Predictors of survival in lung torsion: A systematic review and pooled analysis

Jie Dai, PhD, Dong Xie, MD, Haifeng Wang, MD, Wenxin He, MD, Yiming Zhou, MD, Luis Angel Hernández-Arenas, MD, and Gening Jiang, MD

ABSTRACT

Objective: Lung torsion (LT) is a rare but life-threatening event. The objective of this study was to systematically review the natural history and clinical outcome of LT in published studies.

Methods: A review of publications on LT from January 1950 to December 2014 was performed using 3 databases (PubMed, EMBASE, and Web of Science). The primary efficacy outcome was LT-related mortality. Intervention was classified as direct resection (resection without detorsion), indirect resection (resection after detorsion), and reposition. Univariate comparisons of survival were performed using a logistic regression model.

Results: There were 109 patients from 91 studies identified in this analysis. LT was frequently reported after upper lobectomy (74.4%) and the middle lobe was the most vulnerable lesion (29.4%). The main clinical presentations were dyspnea (38.4%), fever (23.3%), and chest pain (17.4%). Radiologic findings suggestive of LT included worsening consolidation and abrupt truncation/tapering of the pulmonary artery. The overall mortality was 8.3%. Univariate analysis showed that the extent of torsion (whole LT vs lobar torsion) was significantly associated with survival (odds ratio, 5.867; $P = .017$). No significant difference was found between patients receiving direct resection and those receiving reposition; a trend was observed for worsening outcomes in patients treated with indirect resection (odds ratio, 5.300; $P = .060$).

Conclusions: The systematic review reveals the prevalence, key diagnostic tests, and optimal treatment methods for lung torsion. Whole LT is associated with higher mortality rates than lobar torsion. If the tortuous part is viable, reposition and direct resection have similar survival rates; otherwise, direct resection should be performed. (*J Thorac Cardiovasc Surg* 2016;152:737-45)

Time trend in lung torsion.

Central Message

Both direct resection and reposition can be performed in the treatment of lung torsion and the choice depends on tissue viability.

Perspective

Lung torsion is a very rare event. This study assesses the prevalence, key diagnostic tests, and optimal treatment methods for lung torsion. The middle lobe was the most vulnerable lesion. Whole lung torsion is associated with worse survival. If the tortuous part is viable, reposition and direct resection have similar survival rates. Otherwise, direct resection should be performed, instead of indirect resection.

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Lung torsion (LT), a very rare but life-threatening event, with a reported incidence of 0.089% to 0.4%,^{1,2} is defined as parenchymal rotation on the bronchovascular pedicle, accompanied by resultant airway obstruction and

vascular compromise.¹ Epplen and Jacobson³ first described LT in 1930. A survey of 117 thoracic surgeons in the United Kingdom revealed that only 30% had seen cases of LT.⁴ The treatment for LT has not been clearly elucidated. Reposition can retain pulmonary function and direct resection avoids a reperfusion insult.⁵ Indirect resection facilitates surgical operation by detorsing the lung but

From the Department of Thoracic Surgery, Shanghai Pulmonary Hospital, Tongji University, Shanghai, People's Republic of China.

J.D. and D.X. contributed equally to the article.

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Address for reprints: Gening Jiang, MD, Department of Thoracic Surgery, Shanghai Pulmonary Hospital Tongji University, No.507 Zhengmin Road, Shanghai, 200433, People's Republic of China (E-mail: jgnwp@aliyun.com).

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Abbreviations and Acronyms

ARDS	= acute respiratory distress syndrome
CI	= confidence interval
CT	= computed tomography
LT	= lung torsion
OR	= odds ratio
VATS	= video-assisted thoracoscopic surgery

may result in potential complications.⁶⁻⁸ Because of a low awareness of LT and little knowledge on its natural history, a comprehensive review to summarize current understanding is imperative. Therefore, we aimed to systematically review the clinical characteristics of LT, identify the risk factors for poor prognosis, and provide evidence for best treatment.

METHODS

Search Strategy

A systematic search was performed using 3 databases (PubMed, EMBASE, and Web of Science) to identify articles reporting LT. Original articles, case series, and individual reports from January 1950 to December 2014 were considered. Inclusion criteria were publications that (1) involved one or more cases, (2) presented clinical features in each patient, and (3) provided individual clinical outcome. Publications were excluded if the study (1) reported no individual clinical information, (2) lacked survival data, or (3) only described the prevention of LT.

A search strategy using the term “lung torsion OR pulmonary torsion” with the limitation of English language and human only identified 405 articles after removal of duplicates. After reviewing the titles, when appropriate, the abstract or actual articles, 109 articles were selected for further review. Ninety-one articles were used for analysis (Figure E1). The methodological quality of the studies was assessed (Table E1).

