the World Health Organization as a distinctive lesion composed of a myofibroblastic spindle cell population accompanied by an inflammatory infiltrate of plasma cells, lymphocytes and eosinophils. It has been known by different names like inflammatory plasma cell granuloma, pseudotumor, fibroxanthoma, xanthofibroma, xanthoma, xanthogranuloma, etc. It can involve different systems like the lung, eye, gastrointestinal tract, etc.,

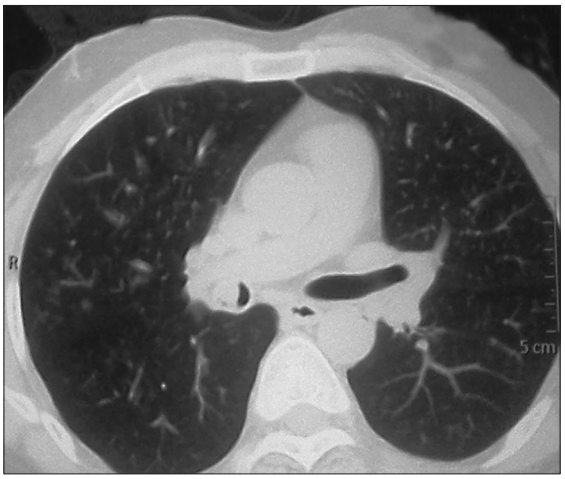

Figure 1: Contrast-enhanced computed tomography of the thorax (axial cut) showing the tumor in the intermediate bronchus of the right lung

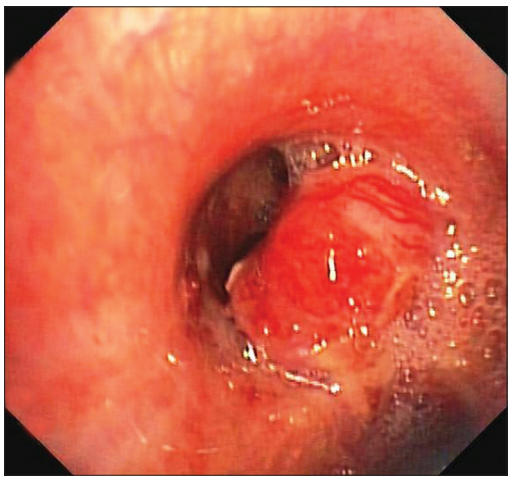

Figure 3: Bronchoscopic view showing the tumor before the procedure

Although considered the most common primary lesion of the lung in children under 16 years of age, overall, across all age groups, it is one of the rarest lung tumors with an incidence varying from 0.04% to 0.7%.[1]

The pathogenesis of IMT is controversial. Some authorities attribute it to non-neoplastic processes like metabolic disturbance, viral origin or antigen–antibody interaction to an unidentifiable agent, while some others attribute it to neoplastic processes.

The clinical presentation of the tumor can be variable, ranging from asymptomatic (70-78%)[2] to symptoms like cough, hemoptysis, chest pain, dyspnea, fever, etc., Radiology of the chest is helpful in localizing the position, which can involve any lobe or segment. It is usually solitary, although multiple lesions involving the same or different lobes of the ipsilateral of contralateral lung may be found.

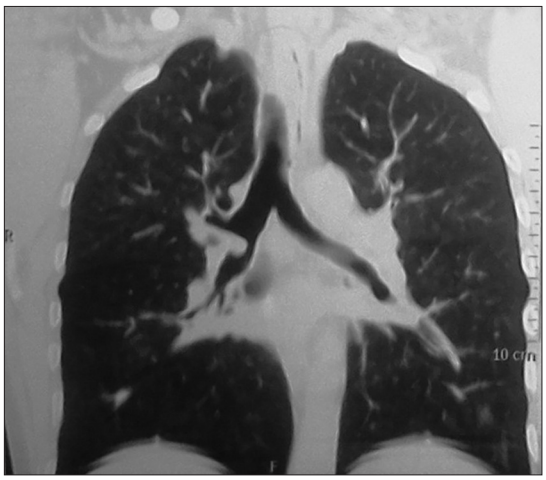

Figure 2: Contrast-enhanced computed tomography of the thorax (coronal cut) showing the tumor in the intermediate bronchus of the right lung

