

Pulmonary Inflammatory Myofibroblastic Tumor: Surgical Resection and Long-Term Surveillance

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Abstract

Objective: Inflammatory myofibroblastic tumor is a very rare mesenchymal tumor, we share our experience of surgical treatment for inflammatory myofibroblastic tumor.

Materials and methods: From January 2005 to January 2015, 32 patients were recruited in this study including 13 males and 19 females who received surgery and diagnosed as inflammatory myofibroblastic tumor. Postoperative follow-up was conducted by thoracic surgeons every 3 months until 12 months after surgery, after that every 6 months until 24 months, and then yearly.

Results: 32 patients (aged: 9-72 yr, average: 37.85 ± 19.42 yr, 13 males and 19 females) received surgical treatment for inflammatory myofibroblastic tumor (ranged: 0.8-8 cm, average: 3.84 ± 2.05 cm), including 15 cases of lobectomy, 7 cases of wedge resection, 4 cases of bronchoplasty, 2 case of tracheal tumor resection, 3 cases of sleeve resection and 1 case of wedge resection with biopsy of the chest wall. 31 patients are still alive till now, while one patient die 6 months after the surgery.

Conclusion: It suggested that complete resection of the IMT lesions may lead to a promising long-term survival, otherwise it may lead to a compromised outcome.

Keywords: Inflammatory myofibroblastic tumor; IMT; Surgical resection; Long-term surveillance

Introduction

Inflammatory myofibroblastic tumor (IMT) is a very rare mesenchymal tumor, which is a diagnostic challenge with myriad clinical presentations and pathogenetic mechanisms [1]. IMT can arise in any site in the body which is characterized by a proliferation of myofibroblastic cells admixed with inflammatory elements [2]. It mostly occurred in the lung, but it has been reported to occur in other areas including orbit, retroperitoneum, gastrointestinal tract, mediastinum, and central nervous system [3]. IMT is mostly seen in the lungs, it comprises only 0.04-1% of all lung tumors [4,5]. IMT is reported to be a borderline tumor with an aggressive biological behavior. Several case reports suggested that IMT may have the potential of metastasis and may lead to a compromised outcome. Literature reports were always single case with the low incidence of the IMT also without long-term follow-up survival data. In this study, we presented 14 cases of inflammatory myofibroblastic tumor occurred in lung and bronchus along with the long-term surveillance.

Materials and Methods

Patient characteristics

This retrospective study included 32 patients treated in the department of thoracic surgery at Shanghai Pulmonary Hospital from January 2005 to January 2015. All 32 patients were admitted in our department for inflammatory myofibroblastic tumor occurred in lung and bronchus.

Preoperative assessment

Preoperative assessments according to the guideline of Chinese Thoracic Society including thoracic computed tomography (CT), pulmonary function and bronchoscope were performed to establish the diagnosis of the single pulmonary lesions of inflammatory myofibroblastic tumor.

Surgical procedures

All patients accepted the curative surgical procedure of lobectomy, wedge resection, sleeve resection and bronchoplasty. All patients were placed in the lateral decubitus position with single-lung ventilation. According to the different locations of the tumors, different approaches for the procedures were performed. Left or right thoractomy were used in the middle or lower tracheal lesions which using the All lesions were resected entirely, with the frozen section confirming the negative surgical margins. The reconstruction of the bronchus was performed after the resection with end-to-end anastomosis with interrupted 4-0 Vicryl absorbable sutures (Ethicon, Inc., Somerville, NJ, USA). Those patients with lower tracheal tumor involved carina were performed the reconstruction with the "flap" technique, which had been described in the published literatures of our department [6,7].

Postoperative management

All lesions were confirmed the diagnosis of inflammatory myofibroblastic tumor by experienced pathologist postoperatively. No routine adjuvant chemotherapy or radiation therapy were conducted.

