

Operated pulmonary inflammatory myofibroblastic tumors: Our experience with 17 cases

Ameliyat edilen pulmoner enflamatuvar miyofibroblastik tümör: 17 olguluk deneyimimiz

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ABSTRACT

Background: In this study, we aimed to evaluate the clinicopathological features of pulmonary inflammatory myofibroblastic tumor cases operated in our clinic.

Methods: A total of 17 inflammatory myofibroblastic tumor patients (5 males, 12 females; median age: 46 years) who were operated in our clinic between February 2000 and July 2019 were included. Data including sex, age, symptoms, accompanying diseases, tumor localization, tumor diameter, endobronchial extension, maximum standard uptake value of the tumors, surgery type, recurrence, and survival data were analyzed.

Results: Two patients were diagnosed preoperatively and two patients were diagnosed during surgery using frozen-section method before resection. Three (17.7%) patients underwent pneumonectomy, five (29.4%) patients lobectomy, three (17.7%) patients segmentectomy, five (29.4%) patients wedge resection, and one (5.8%) patient bronchial sleeve resection. All patients had complete resection with negative margins. None of them had lymph node metastasis. Median follow-up was 122 (range, 8 to 245 months) months. None of the patients received adjuvant therapy, there was no tumor recurrence or tumor-related death.

Conclusion: It is difficult to make a preoperative diagnosis of inflammatory myofibroblastic tumor patients. Systematic lymph node dissection is not required in diagnosed patients. Complete resection is the most important prognostic factor, and it is critical to achieve this with the smallest resection possible.

Keywords: Case series, inflammatory myofibroblastic tumor, rare pulmonary tumors.

ÖZ

Amaç: Bu çalışmada kliniğimizde ameliyat edilen pulmoner enflamatuvar miyofibroblastik tümörlü olguların klinikopatolojik özellikleri değerlendirildi.

Çalışma planı: Kliniğimizde Şubat 2000 - Temmuz 2019 tarihleri arasında ameliyat edilen toplam 17 enflamatuvar miyofibroblastik tümörlü hasta (5 erkek, 12 kadın; medyan yaş: 46 yıl) çalışmaya dahil edildi. Cinsiyet, yaş, semptomlar, eşlik eden hastalıklar, tümör lokalizasyonu, tümör çapı, endobronşiyal uzanım, tümörlerin maksimum standart tutulum değeri, ameliyat türü, nüks ve sağkalım verileri dahil olmak üzere veriler analiz edildi.

Bulgular: İki hastaya ameliyat öncesi ve iki hastaya ameliyat sırasında donuk kesit yöntemi ile rezeksiyon öncesi tanı konuldu. Üç (%17.7) hastaya pnömonektomi, beş (%29.4) hastaya lobektomi, üç (%17.7) hastaya segmentektomi, beş (%29.4) hastaya kama rezeksiyonu ve bir (%5.8) hastaya bronşiyal sleeve rezeksiyonu yapıldı. Tüm hastalarda negatif sınırlarla tam rezeksiyon sağlandı. Hiçbirinde lenf nodu metastazı saptanmadı. Medyan takip süresi 122 (dağılım, 8-245) ay idi. Hiçbir hastaya adjuvan tedavi verilmedi, tümör nüksü veya tümör ile ilişkili ölüm izlenmedi.

Sonuç: Enflamatuvar miyofibroblastik tümörlü hastalara ameliyat öncesi tanı koymak zordur. Tanısı olan hastalarda sistematik lenf bezi diseksiyonuna gerek duyulmadı. Tam rezeksiyon en önemli prognostik faktör olup, bunu mümkün olan en küçük rezeksiyon ile sağlamak önemlidir.

Anahtar sözcükler: Olgu serisi, enflamatuvar miyofibroblastik tümör, nadir akciğer tümörleri.

