

1 SUBMITTED 30 MAY 22  
2 REVISION REQ. 6 JUL 22; REVISION RECD. 1 AUG 22  
3 ACCEPTED 31 AUG 22  
4 **ONLINE-FIRST: DECEMBER 2022**  
5 DOI: <https://doi.org/10.18295/squmj.12.2022.71>

## 7 Nil Intervention is at Times the Best Intervention

### 8 *Benign emptying of pneumonectomy space*

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#### 19 **Abstract**

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

31 **Keywords:** Pneumonectomy; BEPS; Bronchopleural Fistula; India.

32

33 **Introduction**

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

47  
48 **Case report**

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

54  
55 Her history revealed that she was diagnosed to have sputum smear-positive pulmonary  
56 tuberculosis with left-sided secondary spontaneous pneumothorax in august 2017 (Figure1A).  
57 Left-sided chest tube insertion was done at that time for the pneumothorax. She also received  
58 category1 weight-based anti-tuberculosis treatment for 6 months and was declared cured. In  
59 view of left lung persistent hydropneumothorax with pleural thickening (a trapped lung) in  
60 spite of chest tube in situ (Figure1B) and tuberculosis drug treatment she underwent left-sided  
61 pneumonectomy in July 2018 through posterolateral thoracotomy. She was discharged on the  
62 fifth postoperative day without chest tube or any complications. A chest radiograph taken in  
63 her follow up visit two weeks after discharge demonstrated the expected fluid increase in the  
64 post pneumonectomy space (Figure 1C).

65

66 However, a routine chest radiograph taken during this admission revealed a drop in the air-fluid  
67 level in the left hemithorax (Figure 1D). This incidental finding raised the alarms of a possible  
68 bronchopleural fistula. But surprisingly, she had no specific respiratory complaints. Her pulse  
69 rate was 70 beats per minute, blood pressure 110/70 mmHg, room air oxygen saturation was  
70 96%, and respiratory rate 18 breaths per minute. She was afebrile with benign physical  
71 examination findings except for decreased air entry on the left side of chest. So, we elected to  
72 work up and not hurry up with a chest tube insertion. Her total white blood cell count and  
73 routine biochemistry were reported normal. In view of recurrent vomiting episodes, an  
74 ultrasound abdomen was done and reported normal. Pleural fluid diagnostic aspirate was sent  
75 for culture which was reported sterile. The patient did not receive any course of antibiotics as  
76 she did not have any clinical features suggestive of infection. She was afebrile, no purulent  
77 expectoration, normal total leucocyte count and sterile pleural fluid culture report.

78

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

85

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

91

## 92 **Discussion**

93 The possible pulmonary complications of pneumonectomy are pulmonary edema,  
94 postpneumonectomy syndrome and intraoperative spillage. The extrapulmonary complications  
95 include post pneumonectomy empyema, post pneumonectomy syndrome, esophagopleural  
96 fistula, chylothorax, acute hemothorax, contralateral pneumothorax and bronchopleural fistula  
97 is one among these. A drop in the air-fluid level within the post pneumonectomy space in an  
98 erect chest radiograph is often considered pathognomonic of bronchopleural fistula (BPF). It  
99 is a lethal condition and warrants early intervention in the form of prompt and urgent drainage

100 by chest tube insertion. The reported incidence of BPF after pneumonectomy is 0-9% and the  
101 associated mortality rate is 16-23%.<sup>3</sup> Patients with BPF usually have clinical features of cough  
102 with fluid expectoration, fever, and breathlessness. Some may present with new infiltrates in  
103 the contralateral lung due to trans-bronchial spill. Other radiographic presentations include  
104 persistent or increasing pneumothorax despite adequate drainage via chest tube, progressive  
105 mediastinal or subcutaneous emphysema, or mediastinal shift to the contralateral side. A  
106 decrease in the height of the fluid column by 1.5cm or more is suggestive of BPF.<sup>4</sup> A decrease  
107 in the fluid level less than 1.5cm can be ignored unless there is an associated mediastinal shift  
108 to the contralateral side or features of infection.<sup>5</sup>

