CASE REPORT

PNEUMOENCEPHALOMENINGITIS SECONDARY TO INFECTED LUMBAR ARTHRODESIS WITH A FISTULA : A CASE REPORT

E. GUERADO, D. PEÑA

Pneumocephalus associated with spinal problems is very rare. Association with encephalomeningitis secondary to a fistula after an infected elective lumbar spine fusion has not been previously reported. The authors report a case in which the clinical onset of pneumoencephalomeningitis occurred after an airplane flight. CT-scan and lumbar puncture were used to make diagnosis ; the treatment was based on parenteral antibiotics. The symptoms and signs of infection and neurological deficit resolved but the fistula remained. Diagnosis in such cases must be based upon CT-scan and lumbar puncture. Treatment should consist of systemic antibiotic therapy. Surgical management of infection and fistula is desirable, should the status of the patient allow such a treatment. In any case, as airplane flights in such cases may predispose to pneumocephalus, patients with an infected CSF fistula should avoid airplane flights until the problem is solved.

Keywords : pneumocephalus ; infection ; spinal fusion. **Mots-clés** : pneumoencéphale ; infection ; arthrodèse vertébrale.

INTRODUCTION

Pneumocephalus is defined by the presence of intracranial air. It usually appears secondary to fracture or surgery on the base of the skull, thoracotomy, tumor, infection or encephalocele (1). Pneumocephalus associated with spinal problems is extremely rare and, when present, is usually related to thoracic spine fracture with subsequent pleural and dural damage (6). In this paper we present a case of encephalomeningitis with pneumocephalus secondary to a cerebrospinal fluid fistula after infection of an elective fusion of the lumbar spine. We have found no similar cases in the literature.

CASE REPORT

A 54-year-old female who came on vacation from another country in Europe, presented to the emergency unit of our hospital complaining of headache of two months' duration. She felt worse suddenly one week before consulting, during an airplane flight. Her medical history included a lumbar arthrodesis for spondylolisthesis performed three months before, which had developed an infection and, subsequently, a sacral fistula. She was on penicillin V and metronidazole.

On examination she presented fever, dehydration, neck stiffness and a sacral fistula. Blood tests revealed leukocytosis (14,400 leukocytes/mm³) and normocytic anemia (hemoglobin of 10 mg/dl).

Lumbar xray showed an L4-L5 spondylolisthesis with pedicule screw instrumentation. Osteolysis surrounding pedicule screws was evident; one of the screws had pulled out of the pedicle (fig. 1a, b).

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Fig. 1a. — AP view of *in situ* lumbosacral arthrodesis. Peripheral to the screws there is osteolysis. *b.* Lateral view of *in situ* lumbosacral arthrodesis. Peripheral osteolysis around the screws. One of the L4 screws is out of the pedicle.

CT-scan of the brain showed a diffuse pneumocephalus (fig. 2). Lumbar puncture was then carried out revealing purulent cerebrospinal fluid (CSF). Biochemical analysis of the CSF showed glucose 2 mg/dl, protein 207 mg/dl, and a cell count of 1140 leukocytes/mm3 with 98% neutrophils. CSF aerobic culture grew Staphylococcus aureus (resistant to penicillin and ciprofloxacin) and Streptococcus viridans. Culture of fistula effusion showed Proteus mirabilis and meticillin-resistant Staphylococcus aureus. Blood culture was negative. She was diagnosed with pneumocephalus and encephalomeningitis secondary to an infected lumbar arthrodesis with fistula. She was treated with specific parenteral antibiotics (vancomycine, ceftazidime and metronidazol). We also advised her to undergo surgical debridement and osteosynthesis material removal, but she declined any surgical procedure out of her country. After intravenous antibiotic treatment, she had no fever and her meningeal symptoms disappeared. She then returned to her country by airplane, although we advised her against doing so. She apparently returned safely and subsequently underwent removal of the hardware and closure of the fistula.

DISCUSSION

Encephalomeningeal contamination secondary to spinal infection, as well as the presence of gas within the spinal canal and skull is very rare, and the combination of both has not been reported



Fig. 2. — Brain CT-scan showing pneumocephalus

before. It is related to an anatomical lesion of the dura (5). Spinal gas, when appearing, is usually located in the vertebral disc, and its etiology is unknown. Apparently the motion of the lumbar spine induces a pressure gradient, so that the intradiscal gas moves into the spinal canal by a valve-pump mechanism.

