

Surgical treatment outcomes of pulmonary inflammatory myofibroblastic tumors

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Abstract:

BACKGROUND: Pulmonary inflammatory myofibroblastic tumor (PIMT) is an extremely rare disease. The aim of this study was to share the surgical outcomes of these tumors.

METHODS: Patients who were operated for pulmonary myofibroblastic tumors between January 2005 and January 2021 were determined by retrospectively scanning patient files. Patients' demographic characteristics, tumor location, surgical techniques, and other parameters were obtained from the patient files. The KaplanMeier method was used for survival calculations, whereas the log-rank test was used for comparison of survival calculations.

RESULTS: PIMTs were noted in 14 patients (0.12%) in a total of 11,108 thoracic procedures performed in our institution between January 2005 and January 2021. The mean age of the patients was 28.2 (range: 2–67) years. Of the patients, six were male and eight were female, with 50% ($n = 7$) aged under 18 years. A total of 17 surgical procedures were performed on 14 patients. One patient underwent pneumonectomy, two patients lobectomy, ten patients wedge resection, and one patient underwent debulking surgery. A total of 11 patients had complete surgery, whereas three patients had incomplete surgery. The 10-year overall survival was 84.6% and the 10-year disease-free survival (DFS) was 75.0%. Complete resection was found to be the only and significant factor that had an effect on survival ($P = 0.004$) and DFS ($P = 0.012$).

CONCLUSION: PIMTs are extremely rare. Complete surgery should be considered an effective factor in survival and DFS.

Keywords:

Lung neoplasm, myofibroblastic tumor, pulmonary inflammatory myofibroblastic tumor, surgical treatment, survival

Pulmonary inflammatory myofibroblastic tumor (PIMT), first described by Burnn in 1937, is a rare disease. Although it has been described in many different locations such as retroperitoneum, abdomen, larynx, and eyes, the primary site of inflammatory myofibroblastic tumor (IMT) is the lungs. However, they account for only 0.04%–0.2% of all lung tumors.^[1,2]

IMT contains varying proportions of inflammatory cells including histiocytes, lymphocytes, and plasma

cells histopathologically combined with myofibroblasts. Due to this cellular component diversity and the differences in its dominance, many different terms such as xanthogranuloma, inflammatory pseudotumor, and fibrous histiocytoma were used to describe IMT in the past.^[3]

In the 1st year of its definition, IMT was considered to be a benign entity. On the other hand, IMT is currently considered a low-grade malignant tumor due to the demonstration of its local recurrence, invasion of surrounding tissues, and metastasis. Although there are medical treatment options for PIMT such as

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chemotherapy, radiotherapy, and steroids, the key element of treatment is surgery.^[4,5]

The aim of this study was to share the surgical outcomes of these tumors.

Methods

Patients who were operated for pulmonary myofibroblastic tumors in our clinic between January 2005 and January 2021 were retrospectively scanned and determined. Patients' demographic characteristics, tumor location, diagnostic methods, tumor size, surgical technique, and recurrence information were obtained from the patient files.

Regardless of age, the study included all patients (pediatric and adult) with accessible records who were treated surgically for PIMTs. All patients underwent chest X-ray and chest computed tomography (CT). Positron emission tomography (PET)/CT and magnetic resonance imaging (MRI) were used for screening of tumor invasion and metastasis.

As a diagnostic methodology, patients were attempted to be diagnosed using the more noninvasive techniques of bronchoscopy and transthoracic biopsies whenever possible. Those for whom no diagnosis was established were diagnosed using open lung biopsies. Video-assisted thoracic surgery (VATS) and thoracotomy were used as surgical techniques.

In surgical procedures, complete resection was considered R0 surgeries. All R1 and R2 patients were considered to achieve incomplete surgery. As a surgical resection, patients were first scheduled for wedge resection. More severe anatomic resections such as lobectomy and pneumonectomy were preferred in cases where a safe surgical margin could not be achieved due to anatomical location and invasion. Debulking surgery was preferred only for those with unresectable tumors and severe cardiac and pulmonary compression symptoms associated with tumor size.

