Middle Lobe Torsion after Unilateral Lung Transplant

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ABSTRACT

Lobar torsion is well documented after pneumonectomy, but is very rare after lung transplant. To the best of our knowledge, this is the twelfth reported case of lobar torsion after lung transplant. In our case, bronchoscopies and chest radiographs were inconclusive; however, CT scan clearly demonstrated findings consistent with right middle lobe torsion. We review the literature and discuss the epidemiology, clinical presentation, imaging features, and treatment considerations for this condition. We also propose that if a clinical picture could be secondary to torsion and bronchoscopies and chest x ray are inconclusive that a CT scan should be obtained as soon as possible since early recognition increases the likelihood of being able to successfully detorse the lung and avoid lobectomy.

CASE REPORT

The patient is a 72 year old male with a history of end-stage pulmonary fibrosis with progressive deterioration of his clinical status and increasing daily requirements for oxygen presented for bilateral lung transplant. The implantation of the right lung was very difficult due to the large size of the donor lung and deep cavity of the recipient’s right chest. Additionally, patient’s left pleural space was much smaller and the heart was deviated to the left making it technically difficult to transplant his left lung because it would require another run of cardiopulmonary bypass which potentially could compromise the function of the right lung and patient outcome. Hence only a right lung transplant was performed to minimize complications. After the procedure the patient was transported from the operating room to the intensive care unit on mechanical ventilation. Oxygen saturation remained 100% during transfer and there were no immediate post-operative complications. Shortly after arriving to the intensive care unit the patient began having episodes of hypotension. Vaspressors were being administered to maintain mean arterial pressure above 65 mmHg. An arterial blood gas was obtained and revealed proper oxygenation. A bronchoscopy was performed which revealed a normal anastomosis. A chest radiograph was obtained after the procedure which demonstrated right lower lung base peripheral opacity (Figure 1). Patient’s pulmonary arterial pressure was elevated at 35-37 mmHg.

The morning after the procedure the patient was no longer sedated and was being weaned off of vasopressors. Then his oxygen saturation dropped to 78% with thick bloody secretions noted in the endotracheal tube. A repeat bronchoscopy revealed a normal anastomosis and a large left mucous plug, but no conclusive evidence for why the patient was not improving. Clinically, the patient was anxious and this was thought to be contributing the patient’s desaturation. A few hours later the patient was still desaturating with minimal exertion. Venous oxygen saturation was noted to be approximately 40%. He had leukocytosis of 19,000, and he was on antibiotics. The patient was tachypneic with a
respiratory rate of 35 breaths per minute. Patient was re-sedated, was diuresed as blood pressure allowed, and continued on aggressive pulmonary hygiene with nebulizers. A repeat chest radiograph was obtained and revealed persistent right lower lung base consolidation.

On postoperative day two, the patient failed weaning of vasopressors and was given albumin for his labile blood pressure. Patient’s mean pulmonary arterial pressures were still elevated. Patient was on 5 parts per million of nitric oxide and was started on 10mg sildenafil TID for pulmonary hypertension. On post-operative day two, the patient was no longer experiencing desaturations but remained on nitric oxide, vasopressors, and mechanical ventilation.

On post-operative day three the patient started having repeat instances of desaturations with continued labile blood pressures. The patient’s pulmonary arterial pressures were still elevated and were now above 40 with an increasing oxygen requirement on 50% FIO2. A repeat bronchoscopy was performed which demonstrated tapering of the distal airways with bilateral huge mucous plugs. The patient was continued on conservative management.

On post-operative day four, a chest radiograph once again revealed persistent consolidation of the right lower lung zone. Computed Tomography (CT) was obtained which revealed abnormal posterior displacement of the right middle lobe with anterior displacement of the right lower lobe. There was curvilinear bronchovascular structures and tapering of the right middle lobe bronchus compatible with right middle lobe torsion. The patient was taken to the operating room and right middle lobe torsion was confirmed. A right middle lobe pneumonectomy was performed. Three weeks after the procedure the patient is still alive in the hospital and has required mechanical ventilation but is clinically improving.

