epidural PNET. Ishii et al. reported a supratentorial meningeal PNET and postulated that it might have originated from peripheral sensory nerves to the dura.[9]

PNETs have distinct immunohistopathology. These tumors are a group of "small round-cell" tumors which includes the Ewing's sarcoma/PNET family group, neuroblastoma, rhabdomyosarcoma, and malignant lymphoma. They have similar morphology with small round cells that contain fine granules, an inconspicuous cytoplasm, and round nuclei. Differential diagnosis of these tumors by routine histological examination is difficult. The most useful immunohistochemical marker for diagnosis is monoclonal antibody CD99, which is a product of the MIC2 gene. Fluorescence *in situ* hybridization is another diagnostic tool used to detect the specific t(11;22)(q24;q12) translocation in nuclei of neoplastic cells.[2]

Poor outcomes from spinal PNETs have been reported previously. Although multimodality therapy for these tumors has been reported, less than 40% of patients survive 2 years after diagnosis, and approximately 10% of patients remained alive at 3 years.[7] It is well established that treatment of intracranial PNETs with surgical resection and adjuvant therapy with chemotherapy and craniospinal radiotherapy improves survival rates. However, spinal epidural PNETs that do not have cerebrospinal seeding may have a better prognosis. Therefore, whether the epidural group of this type of tumor needs adjuvant radiotherapy is controversial. Shalet et al. reported that children younger than 10 who received spinal radiotherapy had a subsequent skeletal disproportion.[10] In our patient, even though radiotherapy was not performed, the time to local recurrence was similar to that of other reported patients. However, among the reported patients, there was one who did not receive adjuvant therapy and experienced local recurrence of the tumor after 7 months.[3] We recommended radiotherapy to treat the mediastinal part of our patient's tumor; however, her family declined treatment. The mediastinal tumor was enlarged 1 year later and the patient will now undergo an operation and receive adjuvant radiotherapy. In order to prolong survival, further adjuvant chemo-radiotherapy is indicated if radical resection of the tumor cannot be achieved.

Optimal therapy of spinal epidural PNET is yet to be defined. We suggest radical resection of the tumor should play an important role in preventing local recurrence and is the mainstay of treatment. Adjuvant therapy with radiotherapy or chemo-radiotherapy may play an assisting role if radical resection cannot be performed.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.jocn.2009.05.018.

References

1. Dorfmuller G, Wurtz FG, Umschaden HW, et al. Intraspinal primitive neuroectodermal tumor: report of two cases and review of the literature. *Acta Neurochir* 1999;**141**:1169–75.
2. Izycka-Swieszewska E, Stefanowicz J, Debiec-Rychter M, et al. Peripheral primitive neuroectodermal tumor within the spinal epidural space. *Neuropathology* 2001;**21**:218–21.
3. Kumar R, Reddy SJ, Wani AA, et al. Primary spinal primitive neuroectodermal tumor: case series and review of the literature. *Pediatr Neurosurg* 2007;**43**:1–6.
4. Akai T, Iizuka H, Kadoya S, et al. Primitive neuroectodermal tumor in spinal epidural space–case report. *Neurol Med Chir (Tokyo)* 1998;**38**:508–11.
5. Feng JF, Liang YM, Bao YH, et al. Multiple primary primitive neuroectodermal tumours within the spinal epidural space with non-concurrent onset. *J Int Med Res* 2008;**36**:366–70.
6. Liu HM, Yang WC, Garcia RL, et al. Intraspinal primitive neuroectodermal tumor arising from sacral nerve root. *J Comput Tomogr* 1987;**11**:350–4.
7. Virani MJ, Jain S. Primary intraspinal primitive neuroectodermal tumor: a rare occurrence. *Neurol India* 2002;**50**:75–80.
8. Aydin MV, Sen O, Ozel S, et al. Primary primitive neuroectodermal tumor within the spinal epidural space: report of a case and review of the literature. *Neurol Res* 2004;**26**:774–7.
9. Ishii N, Hiraga H, Sawamura Y, et al. Alternative EWS-FLI1 fusion gene and MIC2 expression in peripheral and central primitive neuroectodermal tumors. *Neuropathology* 2001;**21**:40–4.
10. Shalet SM, Gibson B, Swindell R, et al. Effect of spinal irradiation on growth. *Arch Dis Child* 1987;**62**:461–4.

