

## Pneumocephalus and Cerebrospinal Fluid Fistula Following Removal of a Superior Sulcus Tumor (Pancoast Tumor)

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**Pneumocephalus resulting from a subarachnoid-pleural fistula following resection of a pulmonary neoplasm is a very rare postoperative complication: we have found only 17 cases in the literature. We report the case of a 65-year-old man diagnosed with a superior sulcus tumor who underwent a left upper lobectomy and costal resection following neoadjuvant radiation therapy. Soon after surgery, the patient developed a subarachnoid-pleural fistula and pneumocephalus that required reoperation and drainage. Recurrence was suspected when chronic aseptic meningitis developed. Another thoracotomy was required to resolve the complication.**

**Key words:** *Pneumocephalus. Pancoast tumor. Postoperative complications. Subarachnoid-pleural fistula. Meningitis.*

**Neumoencéfalo y fístula de líquido cefalorraquídeo tras resección de tumor del *sulcus* superior (tumor de Pancoast)**

**El neumoencéfalo, como consecuencia de una fístula subaracnoideo-pleural, es una complicación muy poco frecuente tras la resección de una neoplasia pulmonar (hemos encontrado únicamente 17 casos descritos en la bibliografía). Se presenta el caso de un paciente de 65 años, intervenido con el diagnóstico de tumor del *sulcus* superior tras recibir radioterapia neoadyuvante, al que se le practicaron lobectomía superior izquierda y resección costal. Como complicación postoperatoria temprana, desarrolló una fístula subaracnoideo-pleural y neumoencéfalo, que requirió reintervención y drenaje. Una meningitis abacteriana crónica llevó a sospechar su recidiva, y fue necesaria una nueva toracotomía para su resolución definitiva.**

**Palabras clave:** *Neumoencéfalo. Tumor de Pancoast. Complicaciones postoperatorias. Fístula subaracnoideo-pleural.*

### Introduction

Pneumocephalus resulting from a subarachnoid-pleural fistula is a rare complication of thoracotomy.<sup>1-17</sup> Cerebrospinal fluid (CSF) fistulae frequently develop as a result of skull-based fractures<sup>12</sup> and are commonly associated with pneumocephalus. However, CSF fistulae in the pleural cavity are very rare. They are described in the literature following open or closed spinal cord injury<sup>18,19</sup> and less frequently following a thoracotomy for resection of pulmonary or chest wall neoplasms.

The first case of pneumocephalus was reported in 1974.<sup>1</sup> A Medline search reveals that since that first publication, 17 cases of CSF fistulae following resection of pulmonary neoplasms have been reported.<sup>1-17</sup> We present a new case of CSF fistula

following pulmonary resection and discuss the signs and symptoms, diagnostic methods, and management of this rare complication.

### Case Description

A 65-year-old man with no relevant medical history presented with a 2-month history of pain in the scapular region, left shoulder, radiating from the brachial plexus region. Symptoms and radiographs were consistent with Pancoast tumor without mediastinal lymph node involvement and fine needle aspiration led to a diagnosis of non-small cell carcinoma. Following a single radiotherapy cycle of 10 sessions with a total dose of 30 Gy and a positive radiological response, the patient underwent a left upper lobectomy with en bloc resection of the posterior segments of the second and third ribs, adjacent intercostal muscles up to the third thoracic vertebra, which was uninvolved, and mediastinal lymphadenectomy. No invasion of the brachial plexus was found. The initial postoperative course was uneventful, with scarce air leakage through thoracic drainage tubes, such that suction was discontinued. On postoperative day 4, copious serous exudate was drained through the tubes and the patient

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Manuscript received December 24, 2003. Accepted for publication February 9, 2004.



Figure 1. Computed tomography scan of the head showing significant pneumocephalus in the frontal subdural, subarachnoid, and intraventricular spaces. In addition, possible signs of cerebral edema can be observed.

developed cervical pain, headache, temporal-spatial disorientation, and a change in mental status. A CSF fistula was suspected and the patient underwent a second thoracotomy. The fistula tract was found in the costovertebral junction with the third thoracic vertebra (T3) and was filled

with bone wax and fibrin sealant (Tissucol®; Baxter Hyland Immuno, Vienna, Austria). A computed tomography (CT) scan of the head (Figure 1) revealed significant pneumocephalus. Following drainage and antibiotic treatment with ceftriaxone and corticosteroids, the patient's neurological state improved. On postoperative day 4, the chest drainage tubes were removed. Pneumocephalus resolved and the patient was discharged on postoperative day 9.