While patients who achieved complete surgery after surgical treatment received no treatment, chemoradiotherapy was recommended to all patients who had incomplete surgery. All patients undergoing surgery, for whom no routine follow-up protocol has been reported in the literature, were followed up with chest X-ray/CT at 6-month intervals for at least 2 years. All patients with recurrence were evaluated radiologically (CT and PET/CT, MRI), and those without distant metastases were evaluated for surgical eligibility. Oncological treatment was prescribed for patients who were not eligible for surgery. In all of the second surgeries, the thoracotomy approach was used for patients.

IMT is a type of tumor consisting of inflammatory cells (such as histiocytes, lymphocytes, and plasma cells) combined with varying proportions of myofibroblasts. In our study, this tumor was defined according to the World Health Organization (WHO) criteria.^[3] The fluorescence *in situ* hybridization method, which is the gold standard for detecting the EML4-ALK fusion gene, was used for the evaluation of ALK.

Whether the patients with available data were alive or not and the time of death of those who died were determined from the national death reporting system. Data analysis was carried out using IBM SPSS Statistics (version 22.0, Armonk, NY: IBM Corp., USA). The time from surgery to death or the last follow-up time was taken into account for the overall survival (OS) calculation. Disease-free survival (DFS) was calculated based on postsurgical recurrence, death, or last follow-up period, whichever occurred first. While the KaplanMeir method was used for survival calculations, the long-rank test was used for comparison of results.

Results

A total of 14 patients were identified. The mean age of the patients was 28.2 (range, 2–67) years. Of the patients, 6 were male (42.9) and 8 were female (57.1), with 50% (7) aged under 18 years. While 28.6% of the patients were asymptomatic, the most common symptom was dyspnea (35.7%), followed by cough with 28.6% [Table 1].

As a surgical approach, 6 out of 14 patients underwent VATS in their initial surgery, while the thoracotomy approach was used for eight patients. Thoracotomy was the preferred surgical approach for re-resections, debulking surgery, and pediatric patients. One patient had an indication for pneumonectomy due to a hilar tumor invading the left main bronchus, while two pediatric patients had an indication for lobectomy because of a large tumor diameter (7 cm and 8 cm).

A total of 17 surgical procedures were performed on 14 patients. One patient underwent pneumonectomy, ten patients underwent wedge resection, whereas patient number 5 underwent debulking surgery twice to reduce cardiac pressure. A total of two patients underwent lobectomy, but patient number 13 underwent thoracotomy three times, first left upper lobectomy, 10 months later left lower lobe wedge resection, and 11 months later right wedge resection. The radiological evaluation at admission revealed total left lung collapse in patient number 8 [Figure 1] and severe mediastinal and vascular invasion in patient number 5. Complete surgery was performed on 11 (78.5%) patients, whereas incomplete surgery was performed on three patients. Of

Table 1: Clinical characteristics of patients

Case number	Age	Sex	Symptom	Location	Tumor size	Diagnostic method	Surgery
1	49	Female	Asymptomatic	RU	10×10	WG	WG
2	65	Female	Dispine	RU	25×35	TTBx	WG
3	32	Female	Dispine	LL	30×35	WG	LP
4	67	Male	Cough	RU	15×15	TTBx	WG
5	40	Male	Hemoptysis	RU	172×177	WG	DB
6	67	Male	Cough	RL	67×78	TTBx	WG
7	2	Male	Asymptomatic	LU	15×15	WG	WG
8	8	Male	Dispine	LU	44×23	RJB	WG
9	8	Female	Dispine	LU	20×10	RJB	WG
10	20	Female	Cough	LL	45×35	WG	WG
11	10	Female	Asymptomatic	LL	10×8	WG	WG
12	12	Male	Asymptomatic	RL	13×15	WG	WG
13	6	Female	Dispine	LU	80×85	WG	LUL*, LLWG**, RUWG***
14	10	Female	Cough	LL	70×35	WG	LUL

*First surgery, **Second surgery, ***Third surgery. RU=Right upper lobe, RL=Right lower lobe, LU=Left upper lobe, LL=Left lower lobe, WG= Wedge resection (using a stapler or satinsky clamp), LUL=Left upper lobectomy, LLWG=Left lower wedge resection, RUWG=Right upper wedge resection, LP=Left pneumonectomy, DB=Debulking surgery, RJB=Rigid bronchoscopy, TTBx=Trans thoracic biopsy

