Spontaneous middle lobe torsion: An institutional case series

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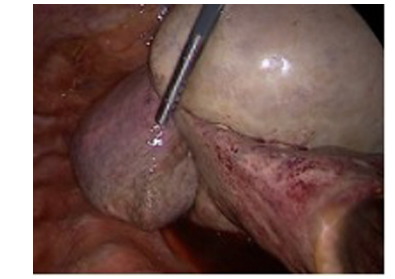
ABSTRACT

Objective: Lobar torsion is a rare occurrence in which a portion of the lung is twisted on its bronchovascular pedicle. The vast majority are observed in the acute postoperative period often following right upper lobectomy. Spontaneous middle lobe torsion independent of pulmonary resection is exceptionally rarer; fewer than 15 cases have been recorded. We present an institutional case series of 2 patients postorthotopic liver transplantation who developed spontaneous middle lobe torsion due to large pleural effusions.

Methods: We provide the medical course as well as intraoperative techniques for our 2 patients along with a review of the literature.

Results: Both patients in this case series underwent orthotopic liver transplant complicated postoperatively by a large pulmonary effusion. Patient one developed an abdominal hematoma requiring evacuation and repair, after which he developed progressive shortness of breath. Bronchoscopy revealed a right middle lobe obstruction; upon thoracotomy, 180-degree torsion with widespread necrosis was evident and the middle lobe was removed. He is doing well to date. Patient 2 experienced postoperative pleural effusion and mucus plugging; computed tomography revealed abrupt middle lobe arterial occlusion prompting urgent operative intervention. Again, the middle lobe was grossly ischemic and dissection revealed a 360-degree torsion around the pedicle. It was resected. He is doing well to date.

Conclusions: As the result of its rarity, radiographic and clinical diagnosis of spontaneous pulmonary lobar torsion is challenging; a high index of suspicion for spontaneous middle lobe torsion must be maintained to avoid delays in diagnosis. Prompt surgical intervention is essential to improve patient outcomes. (JTCVS Techniques 2023;20:176-81)



Initial thoracoscopic appearance of the middle lobe consistent with hemorrhagic infarction.

CENTRAL MESSAGE

Due to its rarity, radiographic and clinical diagnosis of spontaneous pulmonary lobar torsion is challenging. Prompt surgical intervention is essential to improve patient outcomes.

PERSPECTIVE

Spontaneous middle lobe torsion is a rare phenomenon that is difficult to recognize both clinically and radiographically. As a result, the majority of cases require resection and are associated with high morbidity and mortality. A high index of suspicion must be maintained to avoid delays in diagnosis and treatment. Prompt surgical intervention is crucial to improving patient outcomes.

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Abbreviations and Acronyms
CT = computed tomography
ML = middle lobe
OLT = orthotopic liver transplant
POD = postoperative day
RLL = right lower lobe

▶ Video clip is available online.

Middle-lobe (ML) torsion most commonly occurs as a complication of lung resection, often after right upper lobectomy.[1] Nonpulmonary thoracic surgery, large pneumothorax, and thoracic trauma have also been shown to cause a small percentage of cases.[2-4] True spontaneous right lung ML torsion is very rare, comprising less than 5% of all cases of pulmonary torsion. Although definitive nomenclature has not been established, the term "spontaneous" in this setting traditionally refers to lobar torsion that occurs in the absence of previous thoracic surgery or trauma.[2,3] We present an institutional case series of 2 patients with spontaneous ML torsion due to pleural effusions who had recently undergone orthotopic liver transplant (OLT) (Figure 1). To our knowledge, these are the first reported cases of spontaneous ML torsion after OLT. Both patients in the study provided informed written consent for the publication of their study data; therefore, per institution, no institutional review board/ethics review board approval was required.

CASES

Case 1

Patient 1 is a 63-year-old man with a medical history of end-stage liver disease secondary to alcoholic cirrhosis who presented initially for OLT. His surgery and early postoperative course were complicated by hemorrhagic shock, coagulopathy, and respiratory failure. The patient continued to decompensate on postoperative day (POD) 5, and subsequent workup revealed a large abdominal fluid collection suspicious for biliary leak. He was taken back to the operating room on POD 9 for evacuation of infected abdominal hematoma and repair of a biliary anastomotic leak. This operation was again complicated by hemorrhagic and vasodilatory shock.