Eleven days after discharge, the patient came to the emergency department with a holocranial headache and a fever lasting 48 hours. Meningitis was suspected and the patient underwent lumbar needle aspiration. Inspection of the CSF aspirate showed a white cell count of 1500/ $\mu$ L (90% polymorphonuclear lymphocytes; glucose 32 mg/dL), but cultures were negative. A loculated pleural effusion at the apex of the left lung, evident on a chest CT scan, was considered a postsurgical change. The patient responded well to a 21-day course of antibiotic treatment.

Four days after discharge, the patient was readmitted with a fever of 38°C, persistent headache, and stiff neck. A magnetic resonance image of the head and CSF analysis confirmed a recurrence of aseptic meningitis. Magnetic resonance of the cervical and thoracic spinal column revealed a tear in the left posterior mediastinum consistent with a T2-3 fistula (Figure 2). A CSF fistula was confirmed with radionuclide cisternography (with diethylenetriamine pentaacetic acid marked with technetium 99), where radionuclide leakage was observed from the subarachnoidal space into the left posterolateral region (Figure 3). Another thoracotomy was performed over the previous incision in the interscapular region. A chamber of intrapleural and extrapleural fluid was found and the orifice was found to be partially filled with wax. The fistula was cleared of debris and repaired with a free muscle flap and surgical adhesive (BioGlue®; Cryolife, Kennesaw, GA, USA). A chest tube without suction and a lumbar drain for CSF decompression were inserted. The postoperative course was favorable and a follow-up CT scan of the head and chest produced no

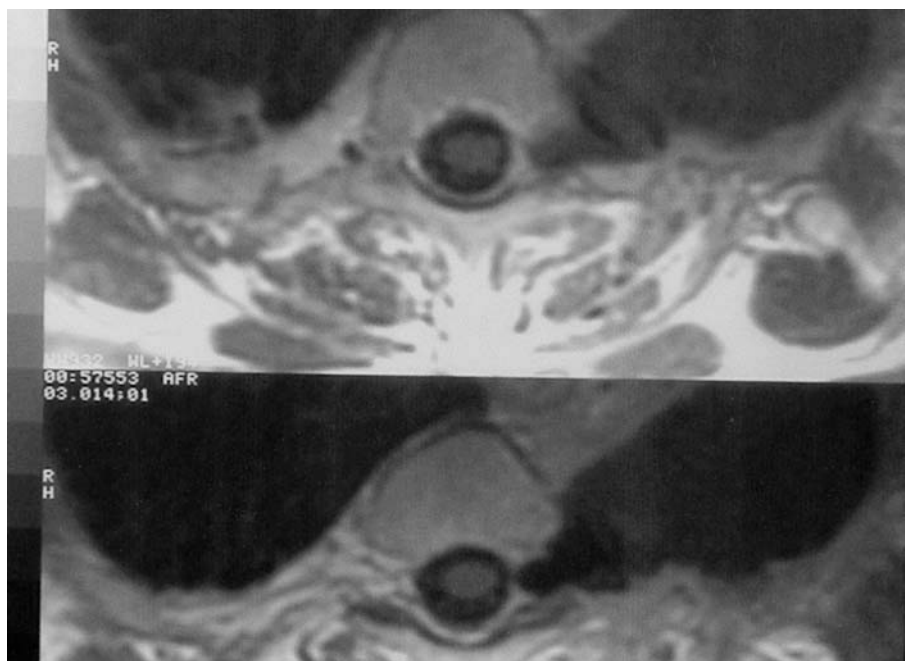


Figure 2. Magnetic resonance image at T2-3 showing a dural tear consistent with CSF fistula.



Figure 3. Isotope cisternography (lateral view) where a radionuclide leak can be observed in the subarachnoid-spinal space at the pleural cavity.

findings, except for postoperative changes. The patient was discharged after 10 days and remained afebrile and asymptomatic at a follow-up examination 3 months later.

## Discussion

Including this case, only 18 cases of CFS subarachnoid-pleural fistulae associated with pneumocephalus following superior sulcus tumor resection have been described in the literature. Nine, our case included, received neoadjuvant radiotherapy.<sup>2,3,5,7,13,14,16,17</sup>

Due to its location, superior sulcus tumors can result in complications and sequelae depending on what structures have been invaded. The tumor must be resected en bloc when ribs and transverse processes are involved in the neoplastic process. There is a risk of tearing the dura mater and the nerve roots when the costovertebral angle is involved.<sup>15</sup> Tears can also occur as a result of costal fractures during thoracotomy.<sup>9</sup> It is likely that the incidence of CSF fistulae is higher than would be indicated by reports in the literature given that small fistulae close spontaneously and remain asymptomatic. Preoperative radiotherapy, which has a necrotizing effect, may inhibit the capacity for the irradiated tissue to repair itself. In this way, radiotherapy may contribute to the formation of fistulae given that 50% of the reported cases, this one included, received neoadjuvant radiotherapy.

