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Case report

Pneumocephalus resulting from chest infection complicated by pleural-subarachnoid fistula

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ABSTRACT

Pleural-subarachnoid fistula is a rare type of Cerebrospinal fluid fistula with less than 60 cases reported in the literature. Here we present a case of 55-year-old female patient, known case of acute myeloid leukemia on chemotherapy, who developed pleural-subarachnoid fistula due to invasive atypical apical lung fungal infection. All of the reported cases in the literature were secondary to trauma or post-surgery. To our knowledge, this is the first reported case of pleural-subarachnoid fistula developed as sequela of fungal infection.

Introduction

Pneumocephalus is the presence of air in the brain or Cerebrospinal fluid (CSF) spaces. It is usually occurring after trauma or post neurological surgery. The presence of fistula between the pleura and subarachnoid space can lead also to pneumocephalus as a rare complication of trauma or spine surgery. None of the reported cases in the literature of the subarachnoid pleural fistula was due to chest infection. Here we are presenting a case of subarachnoid pleural fistula that was a result of aggressive chest infection.

Case report

A 55-year-old female patient known case of acute myeloid leukemia on chemotherapy. She came to our outpatient clinic complaining of headache, vomiting, and blurred vision for one week. Urgent non-enhanced CT head was performed and showed large intraventricular air layering in the bilateral frontal and left temporal lobes. Moreover, multiple scattered air locules were noted involving subarachnoid space, bilateral sylvian and frontal interhemispheric fissures, and prepontine and interpedicular cisterns in keeping with massive pneumocephalus (Fig. 1).

This was unexplained since there was no recent surgery, intervention, or trauma. After reviewing the patient's file and

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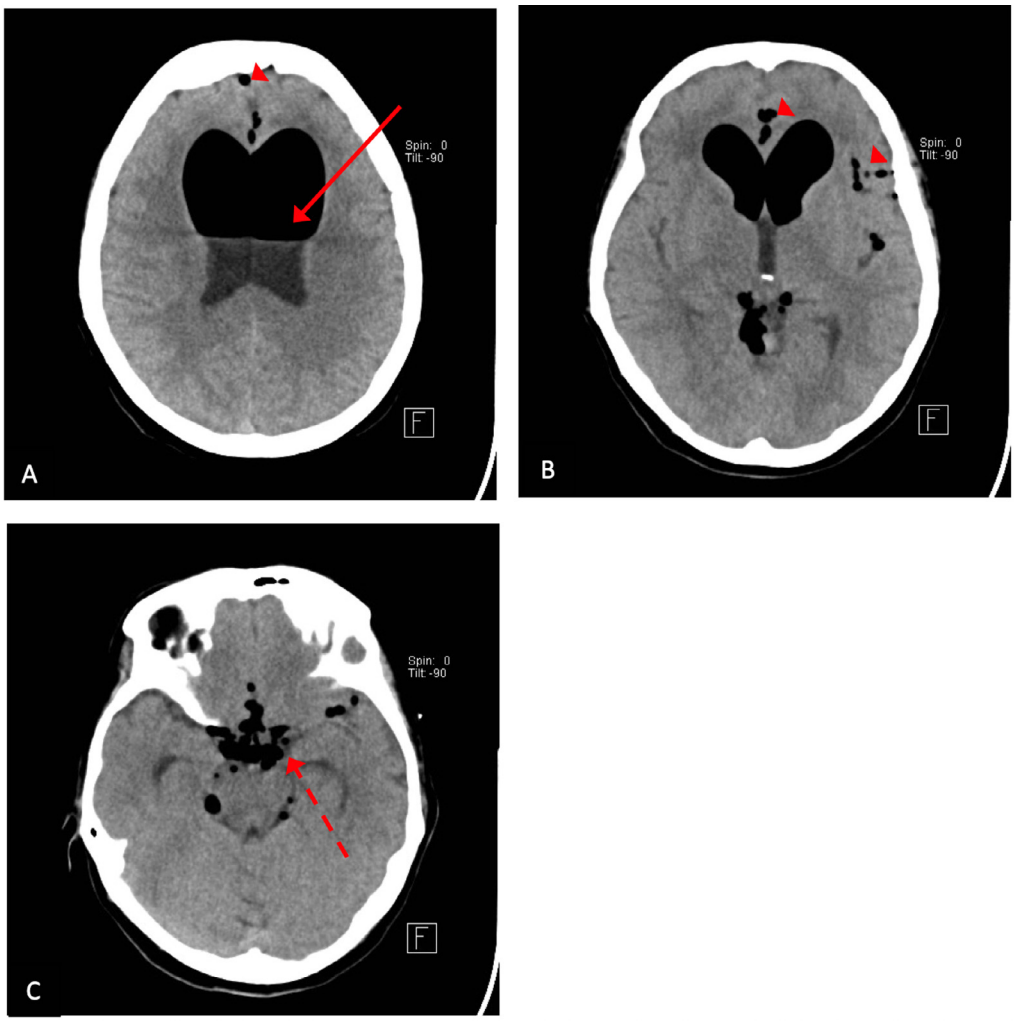