Postoperative follow-up was conducted by thoracic surgeons every 6 months until 12 months after surgery and after that yearly. Standard

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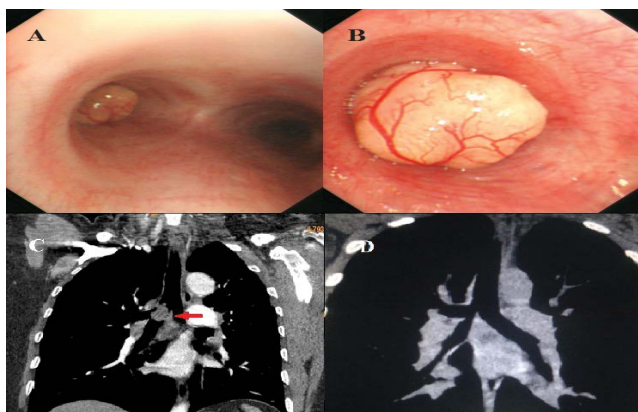


Figure 1: (A-D) 16-years-old female, IMT in the main bronchus of left lung. **A:** Bronchoscope showed the lesion in the main bronchus of Right lung. **B:** The lesion in the main bronchus of left lung. **C:** CT scan preoperatively showed the lesion in the main bronchus of Right lung (red arrow). **D:** CT scan postoperatively showed that the left main bronchus is clear.

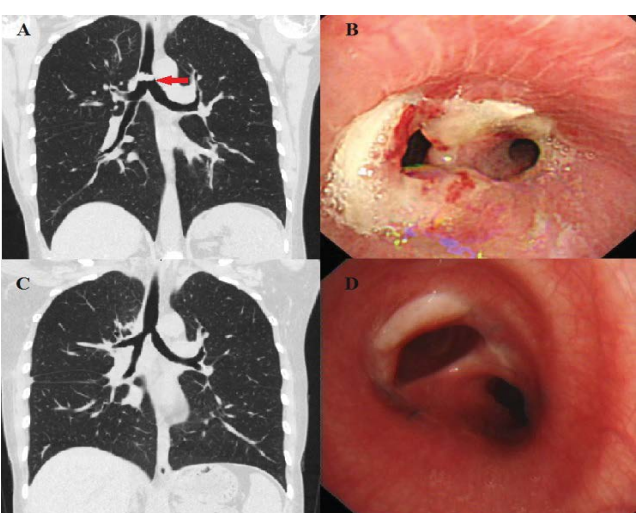


Figure 2: (A-D) 37-years-old female, IMT in the trachea. **A:** CT scan showed the lesion in the lower segment of the trachea (red arrow). **B:** The bronchoscope revealed a mass in the trachea. **C:** CT scan 6 months after surgery suggested that the lesion had been removed. **D:** The anastomosis revealed under bronchoscope 6 months postoperatively.

follow-up consisted of chest X-ray or CT, including bronchoscope and clinical examination.

Results

32 patients (aged: 9-72 yr, average: 37.85 ± 19.42 yr, 13 males and 19 females) received surgical treatment for inflammatory myofibroblastic tumor (ranged: 0.8-8 cm, average: 3.84 ± 2.05 cm), including 15 cases of lobectomy, 7 cases of wedge resection, 4 cases of bronchoplasty (Figures 1A-1D), 2 case of tracheal tumor resection (Figures 2A-2D), 3 cases of sleeve resection and 1 case of wedge resection with biopsy of the chest wall. All patients were followed up till October 2017 (average: 81.52 ± 35.84 months, median: 78 months). 31 patients are still alive, while one patient die 6 months after the surgery.

The dead patient is a 72-years-old male. CT scan showed a 3 cm lesion on the right lower lobe and a mass on the right chest wall. Wedge resection of the pulmonary lesion and biopsy of chest wall mass