Over the course of the following 5 days, he developed progressive shortness of breath accompanied by up-trending leukocytosis. Sequential radiographs of the chest showed worsening right pleural effusion and basilar opacities. A computed tomography (CT) scan confirmed an increasing right pleural effusion and resultant complete collapse of the right lower lobe (RLL), with an abrupt termination of the ML bronchus, which was swirled near the inferior aspect of the hilum. The ML pulmonary vein was abruptly occluded, and no pulmonary artery branch to the ML was visualized. In addition, the ML remained posteriorly displaced with extensive ground-glass opacities and consolidation suggestive of ML torsion and/or infarction (Figure 2). The patient was then taken to the operating room for intervention.

Immediately before the operation, an on-table flexible bronchoscopy was performed. An unusual angulation to the RLL was noted, and the bronchoscope could only be partially advanced into the ML bronchus before a complete obstruction was encountered (Figure 3). We then proceeded with diagnostic thoracoscopy, and a double-lumen tube was positioned. Upon entering the chest, a large pleural effusion was encountered. The ML appeared grossly ischemic and would not decompress, consistent with torsion (Figure 4, *A*). We then converted to a posterolateral thoracotomy, and it was quite clear that the ML was torsed 180° (Figure 4, *B*). ML dissection was carried out, showing a long bronchovascular pedicle with complete fissures. On assessment of the vasculature, the ML vein came off independently from the atrium, which was dissected and freed. The ML pulmonary arteries and bronchus were dissected out and divided, and the ML specimen was removed. Back-table evaluation showed clear evidence of hemorrhagic necrosis throughout (Figure 5). Following resection of the ML, the RLL re-expanded and was in normal anatomic configuration, as was the right upper lobe. The chest was irrigated with antibiotic solution, chest tubes were placed, and the incision was closed in a normal fashion. After replacement of the double-lumen tube with a single-lumen endotracheal tube, bronchoscopy was performed and the patient was transferred back to the intensive care unit intubated but in stable condition. These intraoperative findings are depicted in the associated video (Video 1).

Final surgical pathology of the ML showed extensive hemorrhagic necrosis without evidence of malignancy, compatible with torsion. Over the next several days, the patient developed refractory hypotension requiring pressor support, coagulopathy, and worsening liver and kidney failure. A liver biopsy showed marked bile ductular reaction with periportal fibrosis and prominent hepatocellular and canalicular cholestasis, consistent with biliary outflow impairment. The patient was relisted and underwent repeat liver with kidney transplant. His postoperative course was prolonged as anticipated, with requirement for temporary tracheostomy. The patient, nevertheless, was able to be successfully discharged to a rehabilitation facility on POD 88 without tracheostomy. He is currently doing well.

Case 2

Patient 2 is a 57-year-old man who presented for OLT due to end-stage liver disease secondary to hepatitis C and

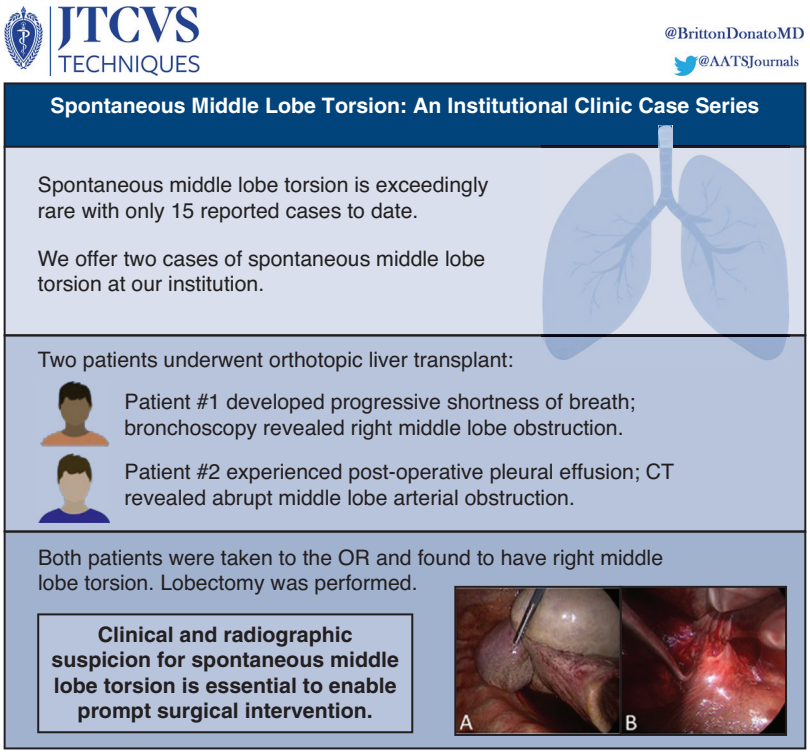


FIGURE 1. Shown are the methods, results, and implications of this study. *CT*, Computed tomography; *OR*, operating room.