Fig. 1 – non-enhanced CT of the brain: (A, B) a Large amount of intraventricular air with air-fluid levels (arrows). (B, C) Also, Multiple scattered locules are present at subarachnoid space in the bilateral frontal and left temporal lobes (arrowheads) and basal cisterns (dashed arrows).

previous studies we found that she has had a right apical cavitary lesion (Fig. 2)and therefore, anti-fungal treatment was given for few days. Unfortunately, she didn't respond well and a CT-guided biopsy was performed through a posterior approach and it was away from the costovertebral angle (Fig. 3). The biopsy result was "Lichtheimia Corymbifera Complex" so, antifungal was changed to "Amphotericin B and posaconazole". Despite that, a few days later, the patient becomes toxic and complain of right-side chest and shoulder pain with right upper arm numbness. physical examination demonstrated diminished air entry on the right side. MRI Thoracic spine was performed and showed a right paraspinal cavitary lesion with fluid collection at T3-4 level and erosion to the right posterior rib of T3-4 as well as an extension to the right posterior paraspinal region. Abscess formation was suggested. However, no intraspinal extension or vertebral body involvement (Fig. 4). The patient continued anti-fungal and aspiration of the fluid was done. After the aspiration, her symptoms improved few days then again presented with a spike of fever. HRCT was repeated and again showed right upper lung fluid-filled collection with extension into the adjacent posterior chest wall and right posterior paraspinal muscles. Newly developed displaced fractures of the posterior aspect of the 3rd and 4th ribs were noted. Moreover, non-displaced fractures of the right lamina and pedicle of the T3 vertebral body were identified (Fig. 5). Re-aspiration of the fluid collection was performed and the patient was discharged one month later on the same medical treatment after which the patient came to the outpatient clinic with the aforementioned neurological symptoms.

Thereafter, we thought about the possibility of Pleural-subarachnoid fistula and initially recommended chest HRCT followed by Thoracic spine MRI as non-invasive modalities. However, no evidence of fistulous tract has been detected and subsequently, CT Myelogram was recommended to confirm the diagnosis. CT Myelogram showed contrast extravasation tracking from the spinal canal to the right pleural cavity through the right neural foramina at the level of T3-T4, likely secondary to dural tear with fistulation with the adjacent cavitary lesion (Fig. 6). The patient was treated conservatively and

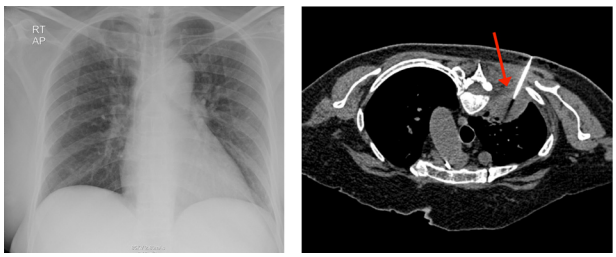

Fig. 2 – AP chest radiograph: right apical cavitary lesion with irregular wall.