In our patient, pneumocephalus was probably the result of the presence of a subarachnoid-pleural fistula and discontinuing suction upon detection of a pulmonary air leak. Most reported cases of pneumocephalus occurred following the removal of the chest drainage tubes and the presence of pneumothorax was discovered with a follow-up x-ray.

Initial pneumocephalus symptoms appear between 1 and 8 weeks after surgery and are characterized by headache (the most common symptom), altered mental state, and, occasionally, reduced states of consciousness. In addition, patients may present with nausea, vomiting, dizziness, stiff neck, aphasia, dysphasia, or hemiplegia.<sup>9</sup> One case reported hyponatremia that may have been related to a syndrome of inappropriate antidiuretic hormone secretion.<sup>14</sup> Symptoms typical of pleural effusion may appear if drainage tubes have been removed. In our case, the first symptom was cervical pain that appeared 4 days postoperatively, followed by headache and altered behavior. The presence of a CSF fistula was suspected based on copious pleural exudate accompanied by deterioration in mental status. Meningitis is the most common complication associated with CSF fistulae and its presence raised the suspicion of fistula recurrence.

Other tests used to confirm the presence of a fistula are based on a pleural fluid analysis characterized by clear transudate, few cells, normal glucose and low protein content, or the measurement of central nervous system-specific ferritin. However, analyses of pleural fluid can produce false negatives.<sup>10,19</sup> The injection of an intrathecal dye, such as fluorescein or indigo carmine, and its subsequent recovery in the chest tubes can also establish the diagnosis.<sup>9</sup> Myelography followed by CT has been found to have better spatial resolution than isotope imaging and has become the definitive test for diagnosing and locating fistulae.<sup>8,9,19,20</sup> However, in this case and in several others reported in the literature, radionuclide cisternography was sufficient to establish the diagnosis.<sup>2,3,7,20</sup> Simple myelography with hydrosoluble contrast can give false positives<sup>2,3,7-9</sup> and CT alone cannot locate the site of the fistula, only yielding evidence of pleural loculations as a result of postoperative changes in the case we report. Magnetic resonance imaging showed better anatomical definition of the fistular area at T2-3.

When a subarachnoid-pleural fistula is recognized during thoracotomy, it should be repaired by ligation with absorbable sutures or, for greater safety, repaired by occlusion with a vascularized muscle pedicle, a free muscle graft, or a free fat graft. Two successful cases of fistula repair have been reported following spinal surgery using a pedicled omental transdiaphragmatic flap.<sup>20</sup> In addition, sealants such as bone wax, fibrin sealant or methyl methacrylate may be used, although use in large volumes can produce brain stem and spinal cord compression. Surgicel<sup>®</sup> and Gelfoam<sup>®</sup> should not be used because of possible postoperative migration and expansion that could lead to brain stem



compression and paraplegia.<sup>21</sup> A CSF leak that is not clear can be managed by applying jugular compression or the Valsalva maneuver.

When a diagnosis is made postoperatively, the treatment depends on the neurological state of the patient and should be individualized. Cases of spontaneous resolution of fistulae with conservative treatment of rest in combination with thoracic<sup>1,4,11,12,17</sup> and lumbar drainage,<sup>15</sup> or, when pneumocephalus was not present, rest in combination with lumboperitoneal drainage,<sup>5</sup> but such cases probably involved small fistulae. When pneumocephalus tension is present with neurological effects, the fistula should be drained. If conservative management fails and the fistula persists, as is often the case, the fistula should be surgically repaired and antibiotic treatment should be administered to prevent infectious complications such as meningitis. In addition to a thoracotomy or thoracoplasty<sup>2,3</sup> and depending on the location of the subdural effusion, the repair can be carried out through a posterior laminectomy<sup>8,9,11,19</sup> and placement of an intradural<sup>7</sup> or extradural patch. In our case, the first approach was not effective, the fistula returned and a muscle flap and BioGlue<sup>®</sup> were required. Following surgery, bed rest is essential and a lumbar drain should be used for decompression of the subarachnoid space.

In conclusion, a CSF fistula following thoracotomy should be suspected when there is a copious amount of clear fluid effusion through the chest tubes and a change in the patient's mental status. The presence of a fistula should be suspected when meningitis is present postoperatively. Prevention is paramount and care should be taken during tumor resection to prevent damage to the dural sleeve. Although radionuclide cisternography can assist in diagnosis, CT myelography is considered the best method to diagnose and locate a CSF fistula.

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