alcoholic cirrhosis. The patient's pretransplant course was complicated by large hepatic hydrothorax requiring multiple thoracenteses. Postoperatively, the patient suffered from intermittent respiratory failure and hypotension requiring reintubation. Bronchoscopy 4 days after transplant showed complete collapse of the ML, and bronchoalveolar lavage was positive for *Stenotrophomonas*. CT of the chest demonstrated a right-sided pleural effusion and opacification of the ML, which was interpreted as mucous plugging. Nine days later, the patient underwent PleurX catheter (BD) placement, with resolution of the effusion noted. Repeat CT demonstrated complete consolidation of the ML with abrupt ML arterial occlusion at the level of the hilum, prompting urgent operative intervention.

At the time of surgery, a limited thoracotomy was performed, and amber-colored pleural fluid was evacuated. The ML appeared initially in proper anatomic position in reference to the upper and lower lobes. The ML was grossly ischemic, with inflammatory adhesions present between the ML and RLL. However, further dissection of the pulmonary hilum and ML bronchovascular pedicle revealed a complete 360° torsion. The ML was detorsed and brought back into

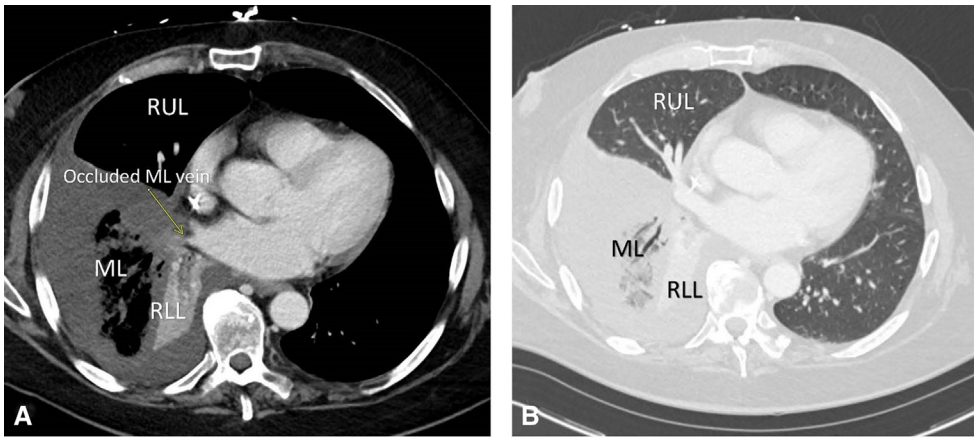


FIGURE 2. Computed tomography images demonstrating (A) an abruptly occluded middle lobe vein (*arrow*) and (B) ML bronchus with dense consolidation of the ML with hemorrhagic infarction consistent with ML torsion. *RUL*, Right upper lobe; *ML*, middle lobe; *RLL*, right lower lobe.

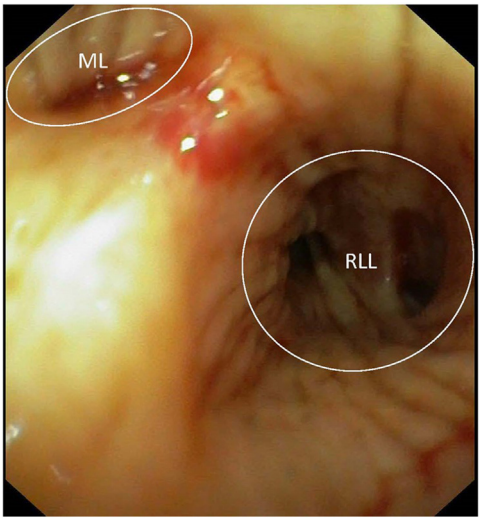


FIGURE 3. On-table bronchoscopy showing an angulated ML bronchus with subsequent occlusion consistent with ML torsion. *ML*, Middle lobe; *RLL*, right lower lobe.

normal anatomic position but did not appear viable and a formal anatomic resection of the ML was performed. The initial postoperative course was uneventful; however, on POD 10, he became hypoxemic and hypotensive, requiring pressor support, reintubation, and subsequent temporary tracheostomy. He eventually improved and was discharged on POD 43.

