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Case Report

A Case of Broncho-Paraspinal Fistula Induced by Metallic Devices : Delayed Complication of Thoracic Spinal Surgery

Seungnam Son, M.D.,[1] Dong-Ho Kang, M.D.,[2,4] Dae Seub Choi, M.D., Ph.D.,[3,4] Nack-Cheon Choi, M.D., Ph.D.[1,4]

Departments of Neurology,[1] Neurosurgery,[2] Radiology,[3] Gyeongsang Institute of Health Science,[4] Gyeongsang National University School of Medicine, Jinju, Korea

We present a case report of a 45-year-old woman with spontaneous pneumocephalus accompanied by pneumorrhachis of the thoracic spine, which is a very rare condition generally associated with trauma and thoracic or spinal surgery. The patient had undergone an operation about 10 years earlier to treat a giant cell tumor of the thoracic spine. During the operation, a metallic device was installed, which destroyed the bronchus and caused the formation of a broncho-paraspinal fistula. This is the suspected cause of her pneumocephalus and pneumorrhachis. To our knowledge, this is a very rare case of pneumocephalus accompanied by pneumorrhachis induced by metallic device, and when considering the length of time after surgery these complications presented are also exceptional.

Key Words : Giant cell tumor · Pneumocephalus · Pneumorrhachis · Spinal surgery.

INTRODUCTION

Pneumocephalus, the presence of air in the intracranial cavity, accompanied by pneumorrhachis, the presence of air within the spinal canal, is very rare condition generally associated with trauma[1)] and thoracic or spinal surgery[3,4)]. We experienced a patient with spontaneous pneumocephalus accompanied by pneumorrhachis of the thoracic spine. The patient had a history of spinal surgery to treat a giant cell tumor of thoracic spine about 10 years earlier. Consequent workup revealed that a broncho-paraspinal fistula was induced by a metallic device installed during the prior operation, and that this in turn resulted in pneumocephalus accompanied by pneumorrhachis. To our knowledge, spontaneous pneumocephalus and pneumorrhachis induced by metallic device are very rare and that presenting a long time after thoracic spinal surgery also rarely occurs.

CASE REPORT

A 45-year-old woman with a history of thoracic spine surgery about 10 years earlier was transferred to our hospital because of a severe headache. About 13 years earlier, she visited our hospital due to back pain and was diagnosed with a compression fracture of the T6 vertebra but did not undergo further investigation at that time. Over the next few years, she visited another hospital and was diagnosed with a giant cell tumor of the thoracic spine. A mass removal and posterior fusion operation was performed twice and she also received radiation therapy. About 2 years from the second operation, she visited our hospital due to sudden paraparesis and was diagnosed with recurrence of the giant cell tumor. A corpectomy from the T5 to T7 vertebrae was performed and a mass was removed. To improve spinal instability, the vertebral body that was removed was replaced with a Harms titanium mesh cage and bone cement. The neurosurgeon also performed an anterior fusion from the T4 to T8 vertebrae using Kaneda screws and rods. After the operation, she had no neurologic sequelae.

About 4 days ago, she developed a high fever with headache and was admitted to the local clinic and diagnosed with an influenza infection. Despite the use of Tamiflu® (Roche, Basel, Switzerland), her fever and headache persisted. The attending clinician started empirical intravenous antibiotic therapy with ciprofloxacin (CJ Pharma, Seoul, Korea) and netilmicin (Kwang Dong Pharmaceutical, Seoul, Korea). After the use of these antibiotics, her fever improved but the headache persisted. A brain computed tomography (CT) scan was performed at the local

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• Address for reprints : Nack-Cheon Choi, M.D., Ph.D.
Department of Neurology, Gyeongsang National University School of Medicine, 79 Gangnam-ro, Jinju 660-702, Korea
Tel : +82-55-750-8077, Fax : +82-55-755-1709
E-mail : brevis1@naver.com

