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## Case Report

# Lumbar puncture as a cause of tension pneumocephalus, pneumorrhachis, and sacral meningocele infection leading to death: An extremely rare case report <sup>☆,☆☆</sup>

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## ABSTRACT

Although it was first described over 100 years ago, lumbar puncture is still an important diagnostic tool for a variety of infectious and noninfectious neurologic conditions. With the widespread use of this common and relatively safe performed medical procedure, minor and major complications can occur even when standard infection control measures and good techniques are used, including post lumbar puncture headaches, infection, bleeding, cerebral herniation, radicular pain, and even pneumocephalus in extremely rare cases. We describe a previously unreported complication of lumbar puncture performed for the diagnosis of meningitis in a 33-year-old woman with no medical history causing pneumorrhachis, tension pneumocephalus, and sacral meningocele infection leading to death. Lumbar puncture is a simple diagnostic procedure with few complications, but if the technique is incorrectly performed, or if it is accompanied by occult congenital malformations such as sacral anterior meningocele in our case, the consequences can be fatal.

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Abbreviations: ASM, anterior sacral meningocele; WBC, white blood count; PR, pulse rate; RR, respiratory rate; *S pneumoniae*, *Streptococcus pneumoniae*; CT, computed tomography.

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## Introduction

Lumbar puncture is a relatively safe procedure that can have at times minor to major complications including frequency: backache (25%), headache (22%), and severe radicular pain (15%) [1].

Pneumocephalus and pneumorrhachis whose main cause is trauma can occur rarely after iatrogenic procedures such as accidental dural puncture, during lumbar epidural anesthesia, and following epidural steroid injections [2].

Anterior sacral meningocele is a congenital defect within the spectrum of spinal dysraphism. It is defined by a herniation of the dural sac through a bony defect of part of the anterior sacral wall, depending on its content: It is referred to as meningocele, myelocele, or meningomyelocele [3].

The literature search revealed only a very few reported cases of pneumocephalus and pneumorrhachis after lumbar puncture [4] and no case of association with anterior sacral meningocele (ASM).

Our case is the first to describe an anterior sacral meningocele infection with tension pneumocephalus following a lumbar puncture which has been associated with multiple unsuccessful attempts at obtaining CSF in a 33-year-old female with no previous history.

## Case presentation

A 33-year-old female patient, with chronic constipation history, without other pathological medical or surgical history, was admitted to the emergency department for management of suspected meningitis with symptoms starting 1 week ago,

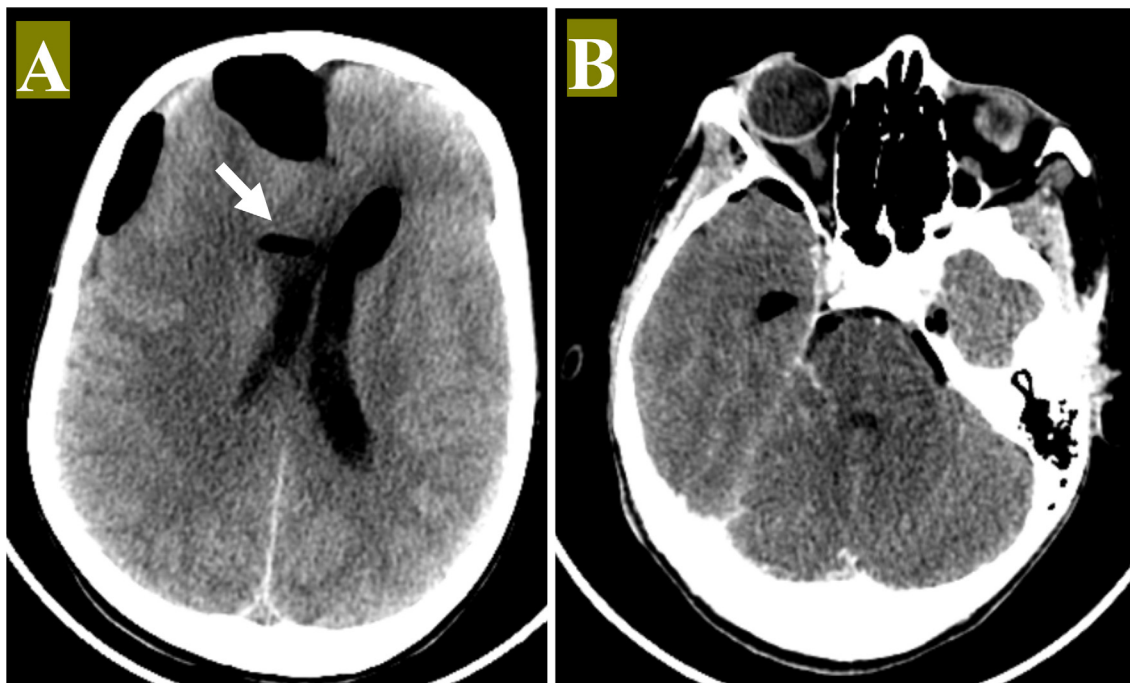
she was first admitted to a private clinic where several attempts of lumbar puncture were realized without success then she was referred to our structure for further care.