Data Extraction

The relevant variables (Table E2) from the selected studies were collected by 2 researchers independently. Extracted data included (1) publication year, (2) patient gender and age, (3) cause of LT, (4) clinical presentation, (5) diagnostic information comprising prospective diagnosis, diagnostic method, and time to diagnosis, (6) pathologic information comprising operative findings, extent of torsion, direction, and degree of torsion, (7) intervention, and (8) clinical outcome. Prospective diagnoses were classified as correct diagnosis, underdiagnosis (patients who were not diagnosed as LT before treatment), and misdiagnosis (patients who were misdiagnosed as other diseases). Time to diagnosis was defined as the interval between the date of operation or trauma according to the different causes and the date of LT diagnosis. Degree of torsion was categorized as partial or complete when the degree of rotation was $<180^\circ$ or $\geq 180^\circ$. Intervention included direct resection, indirect resection, and reposition. Direct resection meant resection of the affected lung directly, and in indirect resection the lesion was initially rotated back to its normal anatomic position and then quickly removed. The primary efficacy outcome was LT-related mortality, which was defined as death as a result of LT during the hospitalization or within 30 days of reoperation.

Data Synthesis

Because most of the published literature were case reports and individual information could be achieved from the original studies, a pooled analysis using a univariate logistic regression model was performed

to compare patient survival between the grouped variables within specific clinical data. The results were presented as odds ratio (OR) with 95% confidence intervals (CIs). For overall case series, baseline characteristics between extent of torsion were compared by χ^2 test for categorical variables and analysis of variance test or nonparametric test (if appropriate) for continuous variables. Statistical analysis was performed using IBM SPSS Statistics (version 19; Chicago, Ill) and graphs using Stata/SE (version 12; College Station, Tex).

RESULTS

Patient Characteristics

One hundred and nine patients in 91 studies were identified from the literature. The mean age was 46.0 ± 23.7 years. There were 60 men (58.3%) among the 103 patients for which gender was reported.

The clinical characteristics are summarized in Table 1. LT occurred in 68 patients (62.4%) postoperatively, in 9 (8.3%) after trauma, and in 32 (29.4%) spontaneously (including 6 after percutaneous lung biopsy). Because video-assisted thoracoscopic surgery (VATS) was first introduced in 1992, the surgical approach after 1992 was compared. LT occurred in 29 patients (78.4%) after thoracotomy and in 8 (21.6%) after VATS. Sixty-eight of 86 patients (79.1%) had obvious clinical presentations, including dyspnea (38.4%), fever (23.3%), chest pain (17.4%), and cough (14.0%); the other 18 (20.9%) were asymptomatic.

Diagnosis and Distribution of LT

Sixty-three patients (57.8%) were diagnosed with or suspected of having LT preoperatively and 20 patients (18.3%) were misdiagnosed as having other diseases, including hemothorax (6 cases), parenchymal infection/hemorrhage (5 cases), atelectasis (2 cases), and tumor (2 cases). In 63 patients who were correctly diagnosed, the diagnostic methods depended mainly on bronchoscopy, thoracic computed tomography (CT), and chest radiography with a median time to diagnosis of 4 days (Table 1). Bronchoscopy examination was performed in 27 patients and showed narrowed orifice in 9, complete airway obstruction in 10, and tortuous bronchus in 8 patients. Thoracic CT and chest radiography was performed in 25 and 11 patients, respectively. Radiologic findings suggestive of LT included consolidation (14 cases), abnormal anatomic structures (8 cases), and lesion shift (3 cases). The consolidation was usually progressive (increase in size). The bronchial or vascular structures were narrowing, blocked, or in an inverse position. Sometimes, the radiologic corresponding lesion changed position on sequential CT scans with the progress of torsion. Pulmonary angiography (6 cases) and CT angiography (6 cases) showed the tortile course of the artery branch, abrupt truncation, and tapering of the pulmonary artery. In patients who were underdiagnosed as LT (10 cases), the most common false-negative CT findings were persistent