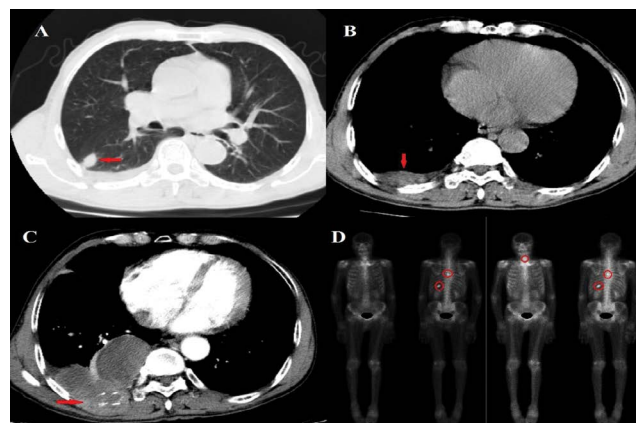


Figure 3: (A-D) The primary and metastatic lesions of the dead patient. **A:** CT scan revealed a 1.5 cm nodule in the right lower lobe (red arrow). **B:** CT scan revealed a 2 cm pleural lesion with pleural effusion (red arrow). **C:** CT scan showed a big mass in the right chest wall with 7th costal invasion which was considered as recurrency of IMT (red arrow). **D:** Bone scan revealed that multiple metastatic lesions on the 7th costa and vertebrae (red circled).

were performed for advantage age and bad physical condition. The pathology results suggested both lesions were IMT. 4 months later, the patient was admitted to our department for aggravated pain and short of breath. CT scan showed a big mass in the right chest wall with 7th costal invasion which was considered as recurrency of IMT. Bone scan revealed that multiple metastatic lesions on the 7th costa and vertebrae. The patient received radiation therapy after that and dead for cachexia and MODS 6 months postoperatively (Figures 3A-3D).

Discussion

Inflammatory myofibroblastic tumor was described in lungs by Brunn in 1939 [8]. The IMT is a rare benign lesion representing 0.7% of all lung tumors. It was previously called inflammatory pseudotumor, plasma cell granuloma, histiocytoma or fibroxanthoma [9,10]. Matsubara et al. reviewed a series of 32 patients and proposed a classification into three groups: organizing pneumonia, fibrous histiocytoma and lymphoplasmacytic [11]. IMTs may present respiratory complaints, such as cough, chest pain, shortness of breath and hemoptysis, also general complaints including fatigue, joint pains, fever, loss of appetite and weight loss. IMTs are found in CT scan as solitary masses with heterogeneous density, smooth and lobulated contours in the lungs or may see as masses with pneumonia looking. Calcifications are particularly found in children.

IMTs used to be regard as a benign tumor. As more cases of metastasis were reported, IMT is now reclassified as a borderline tumor with an aggressive biological behavior. Several cases reports had suggested poor prognosis of multiple lesions of IMT, especially in Brain. Literatures published suggested that if the complete resection of the lesion cannot be achieved than metastasis may occurred, and it may lead to a compromised outcome [12,13]. Also, some reports supported that if the radical resection of the multiple lesions can be accomplished, a promising outcome may achieve [14]. Carolina et al. reported that after the wedge resection of bilateral inflammatory myofibroblastic tumors of the lung along with resection of metastatic lesion on the adrenal gland, patients got a curative result.

Conclusion

In our study, 32 patients with pulmonary IMTs received surgical

treatment in our department. 31 patients received complete resections of the IMT lesion and all of them got a long-term disease-free survival with no recurrence or metastasis. It was reported that the prognosis of IMT is usually good, but it depends on tumor size (less than or equal to 3 cm) and complete resection. The overall 3-year survival rate is about 82% and the overall 5-year survival rate is about 74%. Local recurrence rate of pulmonary IMT after resection was about 6.6% to 13%, especially in those incomplete resection cases [15]. In this study, 13 patients who received complete resection of pulmonary IMT obtained a promising disease-free survival with a long-term surveillance. It may suggest that complete resection of lesion is the prerequisite condition for a better survival. 4 patients undergone wedge resection also obtained good survival. Lobectomy, sleeve resection and bronchoplasty were performed for complete resection with maximal pulmonary function preservation. One patient dead 6 months after incomplete resection because of high age and bad physical condition. The disease advanced aggressively 4 months after surgery. The mass of the chest wall enlarged to 6 cm with pleural effusion. Metastatic lesions on the left lung and cervical vertebra were detected. Radiation therapy showed no efficiency on the IMT. The patients finally dead for cachexia and MODS 6 months postoperatively.

In our experiences, complete resection of IMT is the key point of the treatment. Since no evidence supported corticosteroids administration, chemotherapy or radiations for IMT, surgical procedure is the first choice for the patients. According to the location of the lesions, complete resection with perfect surgical strategy, the patients may obtain a promising long-term survival.

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