Physical examination upon admission revealed a confused patient with a GCS of 14. Hemodynamic and respiratory values were normal, with 115/75 mmHg of blood pressure, pulse rate (PR) of 69, respiratory rate (RR) of 15, and persistent fever at 38.9° (Oral). The neurological examination objectives a persistent meningeal stiffness, segmental muscular strength preserved in the upper limbs and rated at 2/5 in the lower limbs, superficial and deep sensitivity preserved with osteotendinous reflexes abolished in the lower limbs.

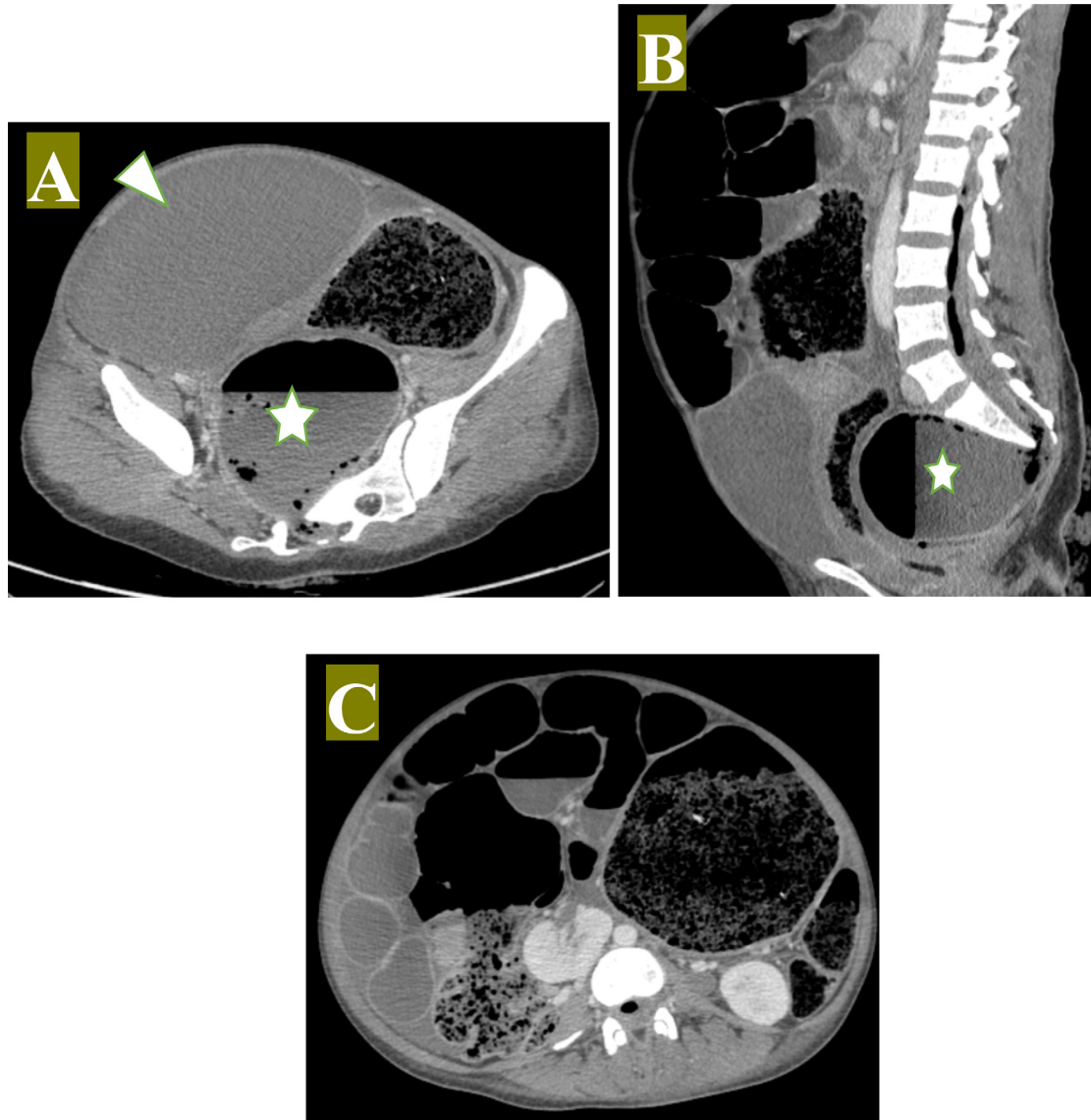
Biologically, the complete blood count cell showed a WBC of  $16 \times 10^9/L$  [Reference range:  $4.5-11.00 \times 10^9/L$ ], a hemoglobin level of 11.9 g/dL [Reference range: 10-15.5 g/dl], and a platelet count of  $185 \times 10^9 /L$  [Reference range:  $150-400 \times 10^9 /L$ ]. C-reactive protein (CRP) was elevated to 350 mg/L [Reference range < 4]. Urea, electrolytes, and creatinine were in normal values. The diagnosis of meningitis was made based on CSF analysis (a lumbar puncture was performed by the anesthesiologist-reanimator at admission), which revealed an elevated protein and white blood cell count, and low glucose level, the culture was positive for *S. Pneumonia*.