DISCUSSION

Torsion of the ML is a very rare occurrence. It is most often observed after pulmonary resection or trauma, with an estimated incidence of 0.0086% to 0.3%.[2,3] Even more rare is torsion independent of thoracic surgery or trauma, with fewer than 15 cases reported to date. Of these, only 2 have been in the setting of large pleural effusions. In our case series, both patients were found to have spontaneous ML torsion due to large pleural effusions after OLT. These are the first reported cases in this setting.

There are several anatomic features associated with lobar torsion. These include long bronchovascular pedicles, complete oblique fissure, narrow ML hilum, and lobar atelectasis.[2,5] Both patients had long bronchovascular pedicles with complete fissures. These anatomic variations, especially in the presence of a large pleural effusion, increase the relative mobility of the lobe, increasing the risk of torsion. The mobility of an atelectatic ML "floating" in a large-volume hydrothorax likely provides the opportunity for torsion along the bronchovascular axis. Pathophysiologically, torsion compromises vascular outflow, resulting in venous congestion and perivascular inflammation, making spontaneous detorsion increasingly unlikely.[3] As this process progresses, both perfusion and aeration of the affected lobe become compromised, ultimately leading to infarction and necrosis.[5] Greater degrees of torsion around the pedicle correlate with increasing severity of vascular congestion and ischemia. Therefore, complete 360° torsion typically results in more severe vascular and bronchial compromise and present more acutely. Conversely, lesser degrees of torsion can have a more insidious presentation, leading to delays in diagnosis and poor patient outcomes.[6-9]

Clinical diagnosis of lobar torsion can be challenging because of the subtlety of early radiographic findings and the relative rarity of this occurrence. The most common radiographic findings are hilar and vasculature displacement, air trapping, opacification of the affected lung, and a collapsed lobe.[5,10] It has been discussed in the literature that the absence of hypoxia with opacification of the affected lung may further support the diagnosis of torsion. If torsion is present, the affected lobe should be completely shunted, as both the airway and perfusing vasculature are typically compromised. This is in contrast to opacification secondary to mucous plugging, where the vasculature

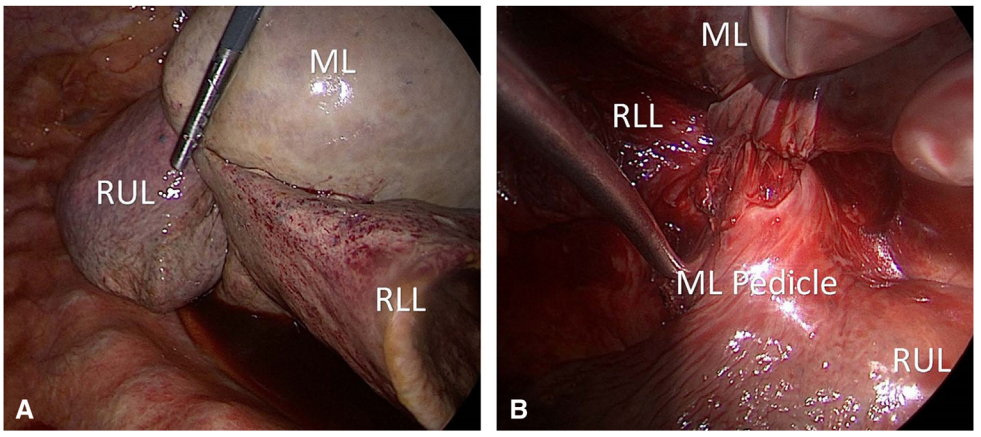


FIGURE 4. Intraoperative images demonstrating ML torsion. A, Initial appearance of the middle lobe consistent with hemorrhagic infarction and associated collapse of the right lower lobe. B, ML bronchovascular pedicle, which is noted to be torsed 180°. *ML*, Middle lobe; *RUL*, right upper lobe; *RLL*, right lower lobe.