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An intradural–extramedullary gas-forming spinal abscess in a patient with diabetes mellitus

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ABSTRACT

Spinal infections are commonly reported to be located in the extradural or intramedullary spaces. Infection involving the intradural–extramedullary space are uncommon. We report a patient with uncontrolled diabetes mellitus and an infected foot ulcer who presented with a cervical cord abscess and intradural gas. Early diagnosis and aggressive treatment are necessary for a favourable outcome in gas-forming intradural spinal abscesses. To our knowledge, a gas-forming intradural spinal abscess has not been reported previously and we discuss the relevant literature.

1. Introduction

Spinal abscesses can be a major cause of morbidity. Most of these abscesses are secondary to a disc space infection, hematogenous

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spread of systemic infection trauma, iatrogenic procedures or surgery. Diabetes mellitus and chronic steroid use compromise the immune system and may act as predisposing factors in the formation of these abscesses. Although most spinal abscesses are extradural, they can also be intradural–extramedullary or intramedullary. Intradural-extramedullary abscesses are uncommon. Spinal abscesses are generally not associated with gas formation. Extradural gas-forming abscesses have been reported in the literature. We present a unique case of a patient with an intradural–extramedullary gas-forming abscess in the cervical spine. To our knowledge, this is the first such report.

2. Case report

A 61-year-old man, suffering from diabetes mellitus, presented with fever, an ulcer on the right foot, neck pain and progressive weakness of all four limbs. He had urinary retention and constipation. The patient was febrile and tachypneic.

Examination revealed spasticity in all four limbs with quadriparesis. Power was grade 2/5 in the upper limbs and 3/5 in the lower limbs. Local examination revealed an ulcer on the right foot which had slough and purulent discharge. The hematological investigations showed an elevated white blood cell count and raised blood glucose.

A CT scan of the spine showed air in the spinal canal behind the second cervical vertebral body (Fig. 1). MRI of the cervical spine showed an intradural abscess from C2 to C7 levels with intradural air (Figs. 2 and 3). There was cord edema and extensive soft tissue inflammation in the prevertebral region from C2 to C7. At surgery, cervical laminectomies (C2–C7) were performed. On opening the bulging and non-pulsatile dura, there was egress of pus, which was evacuated.

The cervical abscess was thoroughly drained and liberally irrigated with saline and betadine solution. Postoperatively the patient required ventilatory support and vasopressors for correction of hypotension due to septicemia. His blood glucose concentrations were grossly abnormal and required a high dose insulin infusion.

The aerobic culture of the pus, from the cervical abscess as well as the foot ulcer, yielded *Staphylococcus aureus* and *Escherichia coli*. The anaerobic culture of the pus was negative. Blood culture yielded similar organisms. The foot ulcer was aggressively treated with surgical debridement and daily dressings. Initially broad-spectrum antibiotics were administered and thereafter the antibiotics were administered according to the antibiotic sensitivity report. Despite the intensive medical management, the patient succumbed on the tenth postoperative day. Histopathology of the tissues showed an acute pyogenic abscess.

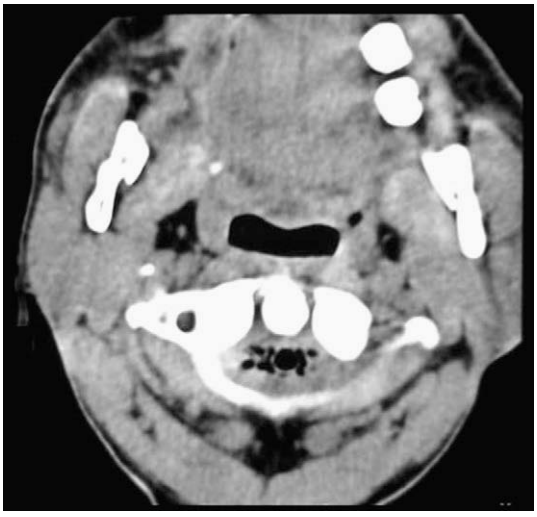

Fig. 1. Axial CT scan at the level of the odontoid process showing bubbles of air within the dura, anterior to the spinal cord.