TABLE 1. Clinical variables

A. Patient characteristics	No. of patients (%)
Age (y), mean $\pm$ SD (n = 108)	46.0 $\pm$ 23.7
Gender (n = 103)	
Male	60 (58.3)
Female	43 (41.7)
Cause (n = 109)	
Postoperative	68 (62.4)
After lung resection	39
After lung transplantation	10
After other thoracic surgery	19
Posttraumatic	9 (8.3)
Spontaneous	32 (29.4)
Clinical presentation (n = 86)	
Asymptomatic	18 (20.9)
Symptomatic	68 (79.1)
Dyspnea	33
Fever	20
Chest pain	15
Cough	12
Blood sputum	3
B. Diagnostic information	
Prospective diagnosis (n = 109)	
Correct diagnosis	63 (57.8)
Underdiagnosis	26 (23.9)
Misdiagnosis	20 (18.3)
Hemothorax/intrathoracic hematoma	6
Parenchymal infection/hemorrhage	5
Atelectasis	2
Tumor	2
Located effusion	1
Emphysema	1
Inadvertent ligation of hilum	1
Leakage at anastomosis site	1
Diaphragmatic herniation	1
Diagnostic method (n = 63)*	
Bronchoscopy	27
Thoracic CT	25
Chest roentgenography	11
Pulmonary angiogram	6
Pulmonary CT angiography	6
Bronchogram	2
Median time to diagnosis (d) (n = 63 patients)	4
C. Pathologic information	
Operative finding (n = 82)	
Congestion	33 (40.2)
Infarction	49 (59.8)
Degree of torsion (n = 70)	
90°	9
120°	4
150°	1
180°	46
270°	1
360°	7

(Continued)

TABLE 1. Continued

C. Pathologic information	
720°	1
1080°	1
Direction of torsion (n = 45)	
Clockwise	30 (66.7)
Counterclockwise	15 (33.3)

SD, Standard deviation; CT, computed tomography. *Data based on the 63 patients who were prospectively diagnosed as lung torsion.

opacification without tracheal deviation and masslike opacity. Likewise, these 2 CT features were easily misdiagnosed as infection and postoperative hemorrhage.

Individual lobar torsion occurred in 93 patients (Table 2). The right middle lobe (24 cases) was the most commonly affected individual lobe after thoracic surgery, and the right upper lobe (8 cases) was the most vulnerable lesion occurring spontaneously. With regard to lobar torsion after lung resection (Table 3), LT after upper lobe resection was more frequently reported; middle lobar torsion after right upper lobectomy (16 cases, 41.0%) and left lower lobar torsion after left upper lobectomy (9 cases, 23.1%) were the 2 main reported situations.

Sixteen patients had whole LT (Table 2); 7 after thoracic surgery, including lung transplantation (2 cases), transthoracic esophageal operation (3 cases), repair of aorta (1 case), and transthoracic correction of hiatal hernia (1 case), 5 secondary to trauma, and 4 spontaneously. There was no significant difference in clinical characteristics except for cause of torsion between individual lobar torsion and whole LT. Six patients with whole LT were treated with direct resection (pneumonectomy), 7 with reposition, and 1 with detorsion and right upper lobectomy. Also, no significant difference was found in the treatment modalities between the 2 groups ($P = .333$, Table E3).

Intervention and Survival Analysis

Treatment methods were reported in 103 patients. The median interval between the date of first operation and the date of torsion was 3 days in cases with postoperative LT. Direct resection of affected lung without detorsion was performed in 56 patients (54.4%) and indirect resection in 13 (12.6%). Reposition was the treatment in 34 patients (33.0%). The operative findings of the lesions in 33 patients (40.2%) showed congestion and in another 49 patients (59.8%) showed infarction. Most of the lesions were found to have a complete torsion (56 cases, 80.0%) in a clockwise direction (30 cases, 66.7%).

Characteristics of 103 cases with different intervention were compared (Table E3). No significant difference in either time to diagnosis or extent of torsion was found between patients receiving different treatments. The lesions with direct resection had a higher proportion of tissue