DISCUSSION

Etiology & Demographics:
Simply stated, lung torsion occurs when a lung lobe is able to twist around its pedicle. Lung torsion is a very rare entity with most cases being documented after lobectomy with only an incidence of less than 0.5% even in this setting (1). Torsion after lung transplantation is much more rare with our research revealing only three documented cases before 2009 (2,3,4). In 2009, there was a case series of four patient’s published documenting lung torsion status post bilateral lung transplantation (5). Since then there has only been an additional four cases published of torsion after lung transplantation for a total of eleven documented cases with eight of them being in the last six years (6,7,8,9). This suggests lung torsion is becoming an increasingly recognized dreaded complication that can occur after lung transplantation. Given the rarity of lobar torsion after lung transplant, information about this condition comes from the study of the collective case reports, and this discussion will review the prior documented cases as compared to our case.

Since torsion is essentially twisting of a lobe or lobes upon its pedicle, the major risk factors for lobar torsion are factors that allow the lung to have increased mobility. This is why the highest reported incidence of lobar torsion is after pneumonectomy. Lung parenchyma is resected and there is relative increased space in the thoracic cavity allowing increased mobility of the remaining lobes of the lung. Thus in this setting fixation of the remaining lobes is routinely performed to decrease the mobility of the respective lobes subsequently decreasing the risk of torsion. However, after transplantation the major risk factors postulated are the normal division of the pulmonary ligament during allograft procurement which normally prevents torsion of the lung around the hilum, size discrepancy between a small donor lung and a relatively larger recipient’s thoracic cavity, and the presence of a complete major fissure (3,4). When reviewing the prior eleven cases, the most common risk factor present was the presence of a complete fissure which was documented to be present in six of the eleven cases. Unfortunately, this fact might not have been known in every case, and in our case it is uncertain if there was a complete fissure or not. The complete fissure being one of the most prevalent risk factors is why after lung transplant that the left lobe is thought to torque most frequently because the incidence of complete fissures is highest on the left ranging from 27-82% (6,10).

The increased risk associated with discrepancy in size between a small donor lung and a relatively larger chest cavity is easily rationalized because the size discrepancy invariably will allow increased mobility of the lobes which increases the risk for torsion (5). For example, Gilkeson’s et al case report was an instance of the donor allograft being significantly smaller than the recipient’s hemithorax and was considered to be the major risk factor for torsion (3). In our particular case, the surgeon actually noted that the transplant of the donor lung was technically difficult due to its large size and the patient having a deep right thoracic cavity. Rather too large of a donor lung could play a role is not as easily rationalized. In our case, considering that the right middle lobe of the allograft is what torsed, it seemingly could be related to the patient’s deep thoracic cavity and/or too large of a donor lung. Seemingly, after transplantation, the large donor lung starts to re-expand and the resulting increased pressure seemingly could have displaced the right middle lobe more posteriorly allowing it to settle into this patient’s deep thoracic cavity. With the resultant displacement of the right middle lobe, the right lower lobe would then be free to move anteriorly. With progressive expansion of the alveoli of the right lung, the right middle lobe would then begin expanding posterior to the right lower lobe and continue to force the right lower lung more anteriorly and cranially torsing the lung counterclockwise. Interestingly there was another case described by Shakoor et al in which the lung allograft was also noted to be large in relation to the patient’s chest cavity. Although, in this case there is a confounding factor because a complete minor fissure was also present (6). Either way, size discrepancy between a donor’s lung and recipient’s chest cavity likely is a major risk factor in the development of torsion after lung transplant.
Clinical & Imaging Findings:
As stated previously, lobar torsion occurs when a lung lobe is able to twist around its pedicle. The degree of torsion, rather it is partial or complete, affects the clinical presentation. Partial torsion is when a lobe or lobes rotate around the hilar structures 90 degrees. Complete torsion is when a lobe or lobes rotate around the hilar structures 180 degrees or more. Torsion is a dangerous complication because it results in compromise of the bronchi, arterial blood supply, and venous-lymphatic drainage with ensuing ischemia and necrosis of the lung parenchyma. However, the lungs are relatively resistant to infarction secondary to a dual blood supply of from the pulmonary and bronchial circulation. Because of this dual blood supply partial torsion does not quickly result in infarction and necrosis and has more of an insidious clinical presentation with clinical signs that can mimic pneumonia or atelectasis such as chest pain, tachycardia, tachypnea, respiratory insufficiency, fever, and leukocytosis. Additionally, ventilation-perfusion defects are generally equally matched in partial torsion, which means there may be no appreciable alteration in any oxygenation or ventilation. Consequently, as in our case, arterial blood gases may be misleadingly normal (12). In complete torsion the onset of pulmonary gangrene is more rapid and the presentation of chest pain, hemoptysis, and hypoxia is more severe. Considering the variability and nonspecific presentation, a high degree of suspicion is needed in every case.