A contrast-enhanced brain CT scan was performed and revealed a significant supratentorial and infratentorial pneumocephalus with a mass effect on the cortical sulci responsible for a tonsillar herniation through the foramen magnum (Fig. 1).

The patient was hospitalized. Treatment with 3rd generation cephalosporin and osmotic therapy was initiated. 3 days later, she developed severe diffuse abdominal pain with the inability to have a bowel movement or pass gas. Contrast-enhanced abdominal CT scan was performed, revealing the presence of a pre-sacral formation with hydro-aerial content



**Fig. 1** – Brain CT scan showing the presence of pneumocephalus in the supratentorial (A) and infratentorial compartments (B). Note the mass effect on the right lateral ventricle which is collapsed (white arrow).



**Fig. 2 – Abdominal CE CT scan showing: (A) The defect in the anterior wall of the sacrum and the anterior sacral meningocele (asterisk), arrow-head: incidental ovarian cyst. (B) This formation fuses posteriorly through the right neural foramen of S3 with an intraductal extension and pneumorrhachis. (C) Enormous distention of the transverse colon in relation to large bowel mechanical occlusion.**

realizing an air-liquid level, with a contrast-enhanced wall, and measuring about  $116 \times 110 \times 105$  mm (transverse diameter, anteroposterior diameter, and height). This formation fuses posteriorly through the right neural foramen of S3 with extension into the spinal canal extending to the level of D5, and individualization of bony defect of the rest of the right sacral wing.

The CT also revealed an important bowel and colonic distension containing hydroaeric levels and measuring about 13 cm in maximum diameter at the level of the sigmoid colon upstream of a recto-sigmoid fecaloma, with this huge distention the risk of colonic perforation is to be expected.

We concluded to a presacral collection with intra-canal extension with sacral bone defect evoking an infected anterior sacral meningocele resulting in sacral meningocele abscess (Fig. 2).

A follow-up brain CT scan was also performed revealing resolving of preceding pneumocephalus, and the apparition of bilateral subdural hygroma relating to brain hypotension (Fig. 3).

On day 7 of her admission, the patient presented a clinical and biological deterioration with a GCS of 9, a WCC of 23.000, and a CRP of 490 complicated by a refractory septic shock leading to death.