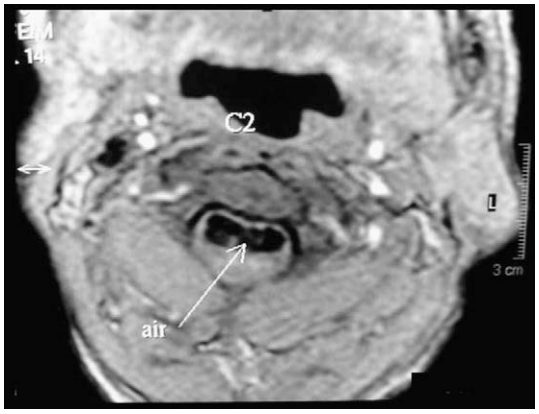

Fig. 2. Axial post-contrast T1-weighted MRI at the C2 vertebra showing intradural air.

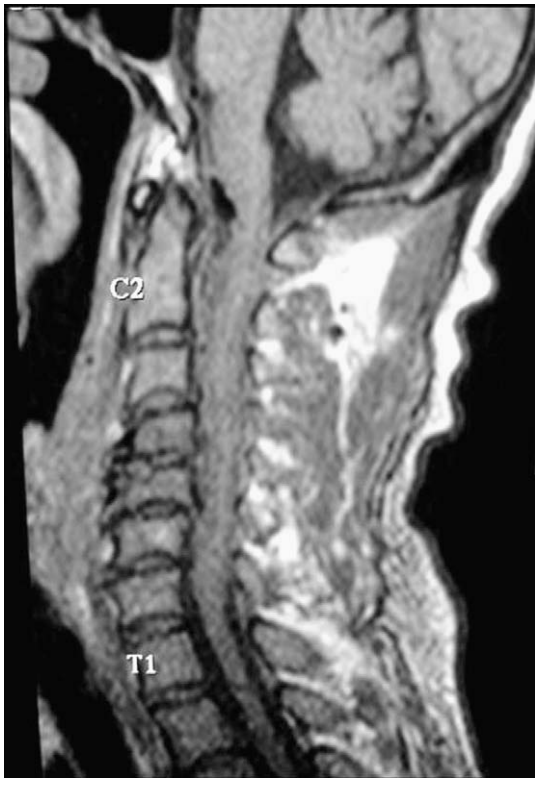

Fig. 3. Sagittal T1-weighted MRI showing the air within the abscess cavity anterior to the cord. There is an intradural abscess from C2–C7 with cord edema.

3. Discussion

Diabetes mellitus is disorder with an increased susceptibility for infection. Gas-forming infections are especially common in these patients and have been reported in the gall bladder, kidney, bladder, spleen and subcutaneous tissues.[1] The organisms usually involved are *Escherichia coli*, *Klebsiella pnuemoniae*, *Proteus mirabilis*, *Enterobacter aerogenes* and *Pseudomonas aeruginosa*.

Many spinal infections are preceded by infections at other sites in the body. Predisposing factors, such as diabetes mellitus, increase patient susceptibility to spinal infections.[2]

Necrotizing infections involving the spine due to foot ulcer in a patient with diabetes mellitus has been reported.[3] However, we there was no report of a patient with diabetes and a foot ulcer and an intradural pyogenic abscess with gas. Although extradural and intradural–intramedullary spinal abscesses are relatively common, intradural–extramedullary abscesses are exceptional.[4] Most intradural abscesses reported in the literature are hematogenous in origin.[5] Intradural abscesses secondary to iatrogenic causes, such as epidural injections, or predisposing conditions, such as dermal sinuses and spinal dysraphism, have been reported.[4–8]

The *S. aureus* and *E. coli* probably spread hematogenously from the foot ulcer and seeded in the spine intradurally, causing an abscess. The blood culture as well as the surgical tissue culture grew *S. aureus* and *E. coli*. *E. coli* was the likely cause of the gas in the spine as *S. aureus* does not commonly form gas. Other factors such as bacterial inoculum, host defences and altered host immunity, which occur in patients with diabetes mellitus, may have had a role.