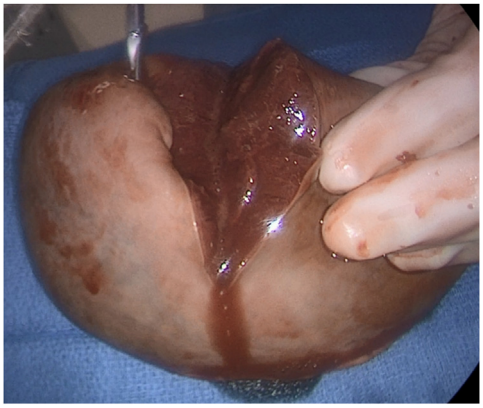
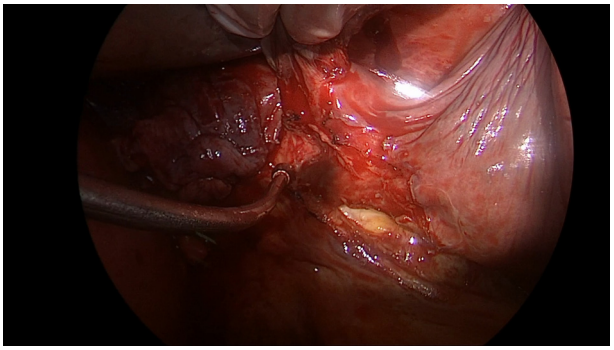


FIGURE 5. Back-table evaluation of the middle-lobe specimen with gross evidence of ischemic infarction and necrosis.

remains patent creating a ventilation–perfusion mismatch promoting hypoxia.[7] Delays in diagnosis secondary to difficulties in radiographic interpretation were observed in both cases in this series.

In retrospect, patient 1 had radiographic evidence of torsion 15 days before his lobectomy. A large right pleural effusion was also noted but had been longstanding and was thought to be a reactive hepatic hydrothorax. Patient 2 had post-OLT imaging that showed a right-sided pleural effusion and opacification with collapse of the ML, which was interpreted as mucous plugging. Despite the lack of usual risk factors, both patients were ultimately diagnosed with ML torsion and hemorrhagic infarction requiring surgical exploration and lobectomy.

Urgent operative intervention with lobectomy is recommended for lobar torsion when ischemic necrosis and gangrene are present. However, when there is partial or $\leq 180^{\circ}$ torsion without evidence of infarction, lobe preservation with detorsion remains controversial.[9] The risks of detorsion without resection include re-expansion pulmonary edema, thromboembolism (most notably pulmonary vein thrombosis), vascular complications, and eventual ischemic necrosis of the preserved lung with resultant sepsis and multiorgan system failure. Sakai and colleagues[9] reported a case of video-assisted thoracoscopic repair of ML torsion, but this is the only successful case to date. If preservation of a torsed lobe is to be considered, a comprehensive assessment of several factors must be undertaken: the degree of torsion around the pedicle, evidence of necrosis or gangrene both visually and biochemically, pulsation of the pulmonary vein, presence and severity of re-expansion pulmonary edema, and the expected postoperative pulmonary function. Additionally, given the significant risk of thromboembolic complications after detorsion, it is recommended that patients be anticoagulated postoperatively.[9] Despite a report of successful operative detorsion, many still believe that resection is the only treatment option for lobar torsion, given the significant risks of preservation.[5] With the ML being composed of only 2 pulmonary segments, its sacrifice, from a pulmonary function standpoint, is likely of lesser import to the patient than the potential significant morbidity associated with attempted preservation. Furthermore, a definitive management such as a formal anatomic middle lobectomy is to be preferred, in most cases, given that patients in these circumstances will often already be acutely ill and have significant comorbidities such as hepatic insufficiency.



VIDEO 1. A short video depicting the intraoperative findings of spontaneous middle-lobe torsion requiring right posterolateral thoracotomy and middle lobectomy. Video available at: https://www.jtcvs.org/article/S2666-2507(23)00129-3/fulltext.

CONCLUSIONS

Spontaneous ML torsion is a rare phenomenon that is difficult to recognize both clinically and radiographically. As a result, the majority of cases require pulmonary resection and are associated with high morbidity and mortality. A high index of suspicion must be maintained to avoid delays in diagnosis and treatment.

Conflict of Interest Statement

The authors reported no conflicts of interest.

The *Journal* policy requires editors and reviewers to disclose conflicts of interest and to decline handling or reviewing manuscripts for which they may have a conflict of interest. The editors and reviewers of this article have no conflicts of interest.

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Key Words: lobar torsion, spontaneous middle lobe torsion, general thoracic surgery, lobectomy, pleural effusion