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## Case Report

# An unusual cause of acute headache: subarachnoid free air secondary to spontaneous bronchopleurodurosubarachnoid fistula from a Pancoast tumor

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## ABSTRACT

Pneumocephalus and pneumorrhachis are related to transgression of the barriers to the central nervous system. We present a patient with a Pancoast tumor treated with palliative chemoradiation who developed symptomatic spinal and intracranial air caused by spontaneous bronchopleurodurosubarachnoid fistula secondary to direct tumor invasion into the thecal sac. © 2016 the Authors. Published by Elsevier Inc. under copyright license from the University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## Introduction

Pneumocephalus and pneumorrhachis are usually associated with traumatic, neoplastic, or iatrogenic violations of the barriers to the central nervous system. A rare cause is bronchopleurodurosubarachnoid fistula, an unusual and difficult clinical problem with limited case reports, commonly related to trauma or posterolateral thoracotomies [1–10]. We present a case of symptomatic pneumocephalus and pneumorrhachis caused by spontaneous nonsurgical, nontraumatic bronchopleurodurosubarachnoid fistula from a Pancoast tumor invading the spine after palliative chemoradiation.

## Case report

A 53-year-old man with a 35-pack-year smoking history initially presented with neck swelling, throbbing upper back pain, right upper extremity paresthesia, hemoptysis, and 25 pound weight loss over 3 months. Computed tomography (CT) of the chest and subsequent cervical spine magnetic resonance imaging revealed a large right superior sulcus tumor with adjacent vertebral invasion with pathologic fracture of T1 (Fig. 1). Transbronchial biopsy confirmed squamous cell carcinoma, staged as T4N3M1b (stage IV). Palliative chemoradiation was instituted a week later.

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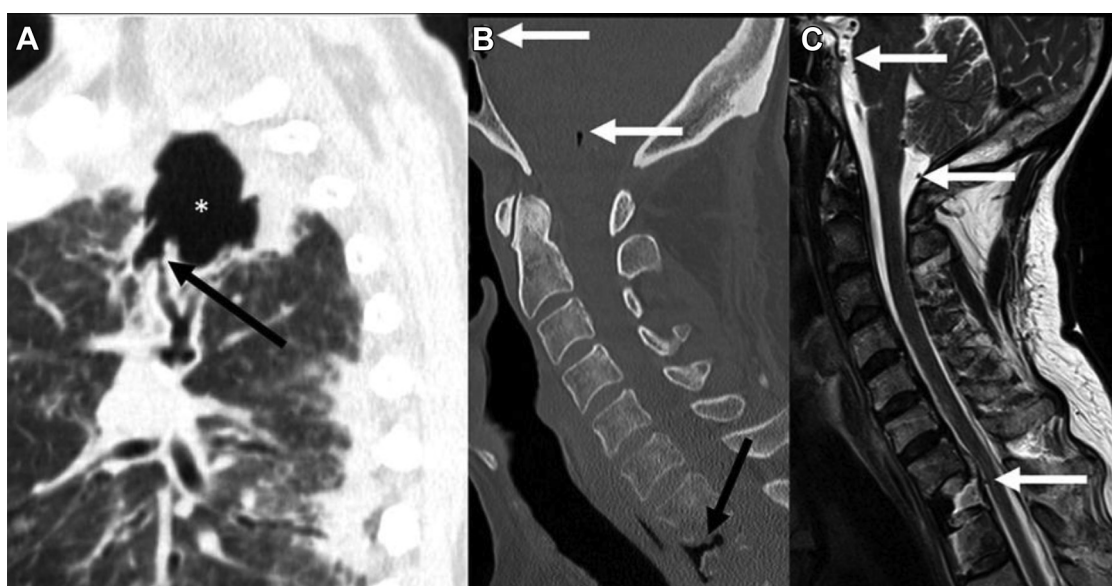
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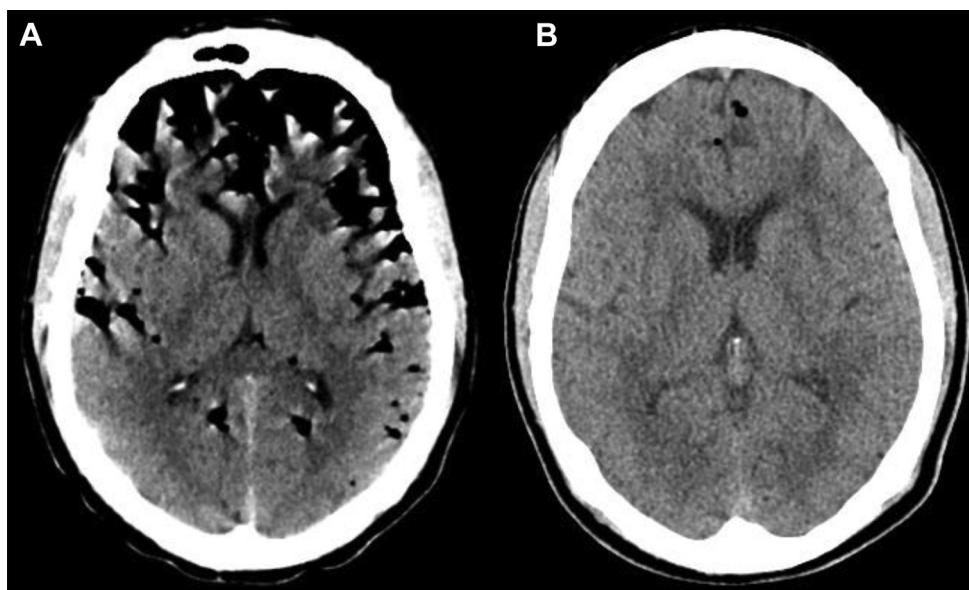
**Fig. 1** – Coronal (A) and axial (B) contrast enhanced CT chest, and sagittal CE T1WI fat suppressed MRI cervical spine (C). Initial presentation: large Pancoast tumor with vertebral invasion (white arrows, A, B) and pathologic fracture of T1 (black arrow, C).