109

110 The confirmation of BPF can be done using bronchoscopy and/or contrast enhanced chest  
111 tomography(CECT). Other methods include contrast bronchography, intrapleural methylene  
112 blue administration, and ventilation scintigraphy.<sup>6</sup> Bronchoscopic examination of the bronchial  
113 stump is the most commonly used method to confirm the diagnosis of BPF. To identify BPF,  
114 bronchial stump immersed in saline is visualized under positive pressure ventilation or can be  
115 done by visualization of continuous air bubbles on lavage. Once diagnosed, the fistula needs  
116 to be repaired. CT chest with airway virtual reconstruction is an alternative and safer option to  
117 confirm or rule out BPF as it is easier to perform, faster and also allows better visualization of  
118 the fistula tract.

119

120 A similar drop in the air-fluid level of pneumonectomy space without the existence of BPF is  
121 termed benign emptying of pneumonectomy space (BEPS). It is a rare condition which may be  
122 misdiagnosed and mismanaged as a BPF. The calculated incidence of BEPS is reported as 0.65  
123 of pneumonectomies.<sup>3</sup> Such patients are often asymptomatic, clinically stable, and do not  
124 require any urgent intervention. They are often afebrile with normal white blood cell count and  
125 sterile pleural fluid culture. Bronchoscopy reveals an intact bronchial stump with no fistula.

126

127 The possible mechanism behind BEPS might be (1) the presence of an occult fistula that heals  
128 spontaneously before seeding and the occurrence of infection, (2) the presence of a valve-like  
129 fistula of a small caliber that allows the passage of only air and does not allow the fluid to enter  
130 the airway or a large number of bacteria to enter the pleural cavity to fuel an infection.<sup>3</sup> A  
131 transient microscopic and spontaneously healing fistula is the most accepted mechanism. The  
132 transit of air into the pleural space causes an increase in the intrapleural pressure forcing the  
133 fluid out of the hemithorax into the surrounding cavities or tissues. Three potential routes for

134 the pleural fluid to break free from the pleural space are (1) congenital diaphragmatic  
135 fenestrations in individuals with porous diaphragm syndrome (2) diaphragm and peritoneal  
136 defects formed at the time of extra pleural pneumonectomy and diaphragm reconstruction (3)  
137 failure to produce watertight chest wall closure helps pleural fluid to escape into the  
138 surrounding soft tissues, often resulting in an entity called woody chest wall.<sup>8,9</sup> (1) and (2)  
139 allow the egress of pleural fluid into the abdomen.

140

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

152

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

158

## 159 **Conclusion**

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

166

167

168 **Authors' Contribution**

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

173

174 **Acknowledgements**

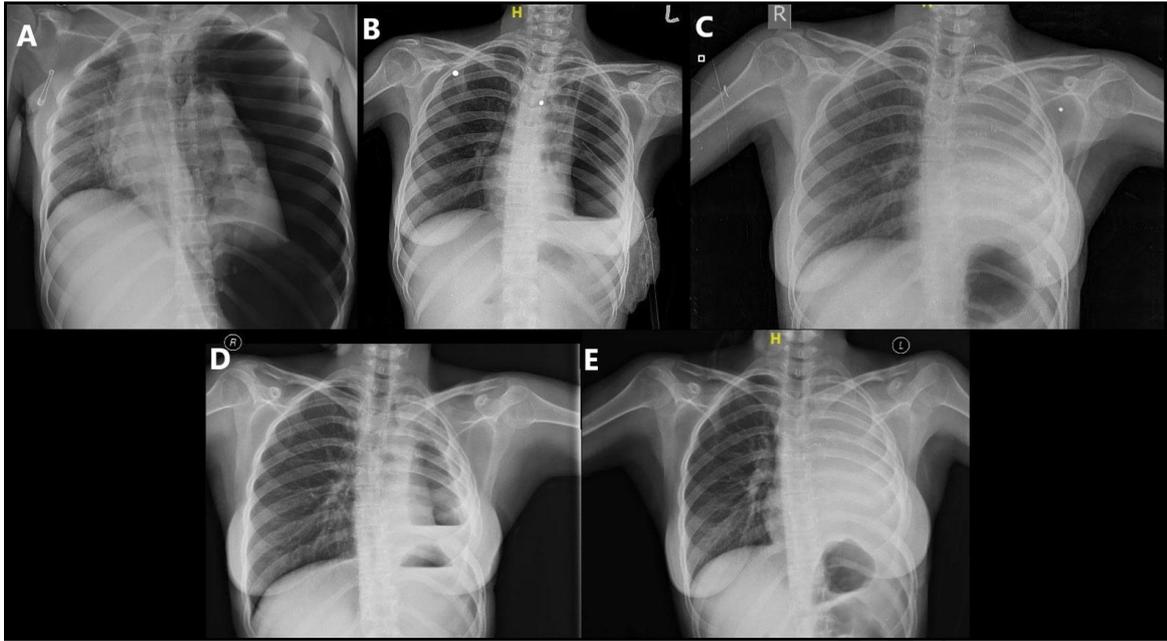
175 We are grateful to Dr Sivaselvi and Dr Lakshmi S residents of the department of Pulmonary  
176 Medicine, JIPMER for the help rendered in diagnosis, workup and patient care.

