Nil Intervention is at Times the Best Intervention

Benign emptying of pneumonectomy space

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Abstract

A sudden drop of air-fluid level in the pneumonectomy space in the absence of a bronchopleural fistula and pleural infection is termed benign emptying of the post pneumonectomy space (BEPS). We herein report a case of benign emptying of pneumonectomy space, briefly review the literature and enumerate possible mechanisms for the same. It is an extremely rare entity and follows a benign course. Patients with BEPS are clinically stable, afebrile with no fluid expectoration, and have a normal white blood cell count. Bronchoscopy reveals an intact bronchial stump and pleural fluid cultures are often sterile. Close monitoring and early detection of a bronchopleural fistula is the management. Clinicians should keep BEPS among the differential diagnosis in case of a drop in the air-fluid level of the post pneumonectomy space. Awareness of this entity is crucial as it helps prevent unnecessary and morbid surgical interventions.

Keywords: Pneumonectomy; BEPS; Bronchopleural Fistula; India.
Introduction

Pneumonectomy, the surgical resection of an entire lung is associated with significant perioperative morbidity and mortality. The understanding of the normal physiology and chest radiography post pneumonectomy is imperative to pick up complications early. Immediately following pneumonectomy, air fills the postpneumonectomy space. Within 24 hours, the ipsilateral diaphragm becomes slightly elevated, mediastinum gets slightly shifted to the contralateral side and fluid accumulation commences in the postpneumonectomy space. In the early postoperative period, fluid accumulation occurs at the rate of one to two intercostal spaces per day.\(^1\) Within 2 weeks, 80-90% of the space gets filled with fluid.\(^1\) A drop in the air-fluid level should evoke the suspicion of a possible bronchopleural fistula.\(^2\) Similar radiologic picture in the absence of bronchopleural fistula and associated infection points towards a lesser known entity, benign emptying of pneumonectomy space.\(^3\) We herein report a case of benign emptying of pneumonectomy space, briefly review the literature and enumerate possible mechanisms for the same.

Case report

A 28-year young woman, a known case of type 1 diabetes mellitus was admitted in view of diabetic ketoacidosis in the month of September 2020. She presented with complaints of multiple episodes of vomiting. She had no history of cough with expectoration, breathlessness, chest pain, or fever. The clinical diagnosis of DKA was made in view of elevated plasma glucose, positive urinary ketones and acidosis in arterial blood gas analysis.

Her history revealed that she was diagnosed to have sputum smear-positive pulmonary tuberculosis with left-sided secondary spontaneous pneumothorax in august 2017 (Figure1A). Left-sided chest tube insertion was done at that time for the pneumothorax. She also received categoryI weight-based anti-tuberculosis treatment for 6 months and was declared cured. In view of left lung persistent hydropneumothorax with pleural thickening (a trapped lung) in spite of chest tube in situ (Figure1B) and tuberculosis drug treatment she underwent left-sided pneumonectomy in July 2018 through posterolateral thoracotomy. She was discharged on the fifth postoperative day without chest tube or any complications. A chest radiograph taken in her follow up visit two weeks after discharge demonstrated the expected fluid increase in the post pneumonectomy space (Figure 1C).
However, a routine chest radiograph taken during this admission revealed a drop in the air-fluid level in the left hemithorax (Figure 1D). This incidental finding raised the alarms of a possible bronchopleural fistula. But surprisingly, she had no specific respiratory complaints. Her pulse rate was 70 beats per minute, blood pressure 110/70 mmHg, room air oxygen saturation was 96%, and respiratory rate 18 breaths per minute. She was afebrile with benign physical examination findings except for decreased air entry on the left side of chest. So, we elected to work up and not hurry up with a chest tube insertion. Her total white blood cell count and routine biochemistry were reported normal. Pleural fluid diagnostic aspirate was sent for culture which was reported sterile.

The patient did not receive any course of antibiotics as she did not have any clinical features suggestive of infection. She was afebrile, no purulent expectoration, normal total leucocyte count and sterile pleural fluid culture report.

Given the ongoing COVID pandemic, a virtual bronchoscopy reconstruction was done from high resolution computed tomography of the thorax instead of diagnostic flexible bronchoscopy to rule out BPF (Figure 2). It showed linear thin fenestrated membranes partially occluding the lumen of the left main bronchus, beyond which soft density was seen, probably collapsed lung parenchyma or scar tissue with suture material. No obvious bronchopleural fistula was noted.

These favored a diagnosis of benign emptying of pneumonectomy space. She was conservatively managed and duly discharged once plasma glucose levels were controlled. Repeat chest radiograph after two weeks revealed reaccumulation of fluid in the postpneumonectomy space, reinstating our diagnosis (Figure 1E). Informed written consent was obtained from the patient and attenders to share patient information and radiology images.