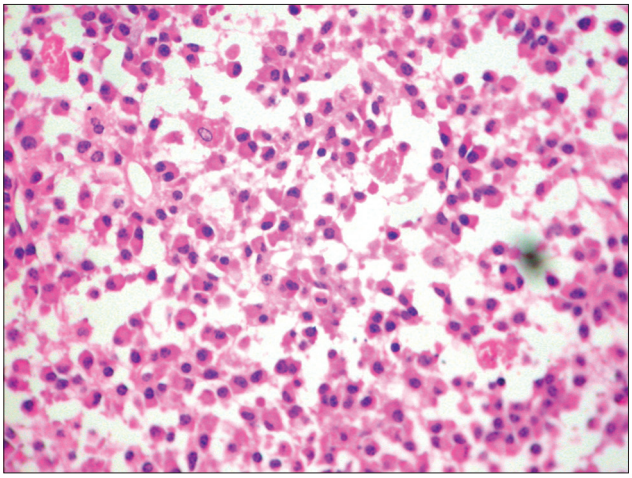

Figure 4: Histopathology of the tumor demonstrating sheets of plasma cell, histiocytes and lymphocytes typical of inflammatory myofibroblastic tumor

For clinching the diagnosis, needle biopsy,[3] wedge biopsy or resectional biopsy have been deemed appropriate. The natural history of IMT is variable. It may remain stable or grow slowly over time or regress. In some cases, it may also show invasiveness and involve the mediastium, diaphragm, chest wall, vertebral bodies, etc. Infrequent reports of distant metastasis have been made. The treatment of IMT usually entails complete surgical resection[4] of the tumor, either by video-assisted thoracoscopy or open thoracotomy. Larger lesions and those with an evidence of local invasion of the surrounding tissues will require a thoracotomy. Obtaining a tumor-negative margin is important to determine the extent of resection. For cases where the disease is deemed unresectable (multiple nodules or extensive involvement), or the patient is medically inoperable, there are sporadic reports of success with corticosteroids, radiotherapy or chemotherapy. Evidence regarding management of tracheobronchial IMT is deficient as very few case reports of such cases exist in the literature. A search was carried out using the Pubmed, Medline and Embase databases to identify cases where IMT of the tracheobronchial tree has been bronchoscopically treated (Table 1 shows the list of such cases). All the cases involved patients who were relatively young, with a mean age of 26 years (range 16-45 years). The majority of the patients belonged to the female gender (4 out of 6, in one case the gender was not specified). In 43% of the cases (3/7) the tumor was confined to the trachea, in 28.5% of cases (2/7) in one of the main bronchus while 28.5% of the cases (2/7) had lesions both in the trachea and in the bronchus. The sizes of the tumors varied between 1 and 2 cm. The major presenting symptom was dyspnea (3/7 or 48% cases), followed by recurrent pneumonia, cough and hemoptysis (2/7 or 28.5% cases). In all the cases, rigid bronchoscopy was employed and in one case Nd YAG laser was used. In 57% of the cases (4/7), post-operative steroids were used. Recurrence was noted in one case months after bronchoscopic resection, requiring surgical resection. Bronchoscopic resection is thus a viable alternative