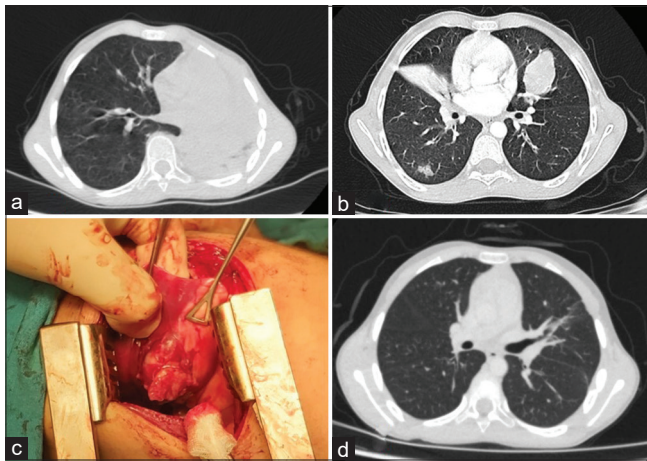


Figure 1: (a) Presentation with left total atelectasis, (b) the mass was detected after rigid bronchoscopy, (c) intraoperative appearance, (d) postoperative control computed tomography

the lesions, six were located in the right lung (four in the upper lobe and two in the lower lobe) and eight were located in the left lung (four in the upper lobe and four in the lower lobe). Six patients had a tumor size <3 cm in diameter, whereas 8 patients had a tumor size >3 cm in diameter. While ALK was positive in six patients, it was negative in eight patients.

Three patients were diagnosed by transthoracic biopsy and two patients by rigid bronchoscopy, but the remaining patients could only be diagnosed intraoperatively. After the atelectasis disappeared following rigid bronchoscopy in the patient with left total atelectasis, a lesion was noted in the left upper lobe [Figure 1]. The granulation-myofibroblastic tumor differentiation of the patient could only be made intraoperatively.

A total of four patients received chemotherapy; one of these patients received chemotherapy for breast cancer

and the other for multiple myeloma. Two patients who underwent incomplete resection for myofibroblastic tumor received chemotherapy, while one patient refused chemotherapy. Vinorelbine and crizotinib were used as chemotherapeutic agents. None of the patients received steroid therapy.

During the follow-up period, two of the 14 patients died. All remaining patients were alive, including the patient who underwent thoracotomy 3 times. The 5 and 10-year OS of the patients with a mean follow-up period of 56.7 months was 84.6%, whereas the 5- and 10-year DFS was 75.0% [Figure 2]. The comparison of the groups with tumor size >3 and <3 cm by the log-rank test for survival analysis showed no difference between the groups [$P=0.17$; Figure 3]. Re-thoracotomy also had no effect on survival ($P=0.19$). Similarly, ALK positivity also had no effect on survival ($P=0.11$). However, complete resection was found to be the only and significant factor that had an effect on survival ($P=0.004$) and DFS ($P=0.012$) [Figure 3].

The correlation analysis revealed that there was no correlation between tumor diameter and complete-incomplete surgery ($P=0.104$), and nonanatomical resections were not a prognostic factor ($P=0.284$).

When evaluating the statistical results, it should be taken into account that the study population consisted of a small group of patients. Future studies with larger patient groups may find a correlation, especially because the vector difference between ALK and survival is lower.

Discussion

PIMTs were first described by Bruno in 1939.^[1] Although IMTs have been reported in many different organs such as the retroperitoneum and abdomen, the primary site is

