Spontaneous Pneumorrhachis and Transverse Myelitis Complicating Purulent Meningitis

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INTRODUCTION
Pneumorrhachis is defined as the presence of air collection in the spinal canal. It represents a rare entity that is mostly iatrogenic. The association of this entity with spontaneous pneumomediastinum without any pneumothorax is rarely reported in the literature. The spontaneous resorption is the usual evolution. The association to acute transverse myelitis is discussed by the authors. Therefore, the purpose of this work is to discuss possibilities and mechanisms of serious and grave multiple complications associated with purulent meningitis (pneumorrhachis). Based on the studied case, conclusion and recommendations that would animate the debate on the topic are also presented.

CASE REPORT
The patient is a 21-year-old male without significant medical history. He was admitted in the emergency department for cephalalgia, fever, and frequent easy vomiting lasting 1 week duration. Symptoms were associated to a puffiness of the face and dyspnea of level I according to Sadoul classification. At admission, the patient was conscious with a fever of 39°C. The blood pressure was 120/70 mmHg with a heart rate of 80 beats/min and a respiratory frequency of 24 cycles/min. The neurological examination objectified signs of meningeal irritation without any motor and sensory deficit. The pleuropulmonary examination found subcutaneous emphysema in the top part of the thorax and upper limbs extending to the cervical region and the face. Associated air effusion syndrome was not evidenced.

The patient is a 21-year-old male with pneumorrhachis associated to a spontaneous pneumomediastinum was admitted at the emergency department for bacterial meningitis. The antibiotherapy has marked the clinical profile by disappearance of the meningeal signs in the 48 h after admission. In contrast, the neurological symptoms were of marked aggravation by appearance of a tetraparesis with a respiratory distress syndrome having required artificial ventilation. The computed tomography (CT) scan showed a typical hypodensity corresponding to paramedullary air extending to several thoracic segments. The spinal magnetic resonance imaging (MRI) showed a high cervical medullary edema without signs of compression. The patient died within 15 days with a profile of vasoparalysis resistant to vasoactive drugs. Pneumomediastinum associated to pneumorrhachis and transverse myelitis complicating purulent meningitis is a rare entity. Although the usual evolution is favorable, the occurrence of serious complications is possible.

Key words: Pneumomediastinum without pneumothorax, Pneumorrhachis, Transverse myelitis

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neutrophils (80%). The glycorrhachia was 0.40 g/l (with concomitant glycemia of 1.20 g/l); total albumin level was 0.98 g/l, and the classical cerebrospinal fluid (CSF) gram’s stain and culture was negative. The immunochemistry study of lumbar puncture was in favor of meningitis diagnosis, which oriented to start the antibiotherapy. The routine biological and hematological studies included blood cells count and blood culture, ionogram, and sedimentation rate were found to be normal. The hepatic assessment was also without abnormalities. The diagnosis of bacterial meningitis was retained. The patient was treated using adequate antibiotherapy consisting of 2.0 g of ceftriaxone every 12 h. Ceftriaxone dose was maintained at the meningitis rate of 100 mg/kg/day.

The thorax X-ray showed a pneumomediastinum in the left paratracheal border along the left edge of the heart; the image also showed subcutaneous emphysema without associated pneumothorax [Figure 1]. Evermore, thorax CT scan confirmed all abnormalities described in the thorax X-ray and revealed the presence of air collection in the spinal canal between C7 and D6 levels [Figure 2]. The laryngoscopy, bronchoscopy, and digestive opacification did not show any abnormality in the respiratory-digestive tracts. These invasive gestures were achieved under general anesthesia without remarkable event.

The clinical evolution within 48 h was marked by the occurrence of apyrexia, coupled with disappearance of the subcutaneous emphysema and the meningeal steepness. However, the neurological status of the patient worsened gradually; a paraparesis occurred and was followed by a tetraparesis. The patient started with presenting neurovegetative disorders with hemodynamic instability which was in favor of central affection. The sepsis was discarded since the patient was afebrile. The patient experienced acute respiratory syndrome that required artificial ventilation. Afterward, a spinal magnetic resonance imaging (MRI) was performed and showed a cervical medullary edema extending from C2 to C6 without any sign of spinal cord compression. This result was consistent with a general clinical profile of acute transverse myelitis [Figure 3]. The hemodynamic support consisted initially of administrating vasoactive Drugs and dopamine then adrenaline since nor adrenaline was not available. The patient died within 15 days with a profile of vasoparalysis resistant to vasoactive drugs.