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Inflammatory myofibroblastic tumor (IMT) is a rare neoplasm that can involve a wide variety of organs, such as the larynx, orbita, intra-abdominal organs, breast, soft tissue, and lungs. Lungs are the most common localization. To date, IMT has been called many different names, such as inflammatory pseudotumor, plasma cell granuloma, fibroxanthoma and fibrous histiocytoma.^[1] It is the most common lung tumor in children.^[2] In adults, it accounts for less than 1% of all lung tumors and is seen equally among men and women.^[3] While half of the patients are asymptomatic, symptoms such as cough, chest pain, fever and shortness of breath may be seen depending on the location and size.^[4] Its natural course and prognosis and, also, the most optimal treatment strategy has not been fully known, yet.

In the present study, we aimed to evaluate the clinicopathological features of pulmonary IMT cases operated in our clinic. To the best of our knowledge, this is the first case series of Turkish IMT patients in the literature.

PATIENTS AND METHODS

This single-center, retrospective study was conducted at Ankara University Faculty of Medicine, Department of Thoracic Surgery between February 2000 and July 2019. A total of 17 IMT patients (5 males, 12 females; median age: 46 years) who were operated in our clinic were included. Data including sex, age, symptoms, accompanying diseases, tumor localization, tumor diameter, endobronchial extension, maximum standard uptake value (SUV_{max}) of the tumors, surgery type, recurrence, and survival data were recorded. A written informed consent was obtained from each patient. The study protocol was approved by the Ankara University Faculty of Medicine Human Research Ethics Committee (date: 24.12.2020, no: İ11-689-20). The study was conducted in accordance with the principles of the Declaration of Helsinki.

All pathology slides were reviewed to confirm the diagnosis. On microscopic examination, predominant cell component (plasma cell type, fibrohistiocytic type or mix), cellularity, border of lesion (ill-defined or infiltrative); presence of endobronchial component, necrosis, mitosis and pleomorphism were recorded. Using the immunohistochemically stained slides at the time of diagnosis, smooth muscle actin (SMA) (n=15), desmin (n=13), cytokeratin (CK) (n=13), CD34 (n=10), S100 (n=12) expression in spindle cells were evaluated. The Ki67 proliferation index was calculated (n=7). Anaplastic lymphoma kinase (ALK) expression was

evaluated by immunohistochemistry in 15 patients with D5F3 (n=3) and ALK01 (n=12) clones. Only in one patient, ALK rearrangement was evaluated by fluorescence *in situ* hybridization (FISH) (Figure 1).

The follow-up time was defined as the time between curative surgery and final control. As there was no recurrence and only one patient died from an irrelevant cause, no survival statistics were performed.

RESULTS

The median follow-up was 122 (range, 8 to 245 months) months. During the study period, 1,952 patients were operated for lung tumors in our clinic, and 17 of them (0.87%) were diagnosed with IMT. There were only two patients in the pediatric age group (3 and 16 years old). The rate of patients under the age of 40 was 41%. One patient who underwent pneumonectomy due to right hilar IMT had a history of chemoradiotherapy after right mastectomy due to breast carcinoma 13 years previously. Nine (52.9%) patients had symptoms such as cough, back pain, and fever. Nine (52.9%) tumors were in the left, whereas eight tumors were in the right lung. There was a predominance of upper lobe localization (47% upper lobes, 29.4% lower lobes, 23.6% hilar and middle lobe). The mean tumor size was 35.4 ± 18.5 mm (range, 10 to 76 mm). Seven (41.1%) patients had endobronchial tumor extension and one (5.8%) patient had a totally endobronchial tumor. Both of the pediatric cases had endobronchial tumor extension. Eight patients had positron emission tomography (PET)/computed tomography (CT), and the mean SUV_{max} of the masses was 9.8 ± 5.4 (3.5-19.7). Eight patients had preoperative biopsy and only two patients had preoperative IMT diagnosis. One patient was diagnosed with bronchoscopic biopsy and one patient with an open lung biopsy in an external clinic. Ten patients had intraoperative frozen-section analysis from their masses and only two of them received an IMT diagnosis (four patients were reported as mesenchymal tumor and four patients as benign tumors). Three (17.7%) patients underwent pneumonectomy (one carinal sleeve pneumonectomy), five (29.4%) patients lobectomy, three (17.7%) patients segmentectomy, five (29.4%) patients wedge resection (one patient with laminectomy) and one (5.8%) patient bronchial sleeve resection. All patients had complete resection with negative margins. Eleven (64.7%) patients underwent lymphadenectomy due to uncertain intraoperative frozen-section analysis and none of them had lymph node metastases. There was no intraoperative mortality. None of the patients received adjuvant therapy and there was no tumor