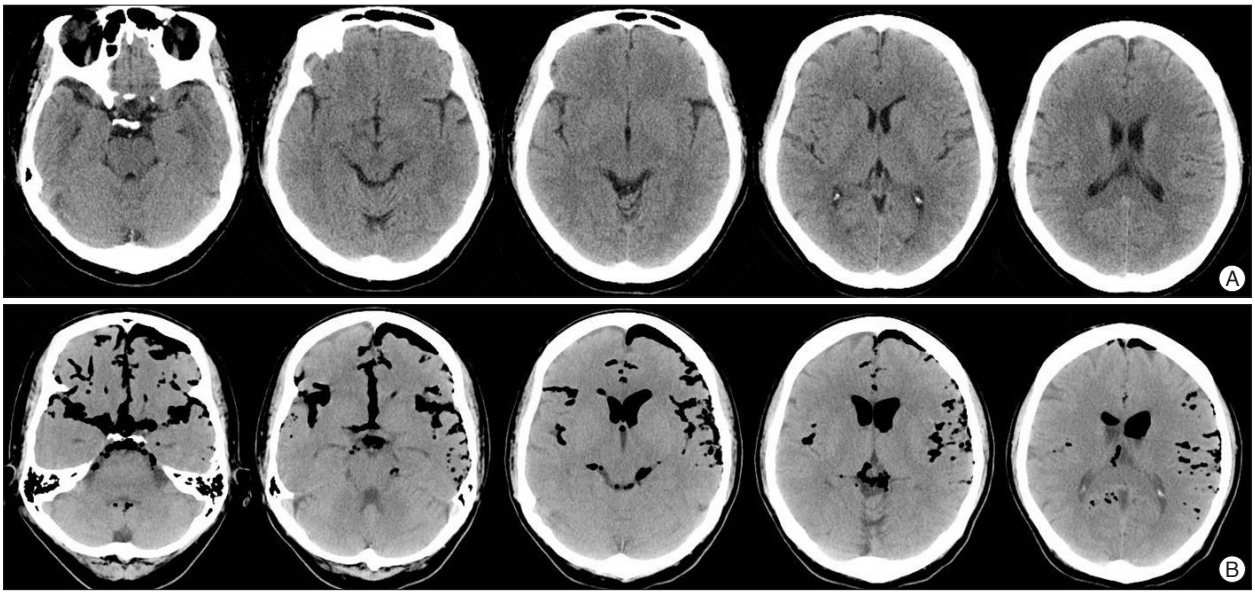

Fig. 1. Brain CT images of the patient, obtained at the local clinic the day before (A) and at our hospital on admission day (B). Newly appeared diffuse and severe pneumocephalus in the subarachnoid spaces and ventricles are shown on the (B).

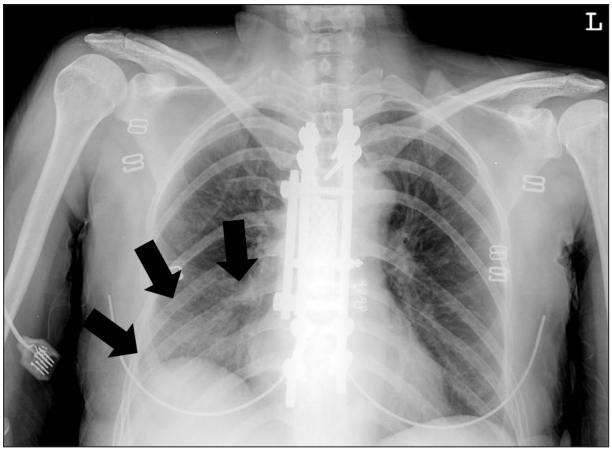

Fig. 2. Plain chest X-ray obtained on admission day. Mild haziness is shown on the right lower lung field (black arrows). Note the installed metallic devices during the prior operation on the thoracic spine.

clinic the day before and showed no abnormal findings (Fig. 1A). She was transferred for further evaluation and management of the persistent headache.

Her vital signs were stable and her body temperature was normal (36.5°C). During a neurological examination, the patient was alert, fully oriented, and had no signs of focal neurological deficits. The only suspicious symptom during the examination was neck stiffness. The laboratory results were unremarkable except increased ESR (76 mm/h) and CRP (31 mg/L). A plain chest X-ray showed mild haziness on the right lower lung field (Fig. 2), but the breathing sound of the patient was clear without rales or wheezing. A brain CT scan was performed again at our hospital, which showed diffuse and severe pneumocephalus in the subarachnoid spaces and ventricles (Fig. 1B). The opening pressure of spinal tap was 75 mmH_2O, the white blood cell count of the cerebrospinal fluid (CSF) was 88/μL (63% PMN, 37% lymphocyte), the protein level was 76 mg/dL, and the glucose level was 46 mg/dL (serum : 130 mg/dL).

A magnetic resonance myelogram was performed. However, the metallic device installed during the previous operation distorted the image and we could not obtain any information. A CT myelogram was performed next. A leakage of radioactive dye was not observed on the CT myelogram, but an air pocket that connected to the right bronchus was seen on the right paraspinal area at the T6 level (Fig. 3). Also, an air bubble was present in the subarachnoid space and spinal canal (Fig. 3).