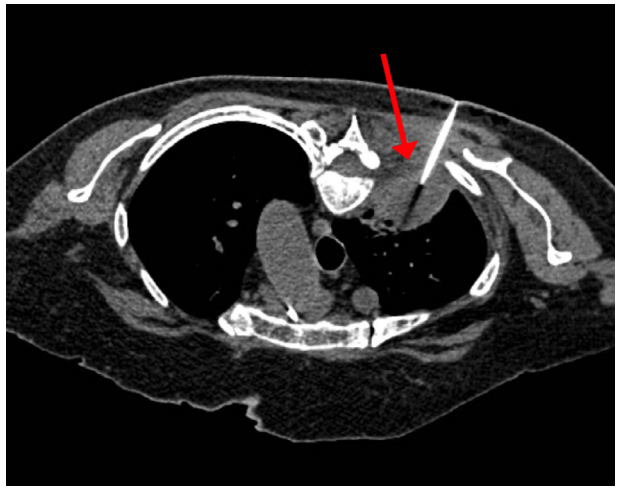

Fig. 3 – non-enhanced CT guided biopsy of right upper lobe posterior cavitary mass (arrow). The image demonstrates the biopsy needle tract away from the spinal canal.

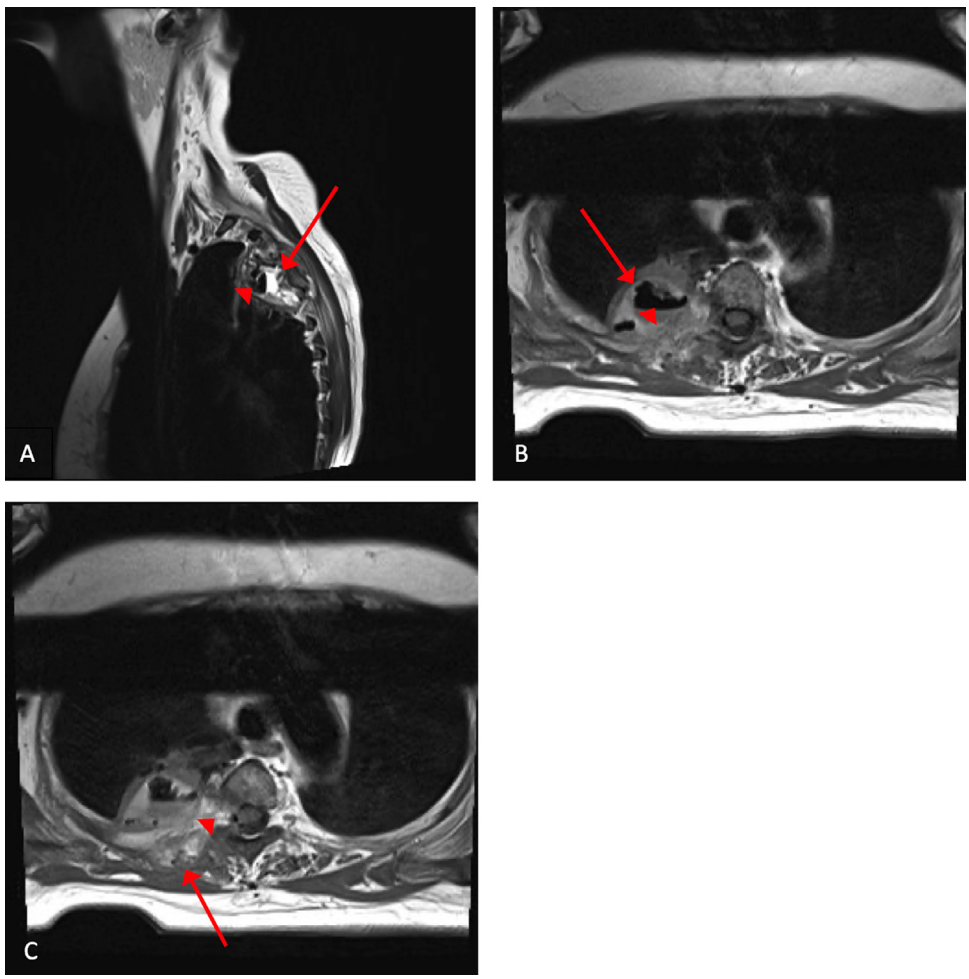

Fig. 4 – Multi sequence MRI of the thoracic spine: (A, B) Well-defined heterogeneous signal intensity cavitary lesion at the right paravertebral region at the upper-level T3 down to lower T4 level (arrows). (C)The lesion showed erosion and involvement of the adjacent posterior right T3-4 with extension posteriorly into the right posterior paraspinal region (arrow). There is limited right neural foramina extension at the T3-4 level (arrowheads). However, no intraspinal extension.