TABLE 2. Extent of lung torsion

	Individual lobe (n = 93)					Whole lung (n = 16)*		P value†
	LUL, n (%) (n = 19)	LLL, n (%) (n = 16)	RUL, n (%) (n = 16)	RML, n (%) (n = 32)	RLL, n (%) (n = 5)	Left, n (%) (n = 12)	Right, n (%) (n = 5)	
Age (y), mean ± SD	45.5 ± 24.9	52.7 ± 14.4	50.6 ± 23.3	51.6 ± 22.4	41.4 ± 26.4	35.6 ± 25.2	59.4 ± 24.8	.434
Gender								
Male	8 (42.1)	8 (61.5)	11 (68.7)	17 (56.7)	4 (100)	8 (66.7)	2 (40.0)	.766
Female	10 (52.6)	5 (38.5)	5 (31.3)	13 (43.3)	0	4 (33.3)	3 (60.0)	
Cause								
Postoperative	15 (78.9)	15 (93.8)	7 (43.7)	24 (75.0)	3 (60.0)	7 (58.4)	1 (20.0)	.002
Posttraumatic	0	0	1 (6.3)	2 (6.3)	1 (20.0)	4 (33.3)	1 (20.0)	
Spontaneous	4 (21.1)	1 (6.3)	8 (50.0)	6 (18.8)	1 (20.0)	1 (8.3)	3 (60.0)	
Clinical presentation								
Symptomatic	9 (69.2)	10 (76.9)	9 (69.2)	23 (92.0)	3 (75.0)	6 (75.0)	3 (60.0)	.303
Asymptomatic	4 (30.8)	3 (23.1)	4 (30.8)	2 (8.0)	1 (25.0)	2 (25.0)	2 (40.0)	
Time to diagnosis, d ± SD	7.7 ± 9.9	3.1 ± 3.3	5.1 ± 4.3	5.6 ± 4.8	7.7 ± 6.0	15 ± 20	7.5 ± 9.3	.312
Prospective diagnosis								
Correct diagnosis	10 (52.6)	9 (56.3)	9 (56.3)	25 (78.1)	5 (100)	4 (36.4)	3 (60.0)	.343
Underdiagnosis	3 (15.8)	4 (25.0)	4 (25.0)	6 (18.8)	0	3 (27.3)	2 (40.0)	
Misdiagnosis	6 (31.6)	3 (18.8)	3 (18.7)	1 (3.1)	0	4 (36.4)	0	
Operative finding								
Congestion	4 (40.0)	4 (36.4)	7 (70.0)	10 (34.5)	4 (100)	3 (25.0)	4 (100)	.840
Infarction	6 (60.0)	7 (63.6)	3 (30.0)	19 (65.5)	0	9 (75.0)	0	
Direction of torsion								
Clockwise	4 (66.7)	5 (55.6)	5 (71.4)	8 (61.5)	1 (50.0)	8 (100)	1 (33.3)	.301
Counterclockwise	2 (33.3)	4 (44.4)	2 (28.6)	5 (38.5)	1 (50.0)	0	2 (66.7)	
Degree of torsion								
Partial	1 (7.7)	3 (27.3)	1 (14.3)	4 (17.4)	2 (50.0)	4 (33.3)	2 (40.0)	.200
Complete	12 (92.3)	8 (72.7)	6 (85.7)	19 (82.6)	2 (50.0)	8 (66.7)	3 (60.0)	

Bold values indicate *P* values with statistically significant difference. LUL, Left upper lobe; LLL, left lower lobe; RUL, right upper lobe; RML, right middle lobe; RLL, right lower lobe; SD, standard deviation. *One patient had bilateral whole lung torsion. †Comparison between individual lobar torsion and whole lung torsion.

infarction compared with those receiving reposition with more pathologic tissue congestion ($P < .001$).

Postoperative complications occurred in 3 patients, including 1 case of aspiration pneumonia and 2 of acute respiratory distress syndrome (ARDS). All 3 patients

recovered after conservative treatment or the application of extracorporeal membrane oxygenation. The LT-related mortality has decreased over time based on the available studies (Figure 1). For this entire cohort, the rate was 8.3%. Nine patients died during the postoperative course,

TABLE 3. Lobar torsion after lung resection

	Total, n (%) (n = 39)	LUL, n	LLL, n	RUL, n	RML, n
Upper lobe resection (n = 29, 74.4%)					
Right upper lobectomy	16 (41.0)				16
Right anterior segmentectomy	1 (2.6)			1	1
Left upper lobectomy	9 (23.1)		9		
Left upper division segmentectomy	1 (2.6)	1			
Left lingual segmentectomy	1 (2.6)	1			
Wedge resection (left upper lobe)	1 (2.6)	1			
Middle lobe resection (n = 3, 7.7%)					
Right middle lobectomy	3 (7.7)			3	
Lower lobe resection (n = 3, 7.7%)					
Left lower lobectomy	3 (7.7)	3			
Mixed lobes resection (n = 4, 10.2%)					
Right upper lobectomy + right superior segmentectomy	1 (2.6)				1
Right upper lobectomy + right lower lobectomy	1 (2.6)				1
Left upper lobectomy + left superior segmentectomy	2 (5.1)		2		

LUL, Left upper lobe; LLL, left lower lobe; RUL, right upper lobe; RML, right middle lobe.

FIGURE 1. Time trend in lung torsion.

including 3 of cardiac arrest,^{6,9,10} 1 from ARDS,⁶ 2 of cerebral embolism,^{7,8} 2 of sepsis,^{5,11} and 1 of an unaccounted reason.¹² Univariate analysis was available for 109 patients (Table 4); results showed that whole LT was associated with a significantly worse survival than lobar torsion (OR, 5.867; 95% CI, 1.381–24.923). No significant difference in survival was found between patients receiving direct resection and those receiving reposition, and a trend was observed for worsening outcome in patients treated with indirect resection compared with patients treated with direct resection, although the difference fell short of statistical significance (OR, 5.300; $P = .060$). However, when a multivariate analysis was performed analyzing variables with more than 80% data provided for the total number of patients included, both whole LT and indirect resection were significantly associated with worse outcome (data not shown). The rest of the clinical variables were not significantly correlated to survival (Figure E2).

