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CASE REPORT

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 pneumonectomy space (BEPS). We report a 28-year-old female patient who presented to a tertiary care referral centre, in Pondicherry, India in 2020 with multiple episodes of vomiting. Subsequent to a left-sided pneumonectomy due to tuberculosis, she was diagnosed with BEPS. Generally, 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. In terms of management, close monitoring and early detection of a bronchopleural fistula are the key points. BEPS should be a 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; Pleura; Hydropneumothorax; Case Report; India.

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 identify complications early. Immediately following pneumonectomy, air fills the post-pneumonectomy space. Within 24 hours, the ipsilateral diaphragm becomes slightly elevated, mediastinum is slightly shifted to the contralateral side and fluid accumulation commences in the post-pneumonectomy space. In the early post-operative period, fluid accumulation occurs at the rate of 1–2 intercostal spaces per day.[1] Within two weeks, 80–90% of the space is filled with fluid.[1] A drop in the air-fluid level should evoke the suspicion of a possible bronchopleural fistula (BPF).[2] A similar radiologic picture in the absence of BPF and associated infection points towards the lesser known entity of benign emptying of pneumonectomy space (BEPS).[3] This report describes a case of BEPS, briefly reviews the literature and enumerates possible mechanisms.

Case Report

A 28-year-old female patient with type 1 diabetes mellitus was admitted in view of diabetic ketoacidosis (DKA) in 2020 to a tertiary care referral centre, in Pondicherry, India. 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 2017 [Figure 1A]. Left-sided chest tube insertion was done at that time for the pneumothorax. She also received category 1 weight-based anti-tuberculosis treatment for six 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* and tuberculosis drug treatment, she underwent left-sided pneumonectomy in 2018 through posterolateral thoracotomy [Figure 1B]. She was discharged on the fifth postoperative day without chest tube or any complications. A chest radiograph taken during 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 BPF. Surprisingly, she had no specific respiratory complaints. She had a pulse rate of 70 beats per minute, a blood pressure of 110/70 mmHg, room air oxygen saturation of 96% and a respiratory rate of 18 breaths per minute. She was afebrile with benign physical examination findings except for decreased air entry on the left side of chest. Therefore, it was decided to evaluate the cause first and a chest tube was not inserted. Her total white blood cell count and routine biochemistry were reported normal. In view of recurrent vomiting episodes, an ultrasound of her abdomen was done; the results were normal. Pleural fluid diagnostic aspirate was sent

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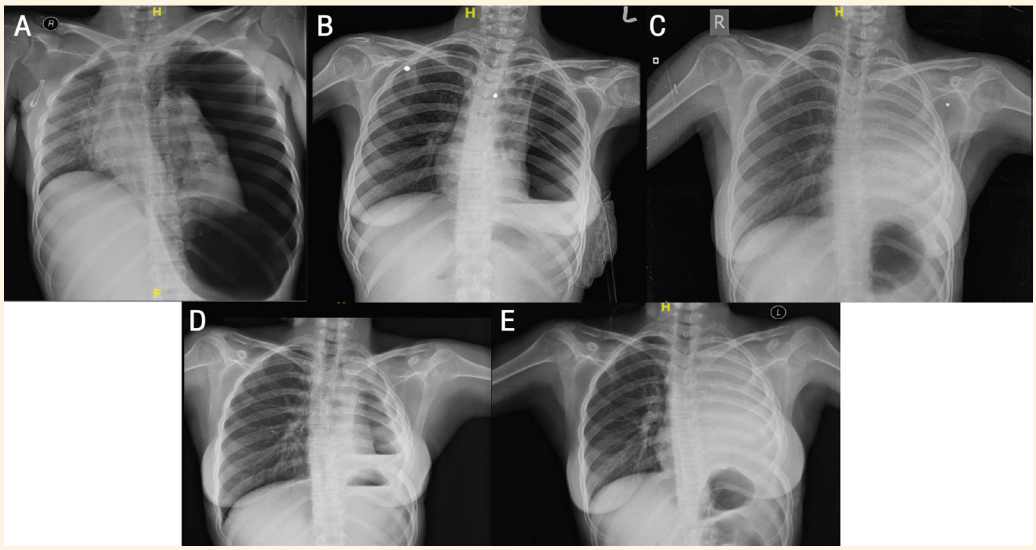