Other pathogenic mechanisms have also been described to account for the presence of gas in the spinal canal following cervical trauma, penetrating injuries of the chest, and after skull fractures (1). Preexisting models of pneumocephalus formation indicate that air tends to rise to the top of the cranial vault when it enters the subarachnoid space, that negative CSF pressure can occur (the negative pressure model) and that fistulas are subject to tamponade depending on their anatomy (the ball-valve model). Although these factors are important in understanding pneumocephalus formation, three other factors are equally important (9): (1) CSF is contained in a compartment that is elastic, (2) air and CSF exchange across a fistula occurs in a discontinuous and not simultaneous fashion, and (3) major shifts in CSF pressure occur from CSF loss and changes in posture and intrathoracic pressure.

Normally CSF pressure is greater than air pressure, so that most fistulas lead to CSF extravasation. If CSF loss exceeds net CSF production, the fistula will cause CSF volume and pressure to drop until CSF and atmospheric pressure equilibrate. Once an equilibrium develops across a fistula any further drop in CSF pressure will lead to pneumocephalus formation. Conversely any transient increase in CSF pressure as provoked by straining or coughing causes transient extravasation of CSF. On relaxation an aliquot of air equal in volume to the lost CSF must enter the CSF compartment to reequilibrate the pressure (9). Intracranial tamponade is an important factor which may retard pneumocephalus formation; however fistulas in the spinal subarachnoid space have no natural mechanism of tamponade (9).

Hopefully, according to the findings with gas in conjunction with herniated discs, pneumocephalus secondary to spinal gas will be reabsorbed spontaneously, and any neurologic deficit will clear (7). However in case of an infected spinal arthrodesis with instrumentation, although gas may have disappeared, removal of hardware, debridement of the wound, and initiation of intravenous antibiotic therapy is mandatory (2), as symptoms are mainly related to infection rather than to pneumocephalus.

In the case presented here, we believe that a pressure mechanism has pumped contaminated gas and CSF to the brain provoking clinical symptoms. Indeed airplane flights in these cases are a factor that predisposes to pneumocephalus (9), and, in the case reported, it should be noted that the patient felt worse immediately after an airplane flight. As patients with fistulous spinal infection have a high risk of developing bacterial meningeal spread and, therefore, infectious encephalomeningitis, we strongly recommend that these patients avoid airplane flights until their problem has been solved.

Pneumoencephalomeningitis should be suspected after acute headache in patients with a history of infected spinal surgery. CT-scan together with lumbar puncture is the most effective procedure (3) and MRI has no advantage over this (4). Antibiotics must be administered intravenously. Even with intravenous administration, vancomycine and metronidazole must be administered very slowly, as it has some neurological effects. Intrathecal and intraventricular aminoglycoside therapy should be considered only for patients with Gram-negative aerobic bacillary meningitis if there is no response to systemic therapy alone. Complications of intrathecal antibiotic administration may be severe. Intrathecal administration of antibiotics carries a risk of local toxicity, subdural effusion and physical alteration of the medulla and is best avoided, as the minimal therapeutic effect does not weigh up against these toxic effects. Intrathecal metronidazole must not be used at all.

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SAMENVATTING

E. GUERADO, D. PEÑA. Pneumo-encephalomeningitis door geinfecteerde lumbale arthrodesis.

De schrijvers brengen een raar geval van pneumoencephalomeningitis met klinische aanvang in aansluiting met een vliegtuigreis. De oorspronkelijke infectieuze haard was een open fisuliserende lumbale fusie. De diagnosis werd gesteld bij middel van Ct scan en lumbale punctie. De behandeling bestond in het toedienen van i.v. antibiotica; heelkundig debridement wordt overwogen, zogauw de toestand van de patiënt dit toelaat.

De sterke luchtdrukwijzigingen tijdens vliegtuigvluchten predisponeren tot pneumo-encephalitis in de aanwezigheid van een cerebrospinale geinfecteerde fistel en moeten dus worden vermeden zolang de fistel bestaat.

RÉSUMÉ

E. GUERADO, D. PEÑA. Pneumo-encéphaloméningite secondaire à une arthrodèse vertébrale infectée et fistulisée.

L'association d'un pneumoencéphale avec une encéphaloméningite secondaire à une fistule après arthrodèse lombaire infectée n'avait pas été rapportée à ce jour. Les auteurs en présentent un cas où les signes cliniques de pneumo-encéphaloméningite sont apparus après un vol en avion. Le CT scan et la ponction lombaire ont permis de poser le diagnostic ; le traitement a consisté en une antibiothérapie parentérale ; il a entraîné la résolution des symptômes neurologiques et de l'infection, mais la fistule a persisté. Le diagnostic dans de tels cas doit être fait par CT scan et ponction lombaire. Le traitement repose sur l'administration d'antibiotiques ; le traitement chirurgical de l'infection et de la fistule s'impose si l'état clinique du malade le permet. Les malades qui présentent une fistule infectée de liquide céphalorachidien devraient éviter les vols en avion jusqu'à ce que le problème soit résolu.