DISCUSSION

This is the first review of the published literature, to the best of our knowledge, to identify the disease distribution and prognostic factors in patients with LT. Given the low incidence of LT, the paucity of large-scale studies makes it difficult to generate sound evidence-based recommendations. However, because surgeons may encounter the disease, guidance should be based on the best available evidence.

Systematic reviews are used to support the development of clinical practice guidelines and inform clinical decision making.¹³ Our analysis was conducted from 91 case reports published between 1950 and 2014, representing 109 patients. The results of this review suggest that (1) the cause of LT does not seem to be associated with survival, (2) whole LT tends toward higher mortality than lobar torsion, (3) survival of patients receiving reposition is similar to that of patients receiving direct resection if the tortuous

part is viable; however, indirect resection might increase risk of death. Further studies are still warranted to validate our findings.

At the early stages after LT, symptoms can present dramatically, including dyspnea, fever, and chest pain, but these symptoms are insufficient for diagnosis.⁵ It is important, then, that the suspicion of LT should be raised on radiologic findings. Moser and colleagues¹⁴ have reported that complete opacification of the affected lung was the most common presentation, followed by inversion of the vascular pattern, which is concordant with our results. Bronchoscopy was an alternative examination, and it was usually performed based on the radiologic manifestation.¹⁵ Apostolakis and colleagues⁷ and Hennink and colleagues² stated that the most suggestive bronchoscopic findings included bronchial occlusion and fish mouth orifice. Therefore, according to our results, a potential diagnostic algorithm of LT (Figure 2) is that patients presenting with dyspnea, fever, or chest pain with contributing factors for LT should first undergo chest radiography. If progressive consolidation is identified on the radiograph, thoracic CT or bronchoscopy is the next method of investigation. Both CT and bronchoscopy are the modalities of choice for prospective diagnosis. If CT shows bronchial or vascular malposition or abrupt truncation, or bronchoscopy shows narrow or distorted bronchus orifice, LT ought to be highly suspected. Additional examinations such as pulmonary angiography and CT angiography are reserved for patients with atypical manifestation. Preoperative evaluation for pulmonary venous thrombus is complicated but still necessary, although it has not been reported in previous studies.

The differential diagnosis of LT includes pneumonia, contusion, and particularly, lung gangrene complicating lung resection. Postoperative pneumonia is often caused by aspiration and accompanied by a series of

TABLE 4. Risk factors on survival

	Number of patients	Mortality (%)	Univariate logistic regression model	
			Odds ratio (95% CI)	P value
Age (y)*	108		1.010 (0.979–1.042)	.530
Gender				
Male	60	8.3	Reference	
Female	43	9.3	1.128 (0.285–4.472)	.864
Cause				
Postoperative	68	8.8	Reference	
Posttraumatic	9	22.2	2.952 (0.497–17.523)	.233
Spontaneous	32	3.1	0.333 (0.038–2.892)	.319
Clinical presentation				
Asymptomatic	18	11.1	Reference	
Symptomatic	68	7.4	0.635 (0.113–3.578)	.607
Extent of torsion				
Single lobe	93	5.4	Reference	
Whole lung	16	25.0	5.867 (1.381–24.923)	.017
Prospective diagnosis				
Correct diagnosis	63	7.9	Reference	
Underdiagnosis	26	7.7	0.967 (0.175–5.331)	.969
Misdiagnosis	20	10.0	1.289 (0.230–7.219)	.773
Time to diagnosis (d)*	82		0.731 (0.492–1.087)	.122
Operative finding				
Congestion	33	3.0	Reference	
Infarction	49	14.3	5.333 (0.624–45.565)	.126
Direction of torsion				
Clockwise	30	10.0	Reference	
Counterclockwise	15	6.7	0.643 (0.061–6.764)	.713
Degree of torsion				
Partial	14	14.3	Reference	
Complete	56	10.7	0.720 (0.129–4.020)	.708
Treatment†				
Direct resection	56	5.4	Reference	
Indirect resection	13	23.1	5.300 (0.933–30.109)	.060
Reposition	34	5.9	1.104 (0.175–6.968)	.916

Bold values indicate *P* values with statistically significant difference. *CI*, Confidence interval. *Age and time window to diagnosis as continuous variables. †Indirect resection represented that the lesion was initially rotated back to its normal anatomic position and then quickly removed.

concomitant presentations (phlegm, fever, increased white blood cell count). Contusion is more frequent after sublobar resection than after lobectomy and usually manifests as airspace consolidation that can disappear spontaneously in a few days.¹⁶ Lung gangrene, resulting from accidental/undiscovered injuries of segmental arteries/veins to the remaining lobe, may mimic LT because both diseases are characterized by arterial or venous obstruction. One common radiologic finding of LT is consolidation (as shown in our review), and lung gangrene may manifest as a radiologic opacification.¹⁶ Bronchoscopy and CT are essential for discriminating the 2 diseases. Bronchoscopy can show airway obstruction and CT can show an inverse vascular pattern in LT.