The patient received 3200 cGy and 2 cycles of carboplatin and/or paclitaxel before presenting to the emergency department a month later with acute upper back pain and

severe positional headache following a bout of coughing. Head CT demonstrated extensive pneumocephalus (Fig. 2). Chest and cervical spine imaging demonstrated enlarged



**Fig. 2** – Sagittal noncontrast CT chest (A) and cervical spine (B), and sagittal T2WI cervical spine (C). Presentation to emergency department with severe headache after coughing: fistula (black arrows) connecting bronchus, necrotic air-filled tumor (\*), and collapsed T1 vertebra (B). Spinal and intracranial free air (white arrows, B, C).



**Fig. 3 – Axial NCCT head (A) massive pneumocephalus on presentation with headache, and (B) 48 hours later, with significant decreased air.**

necrotic mass with progressive vertebral collapse, epidural invasion and air within the T1 vertebra and thecal sac suggestive of bronchopleurodurosubarachnoid fistula (Figs. 2 and 3).

The patient remained recumbent for 5 days. The headaches subsided, with decreased pneumocephalus on repeat imaging (Fig. 3), allowing for discharge home with C-collar and antibiotics.

The patient subsequently represented 3 months later with tumor progression, including further spinal involvement with cord compression. Pneumocephalus and pneumorrhachis had resolved on repeat imaging. Comfort care measures were instituted, and the patient died.

## Discussion

Pneumocephalus and pneumorrhachis are common on imaging after trauma and surgery. Distinction between intradural and epidural spinal air is important, as the former is associated with increased morbidity [1].

Lung squamous cell carcinoma in our patient had eroded through the bronchial system and into the thoracic spine, with dural penetration and subarachnoid extension (Fig. 2). Sequential CT and MR imaging of the craniospinal axis was diagnostic in this case. When the source of the fistula is not clear, CT or radioisotope cisternomyelography may assist in diagnosis [4,7,10].

Due to severe headaches and cord compression, 2 surgical options were considered: posterior decompression with duraplasty and sealing of the vertebral body and/or placing a lumbar drain. Conservative management was elected, given concern for possible tension pneumocephalus caused by positive pressure ventilation in the

operating room, and the possibility of brain herniation before adequate surgical exposure could be achieved. Tension pneumocephalus secondary to communication between an air-containing space and cerebral spinal fluid spaces has been described after thoracic surgery and trauma. Conservative treatment with recumbent head-down positioning has been successful in resolving symptoms [6,8]. Increasing inspired oxygen concentration may reduce the volume of gas collection by driving nitrogen into soluble form; similarly, inhaled nitrous oxide should be avoided to prevent expansion of the gas collections, which could increase intracranial pressure [1].

It is difficult to determine the role that effects of therapy played in the development of the fistula, and whether it would have occurred spontaneously without chemoradiation. Approximately 50% of the reported perioperative cases of fistulas had received neoadjuvant radiation therapy [7]. Only 2 case reports show bronchopleurodurosubarachnoid fistula formation without preceding surgery; both fistulas happened after radiation therapy [5,9].

## Conclusions

Pneumocephalus related to spontaneous bronchopleurodurosubarachnoid fistula from a necrotic Pancoast tumor after chemoradiation is a rare cause of acute headache associated with back pain. Our case illustrates successful conservative management of such fistula. The necrotizing effect of chemoradiation may have predisposed to the development of the fistula. Decision regarding palliative radiotherapy should be made after weighting the risk and benefits for an individual patient.

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