A bronchoscopy revealed a metallic plate on the superior segment and a metallic bar on the basal segment of the right lower lobe bronchus (Fig. 4). The formation of a fistula between the bronchus and the prior operation site within the thoracic spine was suspected. An operation to correct the problem was scheduled but the patient and guardian refused an operation at our hospital, and thus she was transferred for the operation.

DISCUSSION

Two points are of particular interest in this case. First, spontaneous pneumocephalus and pneumorrhachis associated with surgery itself are rare. Second, to our knowledge, this is the first report about the devices installed during the prior thoracic spinal operation that induced delayed complications such as pneumocephalus/pneumorrhachis.

After considering the CT finding and pleocytosis of the CSF at first, we suspected a basal skull fracture and/or a bacterial infection of the central nervous system (CNS). However, the patient denied any history of head trauma and the CT scan of the skull showed no abnormalities. The contents of the CSF showed evidence of a CNS infection. The typical course of a gas-forming bacterial CNS infection is usually accompanied by severe

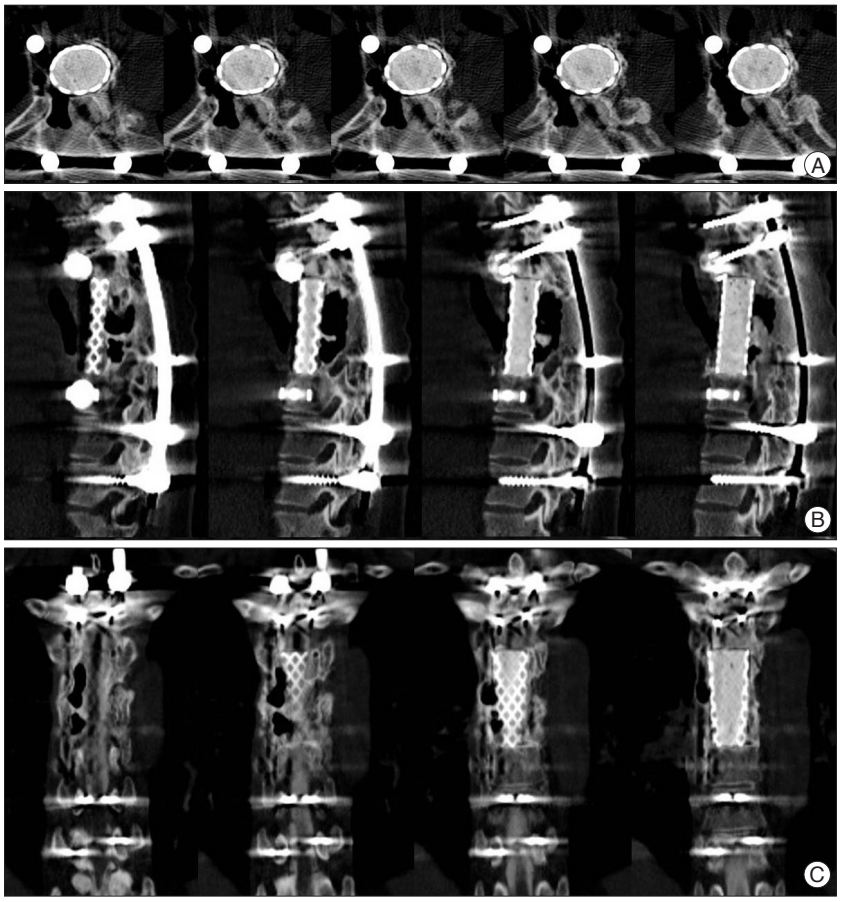

Fig. 3. CT myelography of the patient, axial (A), sagittal (B) and coronal (C) images. An air pocket that connected to the right bronchus is seen on the right paraspinal area at the T6 level. Also, an air bubble is present in the subarachnoid space and spinal canal.