Intraspinal gas is uncommon. The causes of gas in the spine are degenerative disc disease, trauma, infections, spinal tumors, intestinal necrosis, pneumothorax, and various diagnostic and surgical spinal procedures.[9] Intraspinal gas in infections can occur with both aerobic and anaerobic microorganisms. Carbon dioxide and water are the end products of aerobic metabolism. Hydrogen, nitrogen, hydrogen sulfide and methane are produced from the combination of aerobic and anaerobic bacterial infection. These gases, except carbon dioxide, accumulate in tissues because of their reduced solubility in water.

Intraspinal gas is generally considered benign and mostly asymptomatic in patients. In some patients, the gas can cause nerve root compression and sciatica. In our patient an intradural abscess with gas was detected on imaging when he presented with quadriparesis. The patient was managed surgically and the pus was evacuated.

Intradural spinal abscesses with gas can be fulminant, as was observed in our patient: early suspicion with rapid intervention is the key strategy. Thus, these patients should be managed aggressively with thorough debridement and lavage with the administration of culture-specific intravenous antibiotics. They also need intensive care management as these patients may develop septicemia.

References

1. Togninia G, Ferrozzia F, Bovab D, et al. Diabetes mellitus: CT findings of unusual complications related to the disease: a pictorial essay. *Clin Imaging* 2003;**27**:325–9.
2. Hadjipavlou AG, Mader JT, Necessary JT, et al. Hematogenous pyogenic spinal infections and their surgical management. *Spine* 2000;**25**:1668–79.
3. Spock CR, Roberto AM, Shah RV, et al. Necrotizing infection of the spine. *Spine* 2006;**31**:E342–4.
4. Kulkarni AG, Hee HT. Adjacent level discitis after anterior cervical discectomy and fusion (ACDF): a case report. *Eur Spine J* 2006;**15**:559–63.
5. Thome C, Krauss JK, Zevgaridis D, et al. Pyogenic abscess of the filum terminale: case report. *J Neurosurg* 2001;**95**:100–4.
6. Bartels RH, de Jong TR, Grotenhuis JA. Spinal subdural abscess: case report. *J Neurosurg* 1992;**76**:307–11.
7. Carmouche JJ, Molinari RW. Epidural abscess and discitis complicating instrumented posterior lumbar interbody fusion: a case report. *Spine* 2004;**29**:E542–6.
8. Fujiwara S, Morioka T, Ishibashi H, et al. Acute purulent discitis with epidural abscess of the cervical spine in an adult: case report. *Neurol Med Chir (Tokyo)* 1994;**34**:382–4.
9. Frank B, Dorr F, Penkert G, et al. An epidural spinal abscess with caudal symptoms as a complication of Crohn's disease. *Dtsch Med Wochenschr* 1991;**116**:1313–6.

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Oligodendroglioma presenting with intradural spinal metastases: an unusual cause of cauda equina syndrome

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ABSTRACT

We report a 37-year-old man with a primary intracranial oligodendroglioma presenting later with symptomatic multiple cerebrospinal fluid (CSF) intradural drop spinal metastases. This patient initially presented in 2006 with complex partial seizures. Initial histology demonstrated World Health Organization (WHO) grade 2 oligodendroglioma. The patient had further generalised seizures 7 months after initial tumour resection. MRI at that time confirmed tumour recurrence. The patient underwent a repeat craniotomy. Histology showed anaplastic transformation to a WHO grade 3 oligodendroglioma. About 30 months after his initial presentation, the patient developed a focal neurological deficit in the left leg with associated retention of urine. MRI of the neuraxis demonstrated widespread leptomeningeal metastatic drop deposits within the spinal canal. We discuss the mechanisms involved in tumour dissemination throughout the CSF. We also review the relevant literature regarding this phenomenon.

1. Case report

A 37 year-old man presented in January 2006 with a seizure that resulted in a road traffic accident. Subsequently, he had a number of further partial seizures. A brain MRI showed a left frontal lobe intrinsic lesion (Fig. 1). He underwent craniotomy and excision of the lesion in March 2006.

Histological examination showed a widely and diffusely infiltrating oligodendroglioma composed of small cells with uniform round nuclei and clear cytoplasm. There were only a few scattered mitoses of insufficient number to justify a high-grade diagnosis.

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