FIGURE 2. Diagnostic algorithm of lung torsion.

The cause of LT was described in all the studies. It was reported that postoperative occurrence was the most common situation.⁶ With an increasing use of VATS in general thoracic diseases, there is a concern about whether VATS will increase the risk of postoperative LT because of the minimal invasiveness and subsequent less adhesion formation. Our study identified 8 patients with LT after VATS since the first report by Eguchi and colleagues.¹⁷ However, it is inadequate for addressing this issue because this analysis was based on retrospective case reports lacking negative controls, and therefore, further comparative studies are needed. Posttraumatic LTs are even more rare. To our knowledge, there are only 9 case reports describing LTs after blunt chest trauma. The pathogenesis remains elusive. It has been postulated that a traumatic tear of the inferior pulmonary ligament and a resultant hemo-/pneumothorax may allow for increased mobility of the lung.¹⁸ Spontaneous LT may occur in pulmonary conditions such as pneumothorax, pleural effusion, lobar atelectasis, and pulmonary sequestration.^{19,20}

A retrospective study by Cable and colleagues¹ including 7 postoperative lobar torsion patients reported no deaths, which compared favorably with other reports of 12% to 16% mortality in previous studies.^{21,22} Nevertheless, there was a lack of survival data in posttraumatic and spontaneous occurrences. Our study showed the mortality of LT after thoracic surgery, secondary to trauma, and spontaneously was 8.8%, 22.2%, and 3.1%, respectively. Univariate analysis showed no significant survival difference based on the specific causes. The reasons behind this insignificance may be related to the low

incidence of LT. It is plausible that blunt trauma may give rise to more tissue damage and likelihood of comorbidities, resulting in higher mortality. Our finding also showed a trend for worsening survival rates in patients with posttraumatic LT. Therefore, regardless of the cause, timely detection of LT in any clinical scenarios is of paramount importance.

Whole LT was rarely reported. Stratemeier and colleagues¹² first described a whole left LT in 1954. The reported incidence of lobar torsion is 0.2% to 0.3% after thoracotomies⁶; however, there are no reliable data on the incidence of whole LT. Prospective diagnosis of whole LT was challenging because it was easily misdiagnosed as hemothorax²³ or postsurgical pulmonary contusion.²⁴ Furthermore, it was well associated with complete rotation in the present study, producing obstruction of the main bronchus and corresponding arterial and venous circulation and failure in the collateral system,²⁵ which posed a pressing problem for both diagnosis and treatment. So it is reasonable to infer that mortality should be higher in patients with whole LTs than in patients with lobar torsions, as shown in our study.

The acceptable intervention for LT is reposition or resection of the tortuous part. Still, controversy exists over the optimal treatment. Some investigators have argued against reposition, fearing secondary embolism caused by emboli released from the obstructed pulmonary vein^{7,8} and acute inflammatory response caused by inflammatory mediators released from the tortuous segment,²⁶ resulting in ARDS and possibly multiorgan dysfunction.²⁷ However, a series of successful cases with salvaged lobes after reposition refuted this argument.^{19,20,28} Oliveira and colleagues²⁸ advocated that reposition could be performed if arterial flow was shown preoperatively. Trotter and colleagues²⁰ stated that the decision to choose between reposition and resection depended on the viability of the affected pulmonary tissue. Kanaan and colleagues²⁹ believed the time to reoperation was another determinant. Takada and Kuriyama³⁰ and Banki and Velmahos⁵ proposed that repositioning of the affected lung or lobe should be considered within the first day of onset. However, in our analysis, the choice of intervention was determined by the operative finding because infarcted lesions were more often treated with resection. Based on this premise, there was no significant difference in mortality between the 2 interventions. Intraoperatively, infarcted lesions generally appeared as a markedly swollen, airless, blue-black hemorrhagic surface with a complete torsion,^{2,21,24} which might be a clue to assist surgeons not only in judging the viability of the tortuous lung but also, going one step further, in deciding the treatment method. Therefore, we can safely conclude that it is justifiable to preserve the affected lung by performing reposition if the involved

lung is viable, but the temptation to reposition the lung should be resisted if the lesion is as described earlier, indicating hemorrhagic infarction.

The procedures for resection included resection without detorsion (direct resection) and resection with detorsion (indirect resection). Given that the anatomic landmarks were usually obscured in LT, indirect resection could facilitate the surgical operation by detorsing the lung. Nevertheless, it may result in severe complications such as ARDS, secondary to the absorption of necrotic tissue, and an extensive systemic circulation embolism.⁶⁻⁸ Banki and Velmahos⁵ insisted on direct resection to avoid a reperfusion insult. In our analysis, patients with direct resection had relatively better survival, albeit not significant, compared with those with indirect resection. Direct resection would thus be the treatment of choice if resection had to be considered.