**DISCUSSION**

Initially described by Gordon and Hardman, in 1977,[8] pneumorrhachis is defined as the presence of air in the spinal canal, either in the intradural and/or extradural spaces. It is a very rare clinical entity and mostly asymptomatic; hence most probably underdiagnosed.[1]

Pneumorrhachis is commonly classified either spontaneous (nontraumatic), traumatic, or iatrogenic. The first type is
associated with degenerative, malignant, inflammatory, and infectious diseases,\(^9\) while the last variety is generally associated to epidural anesthesis, lumbar puncture, spinal surgery,\(^10\) or after a chest tube insertion.\(^{11}\)

The pathophysiology of pneumorrhachis coupled with a spontaneous pneumomediastinum is not completely understood. The most reported hypothesis in the literature consists of a mechanism involving an increased intrabronchial pressure that leads to alveolar rupture when enough gradient pressure is produced. Then, the air penetrates peribronchial and perivascular spaces to reach the mediastinum.\(^1,^{3,4,10,12-14}\) Afterwards, the air cumulated in the mediastinum separates the mediastinal pleura from the aorta and the parietal pleura from the rachis, and penetrates the epidural space through the conjugate foramina. Additional mechanisms might result from severe asthma attack, coughing, or other efforts while the glottis is closed. More rarely, spontaneous pneumorrhachis occur after grave vomiting efforts that our patient expressed; he manifested vomiting secondary to purulent meningitis.

The association of spontaneous pneumorrhachis with a pneumomediastinum is quite rare and restricted to very few cases in the literature.\(^{1,10}\) To the best of our knowledge, the association of spontaneous pneumorrhachis with pneumomediastinum and transverse myelitis originating of purulent meningitis was never described in the literature. In our patient, we believe that the occurrence of a tetraparesis is not secondary to a spinal cord compression caused by the pneumorrhachis itself, since the level of the intramedullary hypersignal in the MRI images and the clinical profile are supporting the diagnosis of a transverse myelitis associated to pneumorrhachis. This association was never described in earlier literature.

Three possible explanations of this association might be considered. Firstly, both pneumorrhachis and transverse myelitis present are distinct complications of purulent meningitis evolving independently: Pneumorrhachis was caused by alveolar rupture induced by thoracic hyper pressure provoked on its turn by emesis, while transverse myelitis is an independent complication of meningitis related to infectious and/or inflammatory disorders. Secondly, pneumorrhachis and transverse myelitis have been closely laid: The pneumorrhachis has facilitated and/or anticipated the occurrence of spinal cord edema through inflammatory mechanisms or by compromising the venous drainage of the epidural veins. Finally, both pneumorrhachis and transverse myelitis have been probably caused by gas-forming germs. Nevertheless, this last hypothesis might be excluded considering the important

size of the pneumomediastinum and the absence of typical signs of infectious mediastinitis.

The diagnosis of spontaneous pneumomediastinum has to be retained after achieving the complementary investigations. These consist of endoscopy exploration and digestive opacification excluding any esophageal rupture, tracheal branches perforation, or laryngeal fissure.\(^{12}\) The CT scan remains the diagnostic tool of choice.\(^{13}\) However, X-ray radiography is helpful for initial exploration allowing the early detection of pneumorrhachis.\(^{14}\)

Most reports considered the benign course of spontaneous pneumorrhachis which is usually reabsorbed spontaneously and completely without causing any symptoms, and does not recur.\(^{11}\) Indeed, Yoshida et al. reported three observations of compressive pneumorrhachis complicated by lumbar radiculopathy, which were resolved spontaneously.\(^{7}\) Generally, no treatment is recommended in pneumorrhachis secondary to a spontaneous uncomplicated pneumomediastinum. However, in cases of secondary pneumorrhachis, prophylactic antibioticotherapy is recommended to prevent meningitis.\(^1\)

In case of symptomatic pneumorrhachis, several authors discussed the interest of a corticosteroid therapy.\(^8\) Thus, Raynor and Saint-Louis described a case of radicular compression secondary to a pneumorrhachis that occurred 10 days after a lumbar disk surgery; in this case, the evolution was favorable using corticosteroid therapy.\(^9\) Exceptionally, spinal surgery is required for releasing the spinal cord from the air bubbles.\(^{2,11}\)

To date, there are no guidelines for the treatment of this clinical entity due to its extreme scarcity. In general, pneumorrhachis resolves spontaneously once the underlying cause is treated. In addition, the control of infection is more important for the prognosis of the patient.\(^{16}\)

**CONCLUSIONS**

Pneumorrhachis is a rare clinical entity characterized by a favorable outcome. It can be assorted as iatrogenic, traumatic, and spontaneous.

The association of spontaneous pneumorrhachis with pneumomediastinum and transverse myelitis was never reported in the literature. The prognosis is mostly fatal. Therefore, particular care must be undertaken when dealing with patients of such pathological profile. Physiopathology of this entity might be multifactorial and difficult to clarify.
Further reports might better lighten the mechanisms underlying this pathological association as well as the optimal treatment.

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