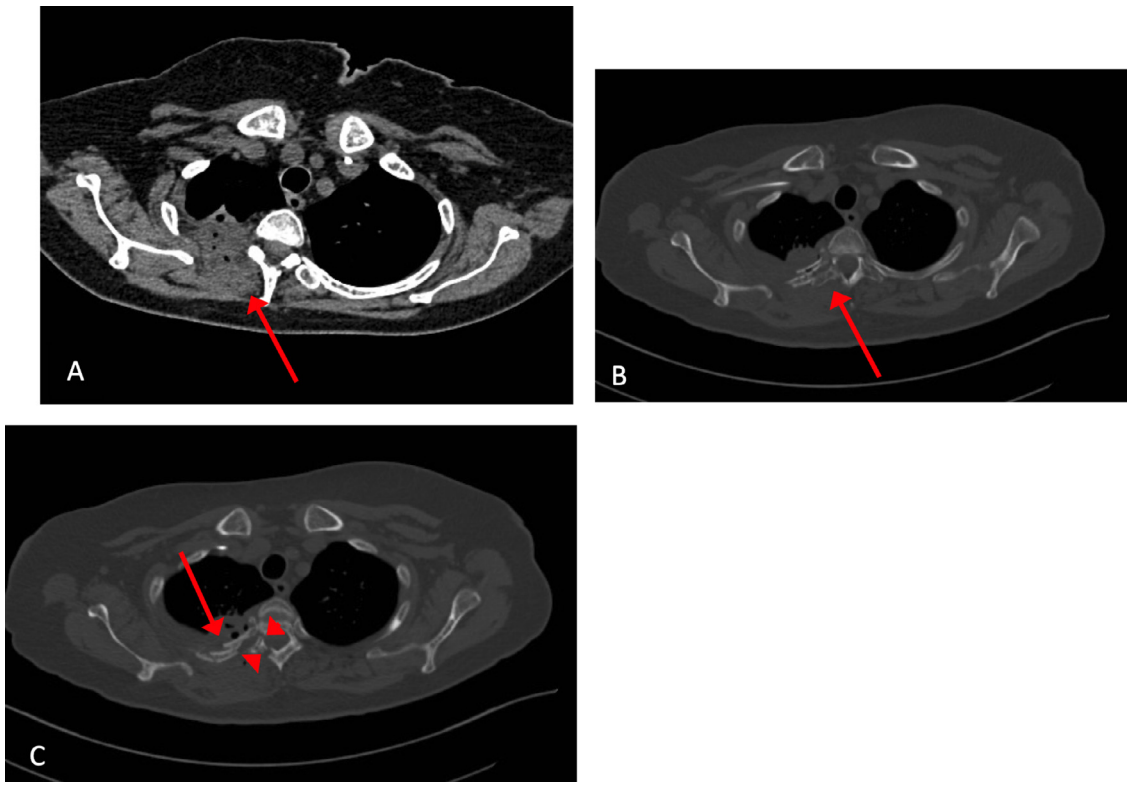

Fig. 5 – Non-enhanced HRCT of the chest: (A) Right apical cavitary lesion with involvement of the right paraspinal muscles (arrow). Multiple air foci are present within the collection and in the paraspinal muscles involvement. (B) Adjacent osseous rarefaction is noted at the posterior aspect of the right 3rd and 4th ribs as well as right transverse processes of the same levels. (C) Displaced fractures of the right 3rd and 4th ribs are present (arrow), with involvement of the T3 right transverse process, lamina, and pedicle (arrowheads).

symptomatically and the fistula closed spontaneously as repeated CT head showed a gradual decrease in the pneumocephalus.

Discussion

Pleural-subarachnoid fistula is one of the rare complications of thoracic and spine surgery, especially when in the paravertebral gutter region or as a result of blunt or penetrating trauma [2,3]. Milloy et al in 1959 are the first ones who report a pleural-subarachnoid fistula [1]. It occurs when attract formed between pleural and meningeal tears [4]. In the literature, there are less than 60 cases of Pleural-subarachnoid fistula. Almost all of the cases are post-surgical complication, post-trauma, or rarely spontaneously such as a result of rupture of a lateral thoracic meningocele into the pleural cavity or secondary to cystic degeneration of a neurofibroma [5]. only one case is found as a complication of superior sulcus tumor post chemoradiotherapy necrosis and did not undergo a thoracotomy or any other operative intervention [6]. None of which reported secondary to atypical fungal infection of the lung as found in our case, which destruct the pleura and the adjacent bone to invade the meninges through the costovertebral angle. However, the upper lobe of the lung with chest wall invasion into the region of the costovertebral angle is the common anatomic location of the cases of en bloc lung resection and presented with Pleural-subarachnoid fistula which represents less than 1% [3].