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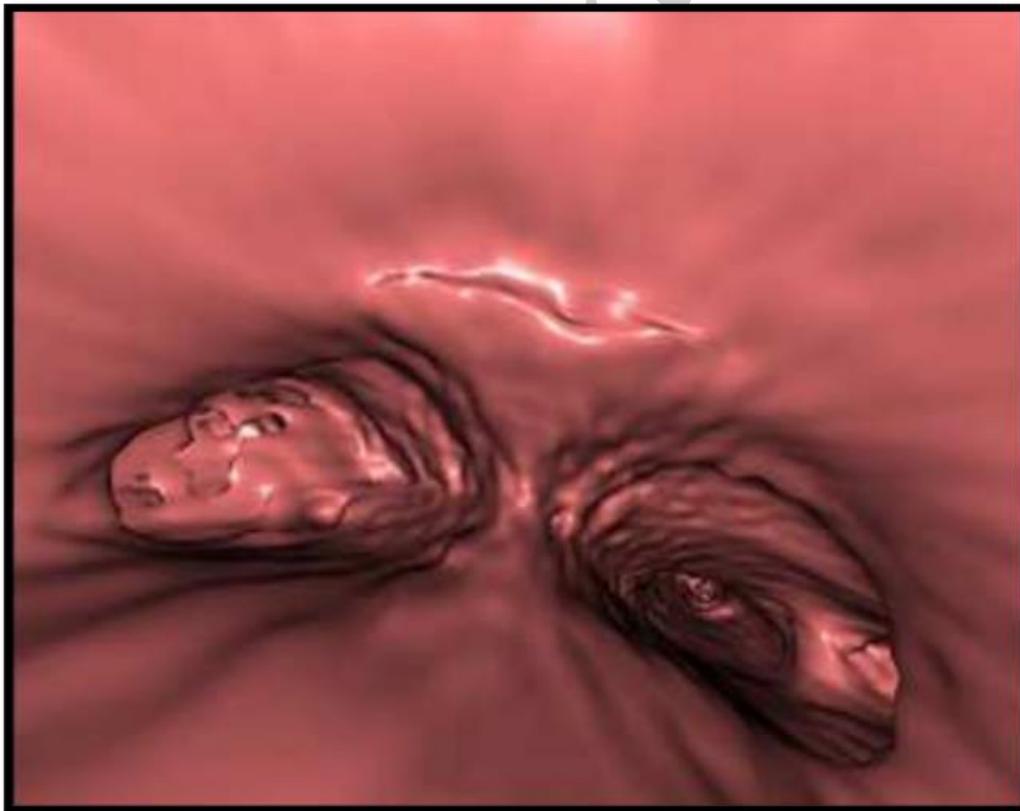
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203 **Figure 1:** Chest radiographs showing A - left side pneumothorax with tracheal and mediastinal  
 204 shift to the right, B – persistent left hydropneumothorax with left chest tube in place, C –totally  
 205 filled up post pneumonectomy space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

208



209

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