**Discussion**

The possible pulmonary complications of pneumonectomy are pulmonary edema, postpneumonectomy syndrome and intraoperative spillage. The extrapulmonary complications include post pneumonectomy empyema, post pneumonectomy syndrome, esophagopleural fistula, chylo thorax, acute hemothorax, contralateral pneumothorax and bronchopleural fistula is one among these. A drop in the air-fluid level within the post pneumonectomy space in an erect chest radiograph is often considered pathognomonic of bronchopleural fistula (BPF). It is a lethal condition and warrants early intervention in the form of prompt and urgent drainage.
by chest tube insertion. The reported incidence of BPF after pneumonectomy is 0-9% and the associated mortality rate is 16-23%. Patients with BPF usually have clinical features of cough with fluid expectoration, fever, and breathlessness. Some may present with new infiltrates in the contralateral lung due to trans-bronchial spill. Other radiographic presentations include persistent or increasing pneumothorax despite adequate drainage via chest tube, progressive mediastinal or subcutaneous emphysema, or mediastinal shift to the contralateral side. A decrease in the height of the fluid column by 1.5cm or more is suggestive of BPF. A decrease in the fluid level less than 1.5cm can be ignored unless there is an associated mediastinal shift to the contralateral side or features of infection.

The confirmation of BPF can be done using bronchoscopy and/or contrast enhanced chest tomography (CECT). Other methods include contrast bronchography, intrapleural methylene blue administration, and ventilation scintigraphy. Bronchoscopic examination of the bronchial stump is the most commonly used method to confirm the diagnosis of BPF. To identify BPF, bronchial stump immersed in saline is visualized under positive pressure ventilation or can be done by visualization of continuous air bubbles on lavage. Once diagnosed, the fistula needs to be repaired. CT chest with airway virtual reconstruction is an alternative and safer option to confirm or rule out BPF as it is easier to perform, faster and also allows better visualization of the fistula tract.

A similar drop in the air-fluid level of pneumonectomy space without the existence of BPF is termed benign emptying of pneumonectomy space (BEPS). It is a rare condition which may be misdiagnosed and mismanaged as a BPF. The calculated incidence of BEPS is reported as 0.65 of pneumonectomies. Such patients are often asymptomatic, clinically stable, and do not require any urgent intervention. They are often afebrile with normal white blood cell count and sterile pleural fluid culture. Bronchoscopy reveals an intact bronchial stump with no fistula.

The possible mechanism behind BEPS might be (1) the presence of an occult fistula that heals spontaneously before seeding and the occurrence of infection, (2) the presence of a valve-like fistula of a small caliber that allows the passage of only air and does not allow the fluid to enter the airway or a large number of bacteria to enter the pleural cavity to fuel an infection. A transient microscopic and spontaneously healing fistula is the most accepted mechanism. The transit of air into the pleural space causes an increase in the intrapleural pressure forcing the fluid out of the hemithorax into the surrounding cavities or tissues. Three potential routes for
the pleural fluid to break free from the pleural space are (1) congenital diaphragmatic fenestrations in individuals with porous diaphragm syndrome (2) diaphragm and peritoneal defects formed at the time of extra pleural pneumonectomy and diaphragm reconstruction (3) failure to produce watertight chest wall closure helps pleural fluid to escape into the surrounding soft tissues, often resulting in an entity called woody chest wall.\textsuperscript{8,9} (1) and (2) allow the egress of pleural fluid into the abdomen.

In 2011, Merritt et al surveyed 28 leading thoracic surgeons across the United States to acquire an estimate of the incidence and varied clinical presentations of BEPS. Based on their observations, a strict clinical and laboratory criterion was described to confidently diagnose BEPS.\textsuperscript{3} The criteria include absence of fever, normal white blood cell count, no fluid expectoration, negative bronchoscopy, and negative pleural fluid cultures if performed.\textsuperscript{3} Our patient met the afore-mentioned criteria. It is important to note that a diagnostic pleural tap could have been avoided in this case as she did not have any clinical features suggestive of infection. Interestingly, a higher incidence of BEPS was noted following right lung resections compared to the left, unlike our case. Right-sided predisposition can be due to the shorter length and lesser concealment of the right bronchial stump and greater chances of ischemia when blood is supplied by a single bronchial artery.

BEPS is managed conservatively, and the pneumonectomy cavity quickly refills over time. Watchful waiting is the right approach. Such patients should be followed up closely with repeat chest radiographs every 1-2 weeks until pleural fluid reaccumulates.\textsuperscript{3} Patients should be counseled of the red flag signs suggestive of BPF and concomitant infection of the pneumonectomy space. Empirical oral antibiotics are not recommended in BEPS.

**Conclusion**

Clinicians should keep BEPS among the differential diagnosis in case of a drop in the air-fluid level of the post pneumonectomy space. A drop in air fluid level of the post pneumonectomy space is not always a pointer to bronchopleural fistula and does not warrant an intercostal drain placement or a surgical intervention. It can just be a benign emptying of pneumonectomy space which does not require any intervention. A right diagnosis of BEPS, when clinched early, will avoid unnecessary, costly, and morbid surgical interventions.
Authors’ Contribution

PU and DPD provided care for the patient. VG and SMP collected the data. DPD supervised the work. MB and PU drafted the manuscript. All authors critically reviewed the manuscript while MB and SMP edited the manuscript. All authors approved the final version of the manuscript.

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References

Figure 1: Chest radiographs showing A - left side pneumothorax with tracheal and mediastinal shift to the right, B – persistent left hydropneumothorax with left chest tube in place, C – totally filled up post pneumonectomy space (July 2018), D – drop in the air fluid level in the pneumonectomy space (September 2020), E – reaccumulation of fluid in the post pneumonectomy space (2 weeks later).

Figure 2: Image of virtual bronchoscopy reconstruction showing linear thin fenestrated membranes partially occluding the lumen of the left main bronchus and an intact left bronchial stump with no evidence of bronchopleural fistula.