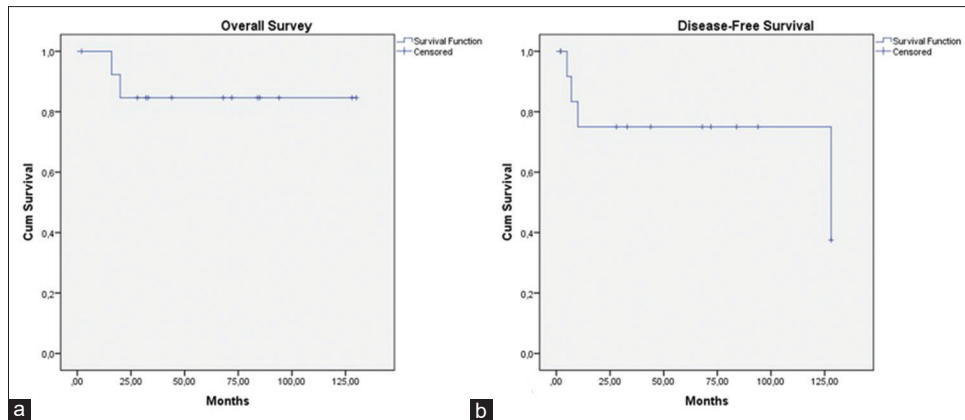


Figure 2: (a) Ten-year overall survival was detected as 84.6%, (b) 10-year disease-free survival was determined as 75.0%

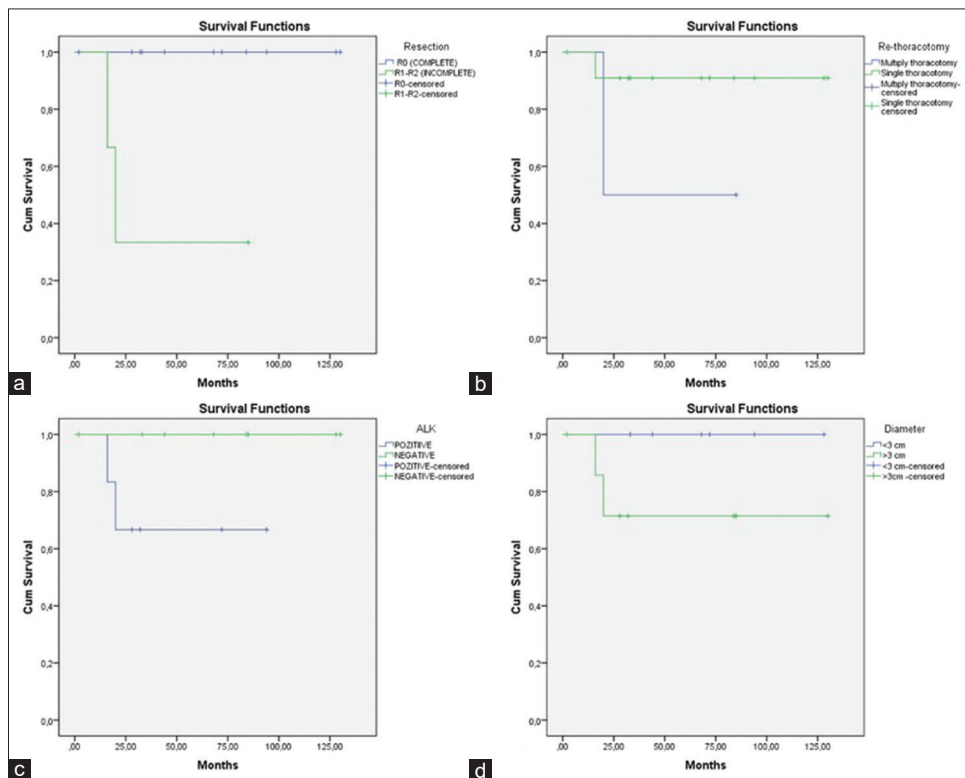


Figure 3: Log-rank test results in overall survival calculations, effect of (a) complete surgery ($P = 0.004$), (b) re-thoracotomy ($P = 0.19$), (c) anaplastic lymphoma receptor tyrosine kinase positivity ($P = 0.11$), (d) tumor diameter ($P = 0.17$)

the lungs, and they are more common in children than in adults.^[2]

Although the actual incidence is not exactly known, Cerfolio *et al.* found an incidence rate of 0.04% with 23 patients in a total of 56,400 thoracic surgery procedures performed in the Mayo Clinic, whereas Melloni *et al.* reported 13 patients (0.3%) in a total of 4120 thoracic procedures performed in his institution.^[6,7] In our study, 14 patients (0.12%) were identified in a total of 11,108 thoracic surgery procedures performed over 21 years.