Felson describes the typical findings of lobar torsion on chest radiograph or computed tomography as collapsed or consolidated lobe that occupies an unusual position, abnormal pulmonary vascular pattern often with inverted hilum, bronchial cutoff or distortion, lobar air-trapping, displacement of the hilum in an inappropriate direction relative to the atelectatic lobe involved, and a non-aerated lobe in an unusual position with or without accompanying compensatory hyperinflation of the surrounding lung (11). In every case reviewed from the literature the chest radiograph revealed consolidation which was often confused with atelectasis. In our case, the chest radiograph demonstrated a right lower lung zone consolidation which similar to the other cases was also thought to most likely represent atelectasis. Retrospectively, the consolidation does not fit the normal appearance of atelectasis and signs of right sided volume loss were not present including the minor fissure was not displaced, there was no right sided rib crowding, and the mediastinum was not displaced to the right side (Figure 1).

In our case, the combination of a chest radiograph revealing a suspicious opacity, persistent leukocytosis despite antibiotics, worsening acute pulmonary hypertension, and hypoxemia collectively raised clinical concern for lobar torsion. Bronchoscopy is usually the first step in evaluation of a patient status post lung transplant that is having respiratory insufficiency because it can be performed easily and quickly at bedside. Generally bronchoscopy is thought to be highly sensitive in detecting torsion and typical findings include occluded or distorted bronchus without evidence of endobronchial lesion or mucous plugging, abnormal rotation of affected bronchus, and hyperemia of the mucosa. In our case, three bronchoscopies were performed which were all inconclusive and did not suggest torsion. Then a CT was obtained which demonstrated a consolidated right middle lobe in abnormal posterior position with corresponding anterior displacement of the right lower lobe. There was narrowing of the right middle lobe bronchus with a curvilinear abnormal posteriorly directed course of the right middle lobe bronchus with a corresponding anterior course of the right lower lobe bronchus. These findings are specific for a diagnosis of right middle lobe torsion (Figure 2, 3, 4, 5).

Reviewing the other eleven documented cases, similar results were obtained with bronchoscopy not always suggesting torsion. Although this is a very small sample size, bronchoscopy was inferior to CT examination in sensitivity and specificity for lobar torsion. Thus, in these rare cases when patient is not improving clinically and bronchoscopy does not clearly delineate a cause for patient’s symptoms, CT scan of the lung should be considered immediately for further characterization.

Treatment & Prognosis:
After torsion was diagnosed in our case, open thoracotomy was performed which is currently the standard of care. Intraoperatively the decision can be made to perform detorsion or lobectomy which is largely based upon the surgeon’s discretion on the viability of the affected lobe. Depending on rather the lung is completely torsed or partially torsed affects the viability of the lung. In patients with complete torsion, the onset of pulmonary gangrene is rapid, and unless immediate surgery is performed, then the lung will not be viable (3). In the eleven cases reviewed after lung transplant, three of them were able to successfully undergo detorsion and all three were alive at follow up. Collin’s et al patient had lobectomy and died sixteen months later of other complications (2). Grazia’s et al patient had lobectomy and died within two months secondary to severe sepsis and acute respiratory distress (4). Detorsion in itself has inherent risks and the revascularization that occurs may cause an acute release of toxic substances which is typical of ischemic lung reperfusion injury. It was generally thought that if detorsion was not done within the first few hours of torsion than lobectomy was preferred to minimize the risk of reperfusion injury. It was suggested by Velmahos et al after a case that required a subsequent lobectomy after detorsion secondary to reperfusion injury that lobectomy was generally preferred over detorsion (12). Grazia et al concurred that lobectomy is preferred over detorsion in his case report (4). However, Souilamas et al performed detorsion as many as five days after lung transplantation on one of his patient’s without any postoperative complications (5). As a rule, patients requiring lung transplant do not have significant quantity of spare pulmonary reserve; thus, detorsion is preferred over lobectomy in cases in which the lung is viable. In our case the patient’s right middle lobe was not viable and lobectomy was required (Figure 6).