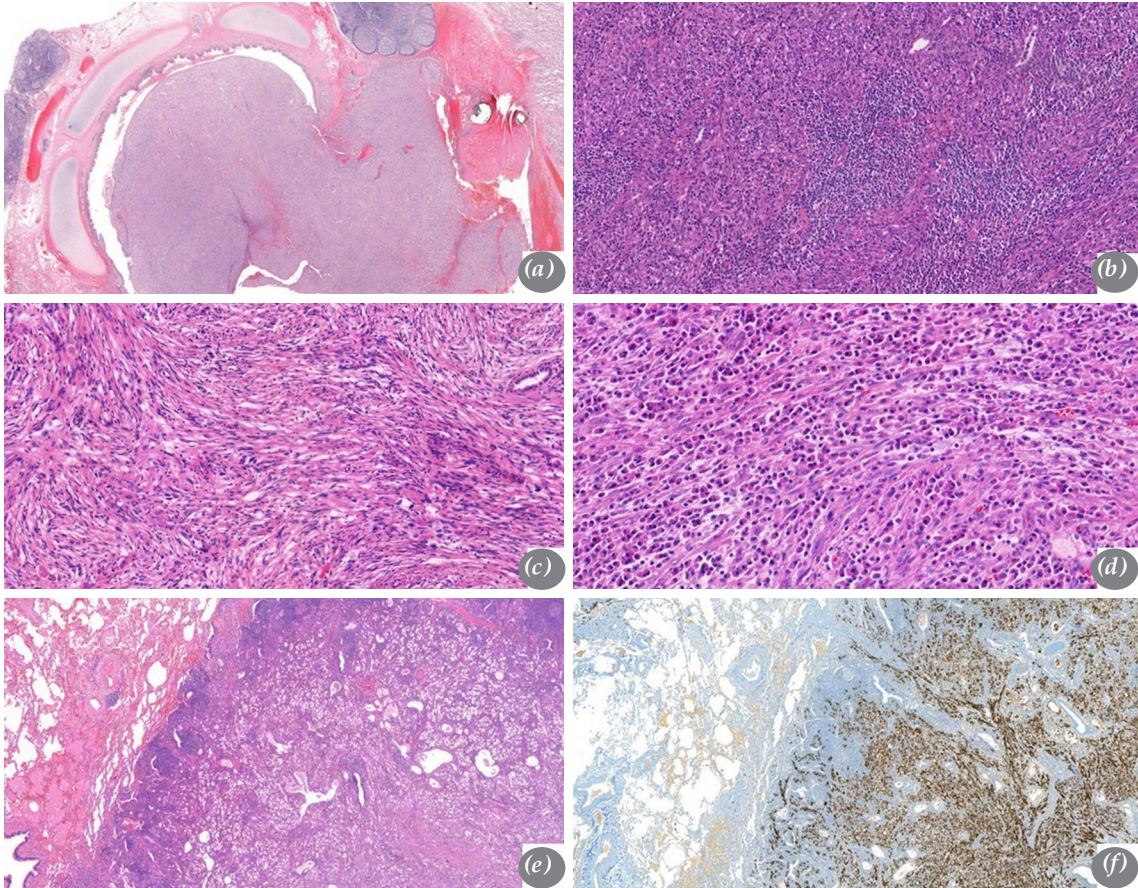


Figure 1. (a) Inflammatory myofibroblastic tumor within bronchial lumen (H-E, 0.9×). (b-d) Inflammatory myofibroblastic tumor mixed, fibrohistiocytic and plasma cell types, respectively (H-E, 10×, 10×, 20×). (e, f) IMT showing ALK positivity with immunohistochemistry clone ALK01(2×). Slides were scanned with digital scanner (Pannoramic 250 Flash III, 3DHISTECH Ltd., Hungary) in 40× objective and photographed with CaseViewer 1.4 (3DHISTECH Ltd., Hungary).

ALK: Anaplastic lymphoma kinase.

recurrence (Table 1). Only one patient (81-year-old) died seven years after the operation due to an unrelated cause.

In one patient who underwent pneumonectomy due to left hilar IMT, Takayasu arteritis was diagnosed postoperatively. A 16-year-old female patient (No. 7) had right thoracotomy with incomplete resection in another clinic. The tumor progressed under infliximab and radiotherapy. Finally, six years later from the initial diagnosis, she was referred to our clinic and right carinal sleeve pneumonectomy was performed. Currently, she is alive with no sign of the disease for 10 years.

In microscopic examination, all tumors were composed of spindle cells and inflammatory cells in variable distribution. Spindle cells showed fascicular pattern in some of the tumors. Necrosis, mitosis, and

pleomorphism were seen in 12.5%, 11.8% and 11.8% of the tumors, respectively. Predominant cell component was plasma cell in 23.5%, fibrohistiocytic in 41.2%, and mixed in 35.3% of the tumors. Cellularity was low in 23.5% and intermediate or high in 76.5% of the tumors. Two (11.7%, No. 6 and 16) patients had infiltrative tumors. Spindle cells were positive with SMA (64.3%), desmin (23.1%) and negative with CK, CD34 and S100. Ki67 index ranged from 1 to 10% with a median value of 5%. The ALK expression was noted in 28.6% of the tumors. The FISH was performed only in one ALK-positive tumor and this tumor had ALK rearrangement based on the FISH analysis.