The tumor structure is characterized by the participation of many inflammatory cells such as plasma cells, lymphocytes, and histiocytes accompanying myofibroblasts at variable proportions. Due to the different cell dominance in the inflammatory component, the disease has historically been described by various terms such as IMT, xanthogranuloma, inflammatory pseudotumor, and fibrous histiocytoma, leading to a definition confusion.^[3]

This confusion ended in 2002 when the WHO described the disease as an IMT.^[8] Moreover, although IMT was

considered to be a benign entity in the past, it was defined by the WHO as a low-grade tumor in 2006 with the data accumulated over time upon the demonstration of its invasion and metastasis, as in our two patients.^[9]

In terms of age distribution, PIMTs mostly occur in the age range of 27–50 years.^[6,7,10] When the literature is reviewed, the age range varies depending on the inclusion of pediatric patients in the centers where the studies are conducted. Seven (50%) of our patients were 18 year–old or younger. While this rate was 26.7% in the series of 15 patients by Lee *et al.*, 6 (26%) of 23 patients were under 18 years of age in the study by Cerfolio *et al.*^[6,10,11] Considering that PIMTs are the most common primary lung tumor in children, it is obvious that they are effective in both incidence and age distribution.^[12,13]

Approximately half of the patients with PIMT are asymptomatic and PIMTs are incidentally detected on chest radiographs. PIMTs may have an endobronchial location. In this case, symptoms such as dyspnea, cough, and rarely hemoptysis are more common.^[3,14,15]

This mesenchymal tumor is radiologically visualized as a single nodule or mass in the lung region. However, it can also be identified as more than one lesion in 5% of the cases. They typically present with lesions with well-circumscribed peripheral borders and sometimes contain calcifications.^[14,16] It has been shown to be of endobronchial origin in patients (0%–12%).^[17,18] It should be kept in mind that PIMTs can cause total collapse if they affect the main airway, as well as advanced invasions in patients with delayed diagnosis.

Due to the histopathological content of the tumor, the number of inflammatory cells is high and variable, which requires large tissue pieces for diagnosis. Therefore, rigid bronchoscopy may be preferred as a diagnostic procedure in eligible patients instead of transthoracic biopsy and fiberoptic bronchoscopy.^[15] Although there are exceptions,^[15] it is seen in the literature that the majority of patients can only be diagnosed intraoperatively.^[10,19] Although eight pieces with the largest one measuring 1.6 cm × 0.6 cm × 0.4 cm were collected by a rigid bronchoscope in patient number 6 in our institution, the differentiation between granulation and myofibroblastic tumor could only be made by intraoperative frozen section.

Different classifications have been described for IMTs by Cerfolio *et al.*, Matsubara *et al.*, and Colby *et al.* and eventually the WHO.^[3,6,20,21] However, these classifications could not be demonstrated to have any prognostic value. That is why they have lost their significance today.

Steroids, chemotherapy, nonsteroidal anti-inflammatory drugs, and radiotherapy have been defined for the

treatment of PIMTs. However, their effects on survival are controversial. The primary treatment method is surgery.^[19,22,23] There is no known standard treatment method for chemotherapy protocols due to the very limited number of patients. There are treatments with different agents such as methotrexate, vinorelbine, vincristine, cyclophosphamide, and doxorubicin. Furthermore, patients are attempted to be treated with different agents such as platinum pemetrexed.^[24] In recent studies, ALK receptor inhibitors such as crizotinib and ceritinib have been reported to be effective in IMTs, and the frequency of use of these agents is increasing, especially in inoperable and metastatic patients.^[25,26] It has been stated in new guidelines that the use of crizotinib, especially in ALK-positive patients, provides excellent disease control with acceptable toxicity.^[27] We used vinorelbine for one of our patients and crizotinib for the other.

While performing surgery, it is important to treat patients' lesions by considering the anatomical location under the guidance of surgical experience and preserving the parenchyma as much as possible. For this purpose, wedge resection is a very appropriate and common treatment modality for these patients. Local recurrence occurs in PIMTs. To avoid this, more major surgical procedures such as segmentectomy, lobectomy, and pneumonectomy can be safely performed when necessary. Sleeve lobectomy should be preferred for endobronchial locations.^[15,19,28] The important point is to perform complete resection.