Pleural-subarachnoid fistula cause hydrostatic pressure differences between the pleural cavity and the CSF spaces, Thus, during inspiration the low intrathoracic pressure acts as a cavum and allows for the CSF to accumulates in the pleura, results in pleural effusion, and vice versa during the expiration where the air will be pushed to the CSF space resulting in pneumocephalous [4,7,8]. Nevertheless, the clinical manifestation is most commonly pleural effusion and pneumothorax respectively. patients could present with hemopneumothorax or widening of the mediastinum [1]. Accordingly, the Patient could present with a variety of symptoms passed on the clinical manifestation, including shortness of breath, chest pain, nausea, vomiting, fever, headache, altered mental status, the declining level of consciousness, or rarely neurological deficit [3,2,7]. Upright position can worsen the symptoms as it leads to increase pneumocephalous [7]. According to the literature, the time of presentation might vary from days to months following fistula formation [2].

Diagnosis of Pleural-subarachnoid fistula can be challenging, a combination of clinical, radiological, and laboratory data would help in reaching the diagnosis and preventing meningitis and progressive myelopathy [4]. Chest radiograph and chest

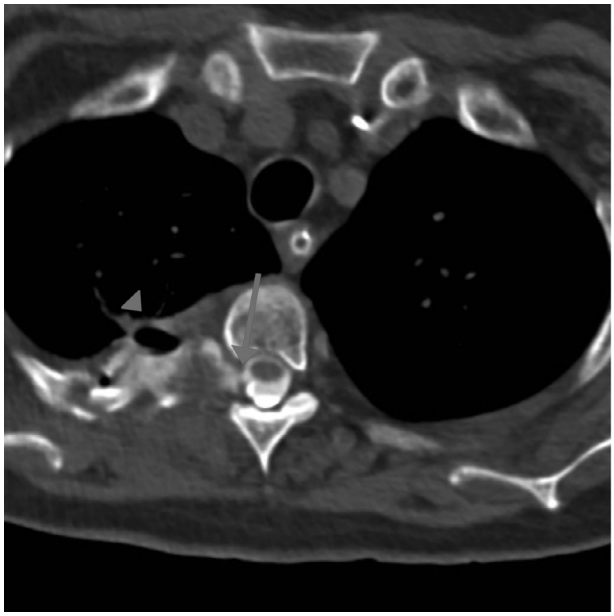

Fig. 6 – Thoracic spine CT myelogram: at the level of T3-T4, there is contrast extravasation noted tracking from the spinal canal through a fistulous tract (arrow) along the right neural foramina into the right pleural cavity (arrowhead) likely secondary to a dural tear.

CT will recognize the pleural effusion, but they are nonspecific, while pleural effusion analysis using β2 transferrin is a highly sensitive and specific test for detecting the presence of CSF [2,1,9]. However, this is not enough alone, since the exact location of the fistula needs further evaluation. Different diagnostic modalities can be used and each carries a different specificity and sensitivity. For example, radioisotope myelography has a 100% positive rate, but on the other hand, it carries high radiation damage to the body comparing to other modalities [1]. CT myelography as obtained in our case is superior to the radioisotope myelography in terms of detailed anatomic location and can aid surgical planning. However, it might have false-negative results [2,3]. These two modalities are the most commonly used and are the best confirmatory tests [3]. Other less invasive although less sensitive modalities include MRI imaging [4,7].

There is no specific treatment or management of Pleural-subarachnoid fistula, as it depends on many factors such as the clinical background of the patient, the location and size of the fistula as well as its progressive course [10,7]. Both surgical repair and conservative management have been reported in the literature, including for example; bed rest, chest tube insertion, external lumbar drainage, positive pressure ventilation, epidural blood patch, or subcutaneous biological glue injection [1,2,7]. Interestingly, in our case, spontaneous closure of the fistula took place which is the least expected pathway as reported in the literature [4], and follow-up CT scans of the brain showed a subsequent gradual decrease in the degree of pneumocephalus.

Patient Consent

Written informed consent for publication was obtained from the patient

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