DISCUSSION

Inflammatory myofibroblastic tumor is a borderline tumor of the lung which have the potential

Table 1. Patient characteristics of operated inflammatory myofibroblastic tumors in our clinic between 2000 and 2019

No	Age/Sex	Symptoms	Medical history	Localization	Diameter (mm)	Endobronchial extension	SUV _{max}	Frozen section analysis	Surgery	Follow-up (month)
1	34/F	-	-	Left upper lobe	40	+	-	-	Lobectomy	245
2	46/M	-	-	Left upper lobe	10	+	-	-	Bronchial sleeve resection	210
3	3/F	Cough	-	Left lower lobe	17	+	-	-	Lobectomy	186
4	42/F	Cough, fever	1992 right mastectomy and right side chemoradiotherapy	Right hilar	19	+	-	-	Pneumonectomy	186
5	36/F	Cough	-	Left lower lobe	17	-	7.3	Mesenchymal tumor	Lobectomy	155
6	49/F	Cough, side pain	Takayasu's arteritis	Left hilar, in the left main pulmonary artery	30	-	10.3	-	Intrapleural pneumonectomy	145
7	16/F	Dyspnea	Previous incomplete resection, infliximab use, radiotherapy	Right hilar	76	+	-	-	Carinal sleeve pneumonectomy	134
8	52/F	-	Hypertension	Left lower lobe	15	-	-	IMT	Wedge	133
9	59/F	Cough, chest and side pain	-	Right upper lobe	60	+	10.0	Mesenchymal tumor	Lobectomy	116
10	24/M	Cough, fever	-	Left upper lobe	40	+	-	-	Segmentectomy	120
11	39/F	-	-	Left lower lobe	30	-	-	Benign	Wedge	122
12	33/F	Cough	-	Right upper lobe	40	-	15.4	Mesenchymal tumor	Segmentectomy	112
13	54/M	-	-	Left upper lobe	60	+	19.7	Benign	Segmentectomy	53
14	55/M	-	-	Right upper lobe	26	-	4.4	Benign	Wedge	84
15	62/F	-	-	Middle lobe	24	-	8.2	IMT	Lobectomy	16
16	81/F	Cough, back pain, difficulty to walk	-	Right lower lobe, spinal canal extension	55	-	-	Mesenchymal tumor	Wedge+ laminectomy	91
17	46/M	-	-	Right upper lobe	20	-	3.5	Benign	Wedge	8