Ultimately, lobar torsion is increasingly recognized as a serious potential complication of lung transplant. The clinical picture is not always clear and a high degree of suspicion is needed to be able to act quickly and possibly avoid infarction and lobectomy. Chest radiographs are very sensitive and
reveal persistent consolidation that is often confused with atelectasis. It is imperative to recognize if other direct or indirect signs of volume loss are present and if not raise the possibility for lobar torsion. In the limited amount of case reports available, bronchoscopy which is largely considered the best next step in patients with respiratory insufficiency does not have as high of sensitivity as computed tomography in detecting torsion. It is very likely that the advances in high resolution computed tomography scans now makes this the preferred study after lung transplant in patients with respiratory insufficiency. Overall, if patients have consolidation on chest radiograph with a clinical picture that could be secondary to torsion, a CT scan should be obtained as soon as possible since early recognition increases the likelihood of being able to successfully detorse the lung and avoid lobectomy in these patients with very little pulmonary reserve.

**Differential Diagnoses:**

In a patient that is status post lung transplant and is having respiratory insufficiency chest radiograph is the best first step. In cases of lobar torsion an opacity was seen with 100% sensitivity in the reviewed case reports. Unfortunately this is not a specific finding and the opacity was often mistakenly considered to be atelectasis. However, this is understandable because in post-operative patient’s atelectasis is very common. The key to avoiding interpreting the opacity as atelectasis is to remember to carefully scrutinize the radiograph for direct and indirect signs of volume loss. Direct signs of volume loss include displacement of the fissures and vascular crowding, and indirect signs of volume loss include elevation of the diaphragm, rib crowding on the side with volume loss, mediastinal shift to the side with volume loss, and hiliar displacement towards the expected area of volume loss. If there is an opacity without these signs then we propose you characterize the opacity which could represent lobar torsion or even pneumonia. However, bronchoscopy has long been considered the next best step. This is reasonable because it can be done quickly at bedside in a patient who might be unstable. In reviewing the prior eleven case reports, bronchoscopy was neither as sensitive nor specific in detecting lobar torsion as computed tomography. In our case three bronchoscopies were performed before computed tomography and none of them suggested lobar torsion. Thus, CT scan should be considered if bronchoscopy is not conclusive for a patient’s respiratory insufficiency.

CT scan will be able to reliably further characterize an opacity detected on chest radiograph. In cases of atelectasis there will be consolidation with normal lobar anatomic position that enhances the same as surrounding lung parenchyma. Additionally there will be the same findings of volume loss as seen on chest radiograph. It is important to note air bronchograms are not seen with central bronchial obstruction, but can be seen in subsegmental atelectasis.

In cases of lobar pneumonia CT scan will demonstrate consolidation and/or ground glass opacities with air bronchograms in a lobe of normal anatomic position.

Another entity in the differential that could present with respiratory insufficiency in a post-operative patient with a peripheral opacity without signs of volume loss on chest radiograph is pulmonary embolus. If the patient is able to tolerate contrast, then a CT scan with contrast is preferred because a pulmonary embolus is readily diagnosed as filling defects within the pulmonary vasculature, and importantly the lobes of the lung will be in normal anatomical position.

In cases of lobar torsion, CT scan will demonstrate a poorly enhancing consolidated lobe or lobes in abnormal anatomic position and possibly tapered obliteration of the proximal pulmonary artery and accompanying bronchus of the involved lobe. CT with contrast would be preferred to assess the vasculature but is not required since the lobes anatomical position and corresponding bronchus can be appreciated without contrast. In reviewing the prior eleven cases, CT was the most sensitive and specific modality used for diagnosis.

**TEACHING POINT**

Chest Radiographs are sensitive in the detection of an opacity in patients with torsion but this finding is not specific and often mistaken for atelectasis. Direct and indirect signs of volume loss are the key, and if are not present, CT scan needs to be recommended since it has a high sensitivity and specificity for detecting complications of lung transplant. In lobar torsion early recognition could allow the surgeon to successfully detorse the lung and avoid lobectomy.

**REFERENCES**


Figure 1: 72 year old male with right middle lobe torsion. There is right lower lung zone opacity (blue star) on postoperative chest radiograph. Atelectasis is common after lung transplant, and so is at the top of the differential for opacity after transplant. However, indirect signs of volume loss are not present. There is no rib crowding on the right. The most clear indicator though is that the right minor fissure (red arrow) is clearly visualized and is not displaced in the direction of suspected volume loss. Additionally, the mediastinum is not shifted towards the right. Thus, further investigation into the cause of the opacity is required.