This review also found that cases with lobar torsion were frequently reported after upper lobectomies. This was consistent with the results of Wagner and Nesbitt,²² who pointed out that lobar torsion after upper lobe resections accounted for 85% of the cases in the literature. So it seems reasonable that preventive measures could be taken after upper lobe resections. Likewise, certain methods ought to be implemented to avoid repeated torsion after reposition. It is a consensus that correct positioning of the remaining lobes before chest closure and a reduction in their mobility are 2 major prophylactic methods. In previous studies, several techniques for fixation of a floppy lobe have been recommended in the setting of long hilar pedicle and large complete fissure, including stitching the lung to the adjacent thoracic structures (eg, pericardial fat pad or pleura), suturing the middle and lower lobes together, or using medical adhesives.³¹⁻³³ Identification of the possible direction of torsion of a floppy lobe is crucial before stitching. In addition, adhesives can be used only when the lung is inflated and any potential torsion is checked.³¹ During VATS, preservation of the pulmonary ligament can also potentially prevent LT.³⁴ In the context of reoperation for reposition, these techniques may also be applicable. Sakai and colleagues³⁵ attached the middle lobe to the lower lobe using over-and-over sutures after detorsion. However, every maneuver has both pros and cons. Interlobular suturing is cost effective but may cause postoperative air leakage and hemorrhage.³² Pleural flap creation is time consuming and not easy to reproduce. Use of medical adhesive is a simple and quick technique but it is costly, with uncertain benefit.³³ Because these methods are commonly introduced by case reports and almost no patient in our review received prophylactic techniques after lung resections, we were unable to evaluate effectiveness and to determine the superiority of one technique over another. Nonetheless, continued attention

should be paid to identifying LT in the early postoperative course.

Study Limitations

The main limitation in our study was associated with the potential publication bias (Figure E3). There must be a strong bias to present an original case, such as a different underlying cause, an unusual presenting feature, or a thought-provoking outcome, rather than an ordinary clinical setting. Although the decreasing mortality was restricted to the published studies, the low mortality with reposition treatment suggested that it can also apply to some candidates with viable torsion parts. Further studies are needed to address this issue.

Another limitation was related to the use of limited data for analysis. Some of the variables such as operative finding and time to diagnosis that might highly affect mortality fell just short of a statistically significant difference because of the incompleteness of the data provided in the articles reviewed. In addition, some of the series included patients from as far back as 1953; variability in treatment standards and diagnosis capabilities could be a factor. Our intuitive experience is that when patients have chest pain, hemoptysis, and dyspnea refractory to oxygen therapy in the early postoperative course, emergency chest radiography should be performed. If the radiologic sign of consolidation (sometimes opacity) cannot be explained by sputum impaction and lobar pneumonia, postoperative LT must be taken into consideration. Subsequent thoracic CT or bronchoscopy is the key test for prospective diagnosis and confirmation. The surgical intervention strategy ought to be based on the viability of the tortuous lobe, and if resection is necessary, we perform a direct resection to prevent inflammatory mediators from flowing into the systemic circulation. Despite some statistical weaknesses, certain conclusions can be drawn from this reasonable attempt.

CONCLUSIONS

LT is a rare event but can be associated with high mortality, and therefore a high index of suspicion and early surgical intervention are warranted. The key diagnostic tests include chest radiography, CT, and bronchoscopy. Whole LT tends toward higher mortality than does lobar torsion. If the tortuous part is viable, reposition and direct resection have similar survival rates. If not, direct resection should be performed, but indirect resection would probably increase the risk of death.

Conflict of Interest Statement

Authors have nothing to disclose with regard to commercial support.

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Key Words: lung torsion, systematic review, diagnosis, intervention, survival

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FIGURE E1. Search algorithm.

FIGURE E2. Univariate analysis for risk factors on lung torsion-related mortality. CI, Confidence interval.

FIGURE E3. Funnel plot of pooled analysis. Case reports were combined in to 5 study groups according to the year of publication (1953–1974, 1975–1984, 1985–1994, 1995–2004, and 2005–2014); however, 1 study group (study period from 1995 to 2004) was not shown in the funnel plot because the mortality was zero, failing to make a comparison between the extent of torsion. *OR*, Odds ratio.