IMT: Inflammatory myofibroblastic tumor.

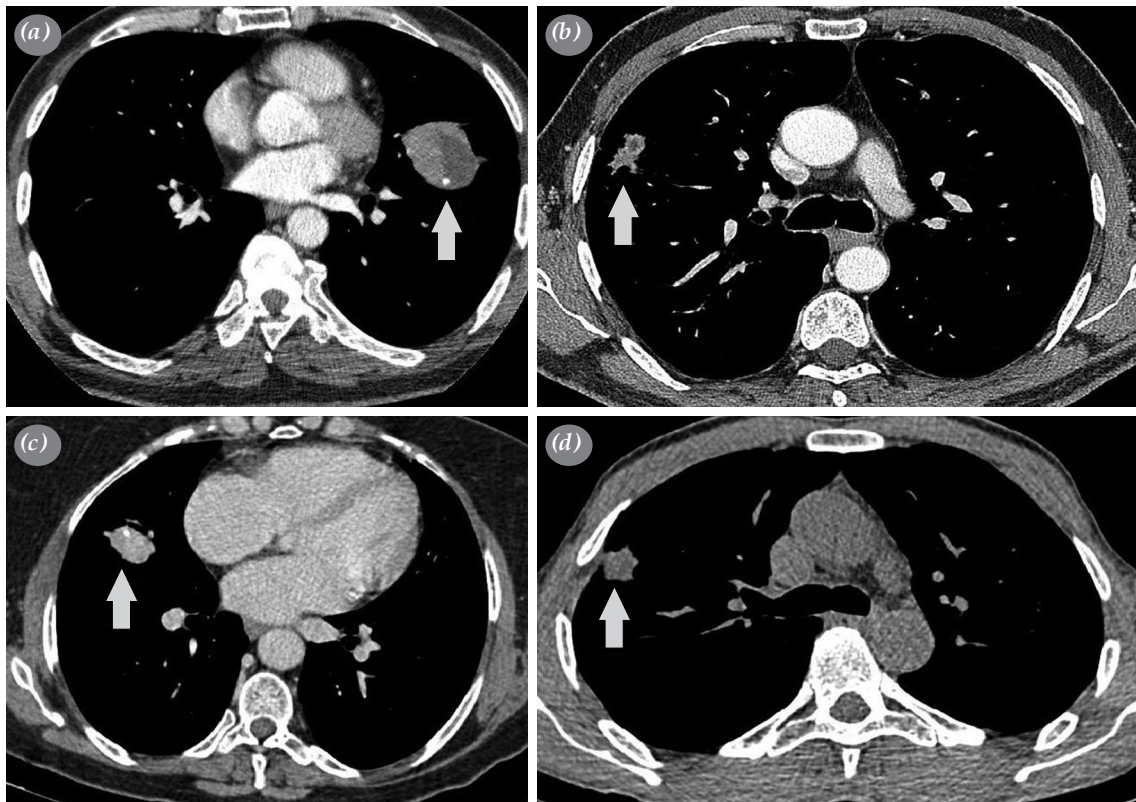


Figure 2. (a) Patient No. 13; left upper lobe mass with necrosis and calcification, (b) Patient No. 14; right upper lobe mass with irregular margins, (c) Patient No. 15; right middle lobe mass with calcification, (d) Patient No. 17; right upper lobe mass with pleural indentation. (Axial computed tomography images on mediastinal window, gray arrows show the masses).