Figure 2: 72 year old male with right middle lobe torsion. There is a consolidated right middle lobe (blue star) in an abnormal posterior position with corresponding anterior displacement of the right lower lobe. Additionally, narrowing of the right middle lobe bronchus with a curvilinear abnormal posteriorly directed course of the right middle lobe bronchus (red arrow) is observed coursing posteriorly to the consolidated right middle lobe. The vasculature is not completely evaluated considering the lack of contrast, but these findings are diagnostic of lobar torsion.

Figure 3: 72 year old male with right middle lobe torsion. There is abnormal posterior displacement of the consolidated right middle lobe (blue star) with corresponding anterior displacement of the right lower lung.
Etiology
Generally it is a complication of lobectomy. It is increasingly recognized as a complication that can occur after lung transplant. It is recognized with a mean of 3.5 days post operation with a range from 1-11 days in the previous documented cases. It can also occur spontaneously or after trauma in rare instances.

Incidence
The highest incidence is after lobectomy and is considered to be less than 0.5%. After lung transplant there are only eleven previously reported cases. Including our case there are now 12 reported cases.

Gender ratio
Including our case there is a 5:1 male to female predilection.

Age predilection
It happens to individuals of all ages after lung transplant.

Risk factors
The two major identifiable risk factors are:
1) Presence of complete fissure in donor lung.
2) Size discrepancy between a small donor lung and relatively larger thoracic cavity of recipient.

Clinical presentation
Chest pain, fever, tachycardia, tachypnea, hypoxia, hemoptyisis, decreased breath sounds.

Laboratory findings
Leukocytosis. Arterial blood gas can be surprisingly normal and clinical deterioration with normal abnormal blood gas should raise concern for torsion.

Imaging findings
Chest Radiograph: Consolidation that is often mistaken for atelectasis but without direct or indirect signs of volume loss present.
Computed tomography: Poorly enhancing consolidated lobe of lung in an abnormal anatomic position. There is tapered obliteration of the proximal pulmonary artery and accompanying bronchus of the involved lobe.

Treatment
Detorsion or lobectomy. The surgeon determines the viability of the involved lobe. If it is viable then detorsion can be attempted. If not viable lobectomy is performed.

Prognosis
Prognosis is generally considered poor. However, of the eleven cases only one patient died within two months secondary to acute respiratory distress syndrome.

Table 1: Summary table for pulmonary lobar torsion.
Opacification which does not demonstrate the direct and indirect signs of volume loss and can be in an atypical lobar anatomical position.

Consolidation with enhancement the same as surrounding lung parenchyma. Same findings of volume loss as chest radiograph. It is important to note air bronchograms are not seen with central bronchial obstruction, but air bronchograms are seen in subsegmentalatelectasis.

Opacity with direct signs of volume loss including displacement of the fissures and vascular crowding and indirect signs of volume loss on adjacent structures including elevation of the diaphragm, rib crowding on the side with volume loss, mediastinal shift to the side with volume loss, and hilar displacement.

Consolidation and/or ground glass opacities with air bronchograms in a lobe of normal anatomic position.

Peripheral opacity without signs of volume loss which is known as the Hampton hump. Sometimes, can also appreciate the Fleishner sign, which is enlargement of a pulmonary artery, or the Westmark sign, which is regional oligemia.

CT with contrast demonstrates filling defects in the pulmonary arteries. Additional signs could include enlargement of the pulmonary artery, straightening of the interventricular septum, and an increase in the ratio of right ventricular cavity to left ventricular cavity >1 which all are secondary to increased pulmonary arterial pressure. Additionally, a peripheral wedge shaped opacity that is pleural based with its apex pointing towards the hilum could be seen and is consistent with pulmonary infarction.

Table 2: Differential diagnosis table for pulmonary lobar torsion.

CT = computed tomography
TID – Three times per day

Lobar torsion; Lung transplant; Complication of Lung Transplant; Atelectasis; Lobectomy

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<th>Chest Radiograph</th>
<th>Computed Tomography</th>
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<td><strong>Pneumonia</strong></td>
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**Abbreviations**

CT = computed tomography
TID – Three times per day

**Keywords**

Lobar torsion; Lung transplant; Complication of Lung Transplant; Atelectasis; Lobectomy