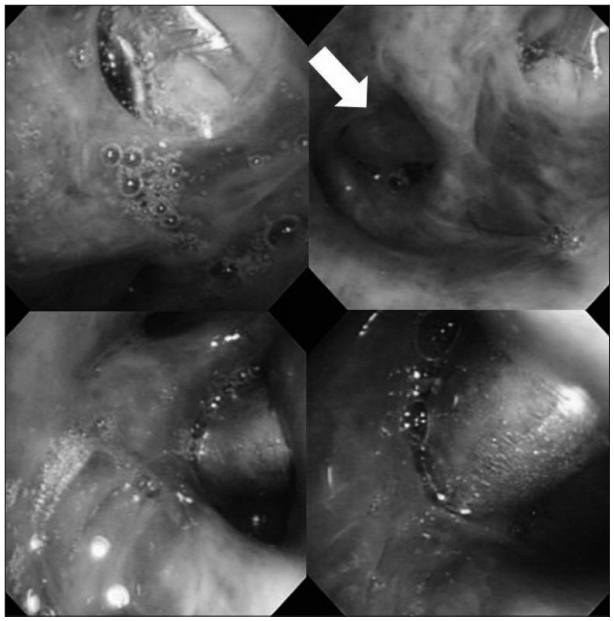

Fig. 4. Images taken during the bronchoscopy. A metallic plate on the superior segment and a metallic bar on the basal segment of the right lower lobe bronchus are shown. Note the granulation tissue due to chronic inflammation on the right-middle lobe of the bronchus (white arrow).

neurological deficits and high mortality[8], however the relatively benign course of her illness did not match the usual course of a gas-forming bacterial infection of the CNS. Nevertheless, we could not exclude the possibility of a gas-forming bacterial infection with confidence. Thus, we started empirical antibiotic therapy against the usual gas-forming bacteria with vancomycin (CJ Pharma, Seoul, Korea), ceftriaxone (Hanmi Pharm, Seoul, Korea), and metronidazole (JW Pharmaceutical, Seoul, Korea).

We believed that something else was likely to be the cause of her pneumocephalus. Therefore, we considered the well-known etiology of spontaneous pneumocephalus, which has several suggested mechanisms for its development[10,11]. One potential mechanism is that air enters the intracranial cavity through a defect in the skull (known as the ball valve mechanism). Another is that a continuous leakage of CSF results in air substituting for the CSF (known as the inverted pop bottle mechanism). Lastly, either the diffusion of anesthetic agents, such as nitrous oxide (N_2O), or a gas-forming bacterial infection of the CNS is believed to be causes of pneumocephalus. We also excluded the role of N_2O anesthesia because the patient had no recent history of an operation or anesthesia.

We noticed haziness on the right-lower lung field on the initial plain chest X-ray, although the patient had no respiratory symptoms. The patient complained of a high fever and headache and denied any respiratory symptoms, such as dyspnea, cough, sputum, and pleural chest pain. We thought the haziness on the X-ray was likely due to small amounts of pleural effusion. Because of this, we doubted the possibility of a fistula formation between the pleural space and the previous operation site. We searched PubMed and found some case reports of pneumocephalus/pneumorrhachis due to a CSF fistula that was associated with surgery. Various types of surgery were reported, but majority were lung resection[3,5,6] and the spinal surgery[4,7,9,11]. In almost all reported cases, the intervals between surgery and the development of pneumocephalus/pneumorrhachis were relatively short, from a few days[5,7,9,11] to a few months[3,4], but the few cases of several years after the surgery were also reported[6,10]. To detect a possible fistula, we tried to obtain a myelogram, first using magnetic resonance imaging (MRI) and then CT.

The mechanism of broncho-paraspinal fistula formation of this patient could not be determined definitively. On the post-

operative CT image obtained about 10 years earlier, the bronchus and the installed rod were situated conterminously. We thought the continuous compressive pressure of the rod to bronchus led to chronic inflammation of the bronchus, which led to the rupture of the bronchus and the formation of a fistula. During the bronchoscopy, a metallic foreign body was seen through the right-lower lobe of the bronchus, and granulation tissue due to chronic inflammation was also observed through the right-middle lobe of the bronchus. We did not know how broncho-paraspinal fistula of this patient reached the subarachnoid space. We thought the possible inflammation between Harms titanium mesh cage and meninges of the thoracic spine led to the extension of a fistula to the subarachnoid space.

CONCLUSION

Giant cell tumors represent approximately 5% of all bone tumors and over 20% of all benign bone neoplasms[2]. Although benign histologically, the tumors have an aggressive tendency to locally invade and recur[2]. At the time of surgery, an extensive surgery using various devices was the best choice due to the aggressive nature of the tumor, and the neurosurgeon could not anticipate future complications such as destruction of the bronchus and the formation of broncho-paraspinal fistula seen in this case. Because of the limited information available from the CT and MRI scans due to the metallic artifacts and the patient's refusal of surgery at our hospital, we will never know the accurate anatomical location of the possible bronco-paraspinal-subarachnoid fistula and the status of that fistula. We, however, believe that the diagnostic approach used in this patient is invaluable to report.

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