## Discussion

Lumbar puncture is an important diagnostic tool for a variety of infectious and non-infectious neurologic conditions. It consists of the collection of cerebrospinal fluid (CSF) from the



**Fig. 3 – Brain CT scan objectiving bilateral frontal hygroma.**

lumbar subarachnoid space for diagnostic and/or therapeutic purposes.

The number of indications for lumbar puncture has decreased with the advent of new neuroimaging procedures including computed tomography (CT) scans and magnetic resonance imaging (MRI), but urgent LP is still indicated in the diagnosis of Suspected central nervous system infection including meningitis [5].

With the widespread use of this common and relatively safe performed medical procedure, minor and major complications can occur even when standard infection control measures and good techniques are used, including post lumbar puncture headaches, Infection, Bleeding, Cerebral herniation, radicular pain or numbness, Late onset of epidermoid tumors and pneumocephalus in extremely rare cases,

Pneumocephalus refers to the existence of air in the cranial cavity. Tension pneumocephalus is applied to intracranial air causing mass effects and abnormal neurological signs. Head trauma with breach or tearing of the dura is the most frequent cause of pneumocephalus, followed by infections tumors eroding the base of the skull, and post-surgical interventions. Pneumocephalus has also been reported following lumbar punctures and lumbar epidural injections and, rarely, after lumbar spine surgery, usually due to a dural tearing [6,7].

Two theories have been proposed to explain the pathogenesis of pneumocephalus:

**Dandy's ball valve theory:** Unidirectional movement of air from the external environment into the cranial cavity when the intracranial pressure is exceeded by the ex-

tracranial pressure frequently found in trauma or skull interventions.

**Horowitz and Lunsford's inverted soda bottle effect:** Excessive loss of cerebrospinal fluid due to physiological drainage during Valsalva or via a lumbar drain results in low intracranial pressure and entrapment of air in the vacuum created within the skull.

→ In our case, we suspect the inverted soda bottle effect with low intracranial pressure can be related to the anterior sacral meningocele accidentally discovered in our patient.

Anterior sacral meningocele is a congenital defect within the spectrum of spinal dysraphism. It is defined by a herniation of the dural sac through a bony defect of part of the anterior sacral wall. Herniation of the meninges through bony defects most often occurs as a result of dorsal deformities located in the posterior part of the lower back. Still, it can also develop along the thoracic or cervical spine, the anterior surface of the sacrum, or through facial bone defects or the skull vault. The sac is made of an external dural membrane and an internal arachnoid membrane and contains cerebrospinal fluid. Rarely, it may also contain neural material, and is then called myelomeningocele [8].

Anterior sacral meningocele is uncommon, and its diverse presentations can bring patients to the attention of a large group of specialists. The most frequent symptoms are lower back and Pelvic pain, constipation, dyspareunia, and urinary incontinence. Such symptoms are due to pressure exerted by the anterior sacral meningocele on the sacral nerve roots, rectum, bladder, or genitals. Two other characteristics that may indicate anterior sacral meningocele are Headaches and Meningitis. Intermittent headaches are due to alternating intracranial hyper and hypotension; intracranial hypertension is due to emptying of the anterior sacral meningocele into the subarachnoid space during an increased intra-abdominal pressure while the intracranial hypotension is secondary to the refilling of the ASM.

Anterior sacral meningocele-associated meningitis can be spontaneous or iatrogenic and is frequently polymicrobial and recurrent [9–11].

## Conclusion

Multiple complications can occur after lumbar puncture, of which pneumocephalus and pneumorrhachis are among the rarer ones. Our patient has the particularity of having an occult meningocele that fuses through the medullary canal. Pneumocephalus, in this case, can be explained according to the theory of inverted soda bottle.

Steps that can be taken that might prevent this from occurring include performing the lumbar puncture in the lateral

position and avoiding iterative lumbar punctures if it is hardly feasible.

## Patient consent

I, the author of the article: "Lumbar puncture as a cause of tension pneumocephalus, pneumorrhachis, and sacral meningocele infection leading to death: An extremely rare case report," approve that the patient gives her consent for information to be published in *Radiology Case Reports*.

## Availability of data and materials

The data sets are generated on the data system of the CHU Hassan II of Fes, including the biological data and the interventional report.

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