for relapse and metastasis.^[5] It can occur in both pediatric and adult populations with a wide age distribution^[1] The incidence of the IMT among lung tumors in our series is 0.87%, compatible with the literature.^[3] The etiology and pathogenesis of IMT has not been still fully understood. Although IMT is usually seen as a well-limited peripheral lesion, it may also occur centrally, may be locally invasive or may be seen as pneumonic infiltration. It can be observed as single or multiple masses.^[5] One (5.8%) patient had spinal canal extension of the tumor as local invasion and all the tumors were presented as single masses in our series. The tumor can be endobronchial in 10 to 20% of the cases and also be localized within trachea.^[6,7] In our series, only one (5.8%) patient had a totally endobronchial tumor. Calcification and cavitation can be seen at a rate of 10%. Mediastinum, chest wall, and pleural invasion are rare (Figure 2).^[8]

In previous studies including 10 or more cases, 41.7% of the patients are females and the mean age is 40.2 ± 11.4 years. In the present study, 70.5% of cases

were females and the mean age was 43 ± 18.3 years. Mean tumor diameter was 38.4 ± 6 mm and 24.5% of tumors were centrally located in the previous literature, whereas mean tumor diameter was 35.4 ± 18.5 mm and 17.6% of tumors were centrally located in this study. One of our patients (No. 6) had a tumor in the left main pulmonary artery. A total of 56% of the patients were symptomatic in the literature and, similarly, 52.9% of our patients were symptomatic. Recurrence was observed in five patients who underwent complete resection and the mean follow-up was 73.8 ± 40 months in the publications (Table 2).^[2-6,8-14] In the present study, the median follow up was 122 months and there was no recurrence.

An 81-year-old patient (No. 16) had walking difficulty, and the tumor had spinal canal extension in this patient. Mass excision was made with wedge resection to the lung and for spinal canal extension by laminectomy. This case is a good example of the extraparenchymal extension of IMT. Takayasu arteritis was detected in the Patient No. 6 after surgery. Although there is no proven relationship between the

Table 2. Case series of inflammatory myofibroblastic tumor including over 10 patients

Author	Patient number	Sex female (%)	Age (mean)	Mean tumor diameter (mm)	Localization (central/peripheral; % central)	Symptoms symptomatic (%)	Histological subtypes	Recurrence	Mean Follow-up (month)
Bahadori and Liebow ^[2]	40	62.5	28.6	42	42.5	40	25 plasma cell	-	40
Spencer ^[9]	27	40.7	36		11.1	51	2 malignant histiocytoma 25 plasma cells/histiocytoma	-	
Matsubara et al. ^[10]	32	51.7	50		12.5	40.6	14 organizing pneumonia 14 fibrous histiocytoma 4 lympho-plasmacytic	1	
Pettinato et al. ^[11]	20	55	26.2	40	20	45	plasma cells	1	44
Agrons et al. ^[8]	61	40.9	28	44	14.7	54	17 fibro-histiocytic	-	
Cerfolio et al. ^[12]	23	52.1	47	40	30.4	78.2	13 organizing pneumonia 11 noninvasive 12 invasive	-	156
Kim et al. ^[13]	28	21.4	37.9	48	21.4	85.7		1	
Melloni et al. ^[3]	18	27.7	57.8	35	11.1	44.4	13 fibrous histiocytic 4 lympho-plasmacytic 1 organizing pneumonia	-	63
Lee et al. ^[6]	15	33.3	31.3	27	46.6	86.6		1	32.4
Chen et al. ^[14]	19	21	53.9	32.8	36.8	42.1	15 organized pneumonia 2 fibrous histiocytic 2 lympho-histiocytic	-	78.3
Fabre et al. ^[5]	25	56	33	35.4	36	52	16 inflammatory myofibroblastic 8 fibrous histiocytic 1 plasma cell granuloma	1	80
Peretti et al. ^[4]	36	38.8	53.5	40	11.1	61.1		-	96.9
<i>Present case series</i>	17	70.5	43	35.4	17.6	52.9	4 plasma cell type 7 fibrohistiocytic type 6 mix type	-	122

two conditions, we believe that this association may be supportive of the link between chronic inflammation and IMT development.