TABLE E1. Assessment of the methodological quality of the 91 studies according to the checklists of the STROBE Statement

	Number			
	Yes	No	Unclear	Not applicable
1. (a) Indicate the study’s design with a commonly used term in the title or the abstract	75	16	0	0
(b) Provide in the abstract an informative and balanced summary of what was done and what was found	65	26	0	0
2. Explain the scientific background and rationale for the investigation being reported	0	0	0	91
3. State specific objectives, including any prespecified hypotheses	0	0	0	91
4. Present key elements of study design early in the paper	0	0	0	91
5. Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	91	0	0	0
6. (a) Give the eligibility criteria, and the sources and methods of selection of participants	0	0	0	91
(b) For matched studies, give matching criteria and the number of controls per case	0	0	0	91
7. Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	91	0	0	0
8. For each variable of interest give sources of data and details of methods of assessment (measurement)	43	48	0	0
9. Describe any efforts to address potential sources of bias	0	0	0	91
10. Explain how the study size was arrived at	0	0	0	91
11. Explain how quantitative variables were handled in the analyses	0	0	0	91
12. (a) Describe all statistical methods, including those used to control for confounding	0	0	0	91
(b) Describe any methods used to examine subgroups and interactions	0	0	0	91
(c) Explain how missing data were addressed	0	91	0	0
(d) if applicable, describe analytic methods taking account of sampling strategy	0	0	0	91
(e) Describe any sensitivity analyses	0	0	0	91
13. (a) Report the numbers of individuals at each stage of the study	0	0	0	91
(b) Give reasons for nonparticipation at each stage	0	0	0	91
(c) Consider use of a flow diagram	0	0	0	91
14. (a) Give characteristics of study participants (eg, demographic, clinical, social) and information on exposures and potential confounders	91	0	0	0
(b) Indicate the number of participants with missing data for each variable of interest	0	91	0	0
15. Report numbers of outcome events or summary measures	91	0	0	0
16. (a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision	0	0	0	91
(b) Report category boundaries when continuous variables were categorized	0	0	0	91
(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	0	0	0	91

TABLE E2. Availability of data for the total number of patients (n = 109)

Clinical variable	% for whom data available (n)
Year of publication	100 (109)
Cause	100 (109)
Prospective diagnosis	100 (109)
Extent of torsion	100 (109)
Clinical outcome	100 (109)
Age	99 (108)
Gender	94 (103)
Treatment	94 (103)
Clinical presentation	79 (86)
Time to diagnosis	75 (82)
Operative finding	75 (82)
Degree of torsion	64 (70)
Direction of torsion	41 (45)

TABLE E3. Characteristics of 103 cases with different treatments

	Treatment*			P value
	Direct resection, n (%) (n = 56, 54.4%)	Indirect resection, n (%) (n = 13, 12.6%)	Reposition, n (%) (n = 34, 33.0%)	
Age (y), mean $\pm$ SD†	42.3 $\pm$ 22.4	47.3 $\pm$ 25.4	50.7 $\pm$ 24.1	.146
Gender				
Male	31 (56.4)	4 (40.0)	19 (59.4)	.553
Female	24 (43.6)	6 (60.0)	13 (40.6)	
Cause				
Postoperative	33 (58.9)	11 (84.6)	22 (64.7)	.441
Posttraumatic	6 (10.7)	0	2 (5.9)	
Spontaneous	17 (30.4)	2 (15.4)	10 (29.4)	
Clinical presentation				
Asymptomatic	6 (12.8)	2 (25.0)	9 (31.0)	.147
Symptomatic	41 (87.2)	6 (75.0)	20 (69.0)	
Prospective diagnosis				
Correct diagnosis	28 (50.0)	7 (53.8)	23 (67.6)	.427
Underdiagnosis	15 (26.8)	3 (23.1)	8 (23.5)	
Misdiagnosis	13 (23.2)	3 (23.1)	3 (8.8)	
Time to diagnosis, d $\pm$ SD†	7.6 $\pm$ 9.1	5.0 $\pm$ 6.5	4.0 $\pm$ 4.5	.103
Operative finding				
Congestion	6 (13.6)	3 (30.0)	22 (88.0)	<.001
Infarction	38 (86.4)	7 (70.0)	3 (12.0)	
Extent of torsion				
Single lobe	50 (89.3)	12 (92.3)	27 (79.4)	.333
Whole lung	6 (10.7)	1 (7.7)	7 (20.6)	
Direction of torsion				
Clockwise	12 (63.2)	4 (66.7)	13 (76.5)	.683
Counterclockwise	7 (36.8)	2 (33.3)	4 (23.5)	
Degree of torsion				
Partial	4 (12.1)	1 (10.0)	7 (29.2)	.197
Complete	29 (87.9)	9 (90.0)	17 (70.8)	

Bold values indicate *P* values with statistically significant difference. *SD*, Standard deviation. *For indirect resection, the lesion was initially rotated back to its normal anatomic position and then quickly removed. †Age and time window to diagnosis as continuous variables.