There are very few publications in the literature regarding the survival in PIMTs, and most of them are the reports of small patient groups such as our patients. The studies of Cerfolio *et al.* with 23 patients, Melloni *et al.* with 18 patients, and Fabre *et al.* with 24 patients reported OS rates of 91%, 74%, and 89%, respectively.^[6,7,29] No mortality was reported in the studies of Thistlethwaite (11 patients) and Lee *et al.* (15 patients), with an OS rate of 100%.^[10,15]

In the literature, comparisons have been made for tumor size, recurrence, and survival. In his study, Melloni *et al.* reported that those with a tumor size of <3 cm in diameter achieved better survival.^[7] Although tumor size has been specified in studies on PIMTs in the literature other than this study, its direct correlation with survival has not been discoursed.^[6,10,15,29] In our study, we could not find any correlation between tumor size and survival.

In the history of IMTs, one of the conditions that caused this disease to be characterized as a neoplasm rather than an inflammatory condition is the identification of a number of chromosomal disorders (chromosome band 2p23) in these patients including the

anaplastic lymphoma receptor (ALK) tyrosine kinase gene locus. This chromosomal abnormality results in ALK overexpression.^[30,31]

Although we did not find any effect of ALK-positive pathological specimens on survival or DFS in our study, two of our six ALK-positive patients died. Tumor aggressiveness has been associated with ALK positivity in the past. However, the study by the European pediatric Soft-tissue Sarcoma Study Group evaluating the data of a total of sixty patients from nine different countries found no effect of ALK positivity on survival in IMTs.^[31] It should be remembered that the patients in this study consisted of the pediatric group, IMTs were not isolated in the thorax, and all tumor locations (head, neck, thorax, and abdomen) were included in the study.

Complete surgery is not always possible in PIMTs. Melloni *et al.* could achieve complete surgery in 13 patients (72%), Cerfolio *et al.* in 78%, and Sakurai *et al.* in 8 patients (89%).^[6,7,19] We were able to achieve complete resection in 11 of 14 patients (78.5%). In our study, complete surgery was found to be the only effective factor in OS ($P = 0.004$) and DFS ($P = 0.012$). In their studies, Cerfolio *et al.* and Melloni *et al.* stated that complete resection had an effect on mortality.^[6,7] Thistlethwaite, on the other hand, stated that complete surgery led to excellent outcomes in PIMTs located in the main airway.^[15] Discourses regarding the effect of complete surgery on survival are similar in other studies in the literature.^[10,29]

In the literature, there is no clear information on recurrent thoracotomies. What we could see was that in the study of Dartevela, thoracotomies performed for tumor recurrence had a negative effect on survival.^[29] However, it should be known that the patients included in this study were re-thoracotomy patients treated for incomplete surgery in the other centers and only one patient died. In our study, 17 surgical procedures were performed on 14 patients and re-thoracotomy had no effect on survival ($P = 0.190$). Both these statistical data and the fact that the patient who underwent thoracotomy three times, one of which was performed on the opposite lung, is still alive, has encouraged us more to consider re-thoracotomies in PIMTs.

Conclusion

The agents defined in the current medical treatment are still preferred for incomplete surgeries and metastatic patients. Although ALK positivity gives a clue about the more aggressive course of the disease, it has not been shown to have an effect on survival including our study. The most definite result on PIMTs, which has been accumulated over time and confirmed by current papers,

is that the primary treatment method of the disease is surgery. Apart from this, complete surgical treatment is the most effective factor in survival.

VATS resection can be safely performed on PIMTs by experienced hands, just as in other lung tumors. Considering that this patient group may have severe invasions and some of the patients are from the pediatric group, VATS should be preferred for eligible patients for reasons such as short length of hospital stay, less postoperative pain and associated complications, and rapid mobilization.

Both our study and other studies in the literature could not demonstrate an obvious effect of ALK positivity on DFS and survival. Nevertheless, we are of the opinion that this group of patients requires particular attention in terms of postoperative recurrence and metastasis, and therefore, a more strict and long-term follow-up of these patients is appropriate.

Limitation

The major limitation of the study is the small sample size. This reduces the reliability of the results in statistical analyses. It causes problems especially in advanced statistical evaluations such as Cox analysis. In our study, the log-rank test revealed the complete surgery as the only effective factor in survival, but this could not be confirmed in the Cox analyses. This statistical evaluation problem arises from the fact that almost all studies in the literature, including ours, were conducted with a maximum of 25 patients. We are of the opinion that if studies on PIMTs could be conducted with a higher number of patients, more parameters affecting survival would be determined.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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