Figure 1: A: Chest radiograph of a 28-year-old female patient showing left side pneumothorax with tracheal and mediastinal shift to the right. **B:** Chest radiograph showing persistent left hydropneumothorax with left chest tube in place. **C:** Chest radiograph showing totally filled post-pneumonectomy space in 2018. **D:** In 2020 there was a drop in the air fluid level in the pneumonectomy space, as seen in the chest radiograph. **E:** Chest radiograph shows reaccumulation of fluid in the post-pneumonectomy space 2 weeks later.

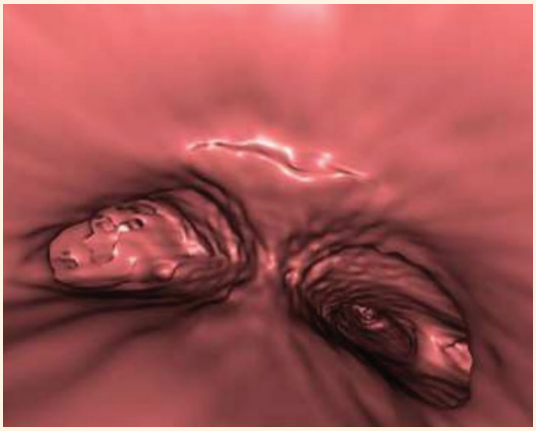

Figure 2: Image of virtual bronchoscopy reconstruction showing linear thin fenestrated membranes partially occluding the lumen of the left main bronchus and an intact left bronchial stump with no evidence of bronchopleural fistula.

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

Given the COVID-19 pandemic, a virtual bronchoscopy reconstruction was done from high resolution computed tomography (CT) 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 findings favoured a diagnosis of BEPS. She was conservatively managed and duly discharged once plasma glucose levels were controlled. Repeat chest radiograph after two weeks revealed a reaccumulation of fluid in the post-pneumonectomy space, confirming the diagnosis [Figure 1E].

Informed written consent was obtained from the patient and attendants for publication purposes.

Discussion

The possible pulmonary complications of pneumonectomy are pulmonary oedema, post-pneumonectomy syndrome and intraoperative spillage. The extrapulmonary complications include post-pneumonectomy empyema, post-pneumonectomy syndrome, oesophagopleural fistlula, chylothorax, acute haemothorax, contralateral pneumothorax and BPF. A drop in the air-fluid level within the post-pneumonectomy space in an erect chest radiograph is often considered pathognomonic of 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%.[3] 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 1.5 cm or more is suggestive of BPF.[4] A decrease in the fluid level less than 1.5 cm can be ignored unless there is an associated mediastinal shift to the contralateral side or features of infection.[5]

The confirmation of BPF can be done using bronchoscopy and/or contrast enhanced chest CT. Other methods include contrast bronchography, intrapleural methylene blue administration and ventilation scintigraphy.[6] 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 visualised under positive pressure ventilation or can be done by visualisation 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 visualisation 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.[3] Such patients are often asymptomatic, clinically stable and do not require any urgent intervention; they are often afebrile with a 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 or (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.[3] 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; and (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.[8,9] The first and second routes 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.[3] The criteria include absence of fever, normal white blood cell count, no fluid expectoration, negative bronchoscopy and negative pleural fluid cultures if performed.[3] The current 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 in the present case. Right-sided predisposition can be due to the shorter length and lesser concealment of the right bronchial stump and greater chances of ischaemia 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 correct approach. Such patients should be followed-up closely with repeat chest radiographs every 1–2 weeks until pleural fluid reaccumulates.[3] Patients should be counseled about the symptoms suggestive of BPF and concomitant infection of the pneumonectomy space. Empirical oral antibiotics are not recommended in BEPS.

Conclusion

BEPS should be a differential diagnosis in cases where there is 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 an indicator of bronchopleural fistula and does not warrant an intercostal drain placement or a surgical intervention. Instead, it may indicate a benign emptying of pneumonectomy space which does not require any intervention. A correct diagnosis of BEPS, when diagnosed 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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