Furthermore, IMT is composed of myofibroblastic spindle cells, which are arranged in fascicular pattern, with eosinophilic cytoplasm and ovoid vesicular nuclei. Mitotic rate is variable, nuclear atypia is absent or minimal.^[11] Spindle cells are associated with a plasma cell predominant chronic inflammatory infiltrate. Foamy histiocytes, multinucleated giant cells, or neutrophils can accompany to the infiltrate. Spindle cells can show positivity with vimentin, SMA, desmin, muscle-specific actin (MSA), and focal positivity with epithelial markers such as CK and epithelial membrane antigen (EMA). Half of IMTs have ALK gene rearrangement and spindle cells can show positivity with ALK antibody in immunohistochemical analysis. Histopathological subtypes varied significantly due to the different classifications used. The most commonly preferred classification of IMT is based on predominant cellular component as fibrohistiocytic type and plasma cell type; however, histopathological classification has no prognostic value.^[15] In this study, predominant cell component was fibrohistiocytic in the majority of tumors (41.2%), and two (11.7%) patients had microscopically infiltrative type tumors, but all the patients had excellent outcomes independent of their histopathological types.

It is quite difficult to obtain a definite preoperative diagnosis in IMT.^[12] Therefore, routine examinations performed before lung surgery should be requested. It would be appropriate to perform blood tests, pulmonary function tests, cranial CT, thoracic CT and PET/CT. Frozen section is important both to clarify the diagnosis and to determine the width of resection intraoperatively. In Peretti *et al.*'s^[4] study, 35 patients underwent intraoperative frozen-section analysis and only five (14.2%) patients received IMT diagnosis. In our series, we had 10 frozen-section analysis cases and only two (20%) of them were reported as IMT. If there is a need for extended resection or pneumonectomy and the frozen section analysis is indeterminate, a second surgery after definitive diagnosis may be plausible. Of note, IMT may mimic inflammatory reactions and hematomatous proliferations. Microscopic features, immunohistochemical and molecular profile can help in the differential diagnosis. Spindle cell proliferation admixed with inflammatory cells and positivity of spindle cells with SMA, desmin, MSA showing myofibroblastic

differentiation are diagnostic clues. The ALK expression or ALK gene rearrangement in the spindle cells support the diagnosis of IMT.^[16]

No significant difference was observed in survival and recurrence in 17 patients who underwent major (pneumonectomy, lobectomy) or minor (segmentectomy, wedge resection) resections in the case series of Chen *et al.*^[4] This result is also consistent with the results of our study. Wedge resection is sufficient in small and peripheral tumors; for larger and aggressive tumors, lobectomy, pneumonectomy or extended resections can be performed. Complete resection is the most important prognostic factor and incomplete resection may result in tumor recurrence as seen in one of our patients (No. 7). Although there are reports about spontaneously disappearing tumors or good response to steroid therapy, main method of treatment is surgery.^[13] Radiotherapy can be considered for incomplete resections or inoperable cases. Lymph node metastasis was not observed in our patients, similar to the aforementioned series. It is not necessary to make a systematic lymph node dissection for IMT patients, if a pre- or intraoperative diagnosis of IMT can be achieved.

There are two major limitations for this study. This is a retrospective case series study and as there is one exitus and no recurrence in the study population, survival analysis could not be done.

In conclusion, it is difficult to make a preoperative diagnosis of inflammatory myofibroblastic tumor. Systematic lymph node dissection is not required in diagnosed patients. Complete resection is the most important prognostic factor, and it is of critical importance to achieve this with the smallest resection possible. Further studies are warranted to confirm these findings.

Declaration of conflicting interests

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