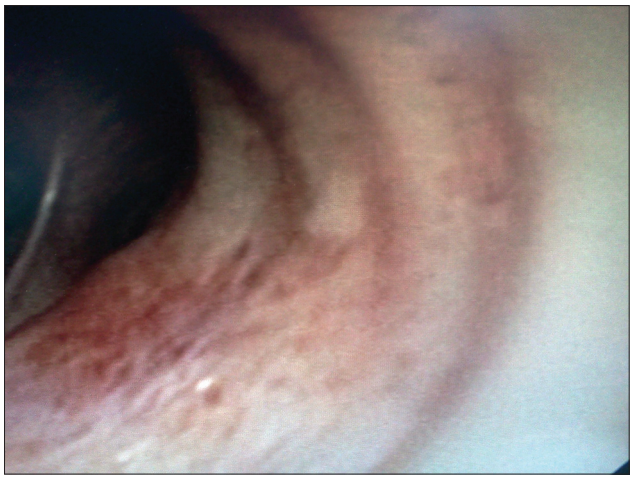

Figure 5: Bronchoscopy after the procedure demonstrating almost complete removal of the tumor

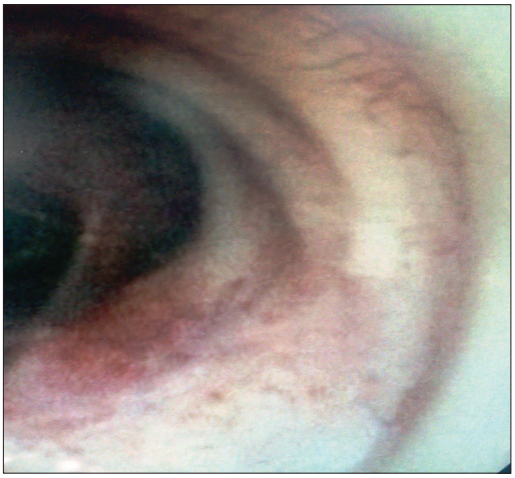

Figure 6: Bronchoscopy 7 months after the procedure showing no signs of recurrence

Table 1: Review of the literature of cases of tracheobronchial IMT treated with bronchoscopic resection

Authors	Age	Sex	Location	Size	Presentation	Procedure	Steroid use	Follow-up period
Kim *et al*. (2002)[5]	17	M	Trachea	1.5 cm×1.5 cm×2 cm	Hemoptysis, dyspnea	Br. resection	Not mentioned	Not mentioned
Nikanne *et al*. (2004)[6]	21	NA	Trachea	NA	Dyspnea, cough	Br. resection	NA	NA
Certfolio *et al*. (2005)[7]	16	F	LMB	Not available	Recurrent pneumonia	Br. resection followed by surgical resection	Yes	Months, 1 year (after surgery)
Ono *et al*. (2006)[8]	45	F	Trachea (2 cm below the vocal cord)	1 cm	Dyspnea	Br. resection/ Nd YAG laser	Not mentioned	Not mentioned
Andrade *et al*. (2010)[9]	31	F	Trachea, LMB, RMB, carina	40% lumen of distal trachea	Recurrent pneumonia	Br. resection	Yes	31 months
Oztuna *et al*. (2012)[10]	20	F	2 cm below the vocal cord, bronchus at the level of carina	Not mentioned	Cough, hoarseness, dyspnea	Br. resection	Yes (30 mg deflazcort)	6 months
Ray *et al*. (2013)*	32	M	Right intermediate bronchus	2 cm×1 cm×1 cm	Hemoptysis	Br. resection	Yes	7 months

NA: Not available, *Present case, Br. resection: Bronchoscopic resection, IMT: Inflammatory myofibroblastic tumour

in cases of IMT confined to the tracheobronchial tree. Although large-scale trials are required to compare the relative efficacy of bronchoscopic resection vis-a-vis surgical resection, it can be appreciated that enrollment of sufficient cases would be difficult given the rarity of this entity. The available literature suggests that such cases can be treated effectively with bronchoscopic resection.

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How to cite this article: Ray A, Suri JC, Bhattacharya D, Gupta A. Bronchoscopic resection of endobronchial inflammatory myofibroblastic tumor: A case report and systematic review of the literature. Lung India 2014;31:172-5.

Source of Support: Nil, **Conflict of Interest:** None declared.