Rapid recovery after delayed myelopathy from electrical burns

Sir: We report a 34 year old man who was transferred to the Burns Unit, Concord Hospital, Sydney, Australia after accidentally flying his hang-glider into 22,000 volt power lines from which he had remained suspended for twenty minutes. On arrival he was noted to have sustained full thickness burns to the medial and ventral aspects of both forearms as well as the lateral aspect of the left thigh. Apart from weakness of right wrist extension, on initial examination power in all muscle groups was normal. He underwent extensive emergency fasciotomies of both arms and the left thigh. During his first few days in hospital he developed rhabdomyolysis (creatinine kinase 132,000 units/l), myoglobinuria and mild renal impairment (serum creatinine 0.15 mmol/l).

Two weeks after his admission, when his medical condition had improved, he complained of bilateral leg weakness and numbness of the left buttock. Neurological examination revealed signs of a myelopathy with bilateral lower limb spasticity and grade 4/5 weakness in a pyramidal distribution in both legs. There was also grade 4/5 to 4+/5 weakness of the intrinsic muscles of both hands and wrist extension and shoulder abduction of the right arm. The right supinator jerk was reduced. All lower limb reflexes were pathologically brisk and remaining reflexes were normal. Plantar responses were bilaterally extensor. Sensory examination revealed diminished sensation to pin and light touch over the median nerve territory of the right hand. There was no evidence of a sensory level and joint position sense was normal. Bowel and bladder function was unaffected. The patient's gait was markedly spastic and he was unable to walk unaided. Magnetic resonance imaging of the cervical and thoracic spinal cord was normal. Nerve conduction studies were not possible because of the burns to both forearms. However, somatosensory evoked potentials from both posterior tibial nerves revealed delayed peripheral as well as an absent cerebral (N/P37) response bilaterally (fig). In addition median nerve somatosensory evoked responses showed an absence at Erbs point, as well as low amplitude prolonged cervicomedullary P/N13 (17.4 ms; normal < 15.4 ms) and cerebral N19 (22.6 ms; normal < 21.9 ms) on the right side and on the left a prolonged low amplitude Erbs point potential (12.6 ms; normal 12 ms) with low amplitude cervicomedullary and cerebral responses of normal latency.

Most of the 40 cases of myelopathy follow-

ing electrical injury were described before World War II.¹ In a more recent series in one major US Army trauma centre myelopathy was found in only two of 111 patients presenting with electrical burns.² A delay between electrical injury and development of the myelopathy is characteristic. The reason for this is not known but it is speculated that as the electrical current passes through the spinal cord it damages its blood supply leading to a delayed vasculopathy.^{3,4} As with our patient, most cases have little sensory and no autonomic disturbance.^{2,3,5} Our patient's right median nerve sensory loss was probably related to carpal tunnel compression by oedema occurring after his burns, or to direct current injury. Somatosensory evoked potentials revealed upper and lower limb peripheral conduction abnormality as well as central conduction delay. This not only reflects damage to the spinal cord but also involvement of nerve roots and peripheral nerves lying in the path of the electrical current.¹ Recovery, if it occurs, has been said to be incomplete, not infrequently extending over years.¹ Several more recent reports attest to the fact that, as with our patient, a relatively rapid and almost complete recovery can occur from this unusual myelopathy.^{3,5}

Fig Posterior tibial somatosensory evoked potentials from right and left lower limbs demonstrating bilateral delay of the lumbar potential (LV1-IC) (marked 2) at 26.4 and 26.0 ms respectively (normal less than 24.0 ms). A cerebral response (CZ-FZ) was absent bilaterally.

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Intradural spinal lipoma with enlarged intervertebral foramen.

Sir: Spinal lipomas are benign tumours, usually divided into extradural and intradural types.¹ Spinal lipoma with spina bifida (lipomyelomeningocele) is distinguished from spinal lipoma without spina bifida.¹ Spinal lipoma without spina bifida is a rare benign tumour comprising about 1% of all spinal tumours.¹ Spinal hourglass tumours (dumb-bell tumour) occurs simultaneously both inside and outside the dura mater, or may develop an extravertebral extension through one or more intervertebral foramen,² and is a neurinoma in the majority of cases. Dumb-bell extension has rarely been reported in cases of lipomyelomeningocele and extradural spinal lipoma.³ To our knowledge, there are no case reports of intradural spinal lipoma in the absence of spina bifida, which extend through the intervertebral foramen and cause radiologically-proved widening of the intervertebral foramen. We report such a case of intradural spinal lipoma with an enlarged intervertebral foramen.

An 18 year old Japanese man was admitted to our hospital in August 1985, because of a gait disturbance. His foot had an equinovarus from childhood. He complained of back pain at the age of 15 years. In August 1984 he noticed weakness in the right and left leg. In December 1984 he visited another University Hospital and was diagnosed as having "familial spastic paraplegia".

Letters

The weakness in the lower extremities slowly progressed. He became aware of some loss of sensation in both legs in June 1985. On admission, examination revealed scoliosis and pes equinovarus. He did not have signs which suggested spina bifida, such as a dimple, abnormal pigmentation, or a subcutaneous lipoma. Neurological findings showed a spastic and ataxic gait, positive Romberg's sign, slight weakness in both legs, spasticity and hyperreflexia in the lower extremities and bilateral positive Babinski's signs, sensory disturbance in all modality below mid-thoracic level, and slight bladder dysfunction. Lumbar puncture demonstrated a block with xanthochromic fluid. The protein concentration in the cerebrospinal fluid was 26 g/l. A spine radiograph showed an enlargement of the right intervertebral foramina of C₇ and Th₁ (fig 1), which was confirmed by CT. A metrizamide myelogram by lumbar puncture disclosed a complete block at Th₁₁. Myelography by high cervical puncture showed a complete block at the lower margin of C₄. T₁-weighted magnetic resonance imaging (MRI) revealed a high intensity signal area in the cervical and thoracic segment of the spinal cord (fig 2).

Thoracic spine tomography demonstrated scalloping of the posterior wall of the vertebral body in Th₂, Th₃ and Th₄. Thoracic and lumbar spine radiography showed scoliosis but no evidence of spina bifida. He was diagnosed as having intradural spinal cord tumour with dumb-bell extension and a lipoma was suspected from the MRI findings. An operation was carried out for the purpose of removing the tumour on 22 October 1985. A skin incision was made from the spinous process of C₂ to that of Th₁₂. Subcutaneous tissue was well developed, but no subcutaneous lipoma was found. There was no defect of the spinous process nor any defect of the vertebral arch. Laminectomy from C₅ to Th₁₁ was carried out. There was no evidence of fibrous attachment to the posterior elements of the vertebral canal. The dura mater was considerably swollen. Small lipomatous tumours were found in the extradural space. There was no lipoma in the subdural space but incision of the arachnoid showed a lipoma, tightly adherent to the spinal cord. Though it was impossible to remove the tumour, it was considered that decompression was achieved. Histological studies confirmed that the tumour was a lipoma. After the operation, the patient's gait disturbance, spasticity and sensory disturbance improved. About two and a half years after the operation, in April 1988, the patient could walk without assistance and enjoyed daily life.

Enlargement of an intervertebral foramen

Fig 1 Spine radiograph, showing the enlargement of the right intervertebral foramen of C₇ and Th₁ (arrow).

Fig 2 T₁-weighted magnetic resonance imaging (MRI), showing a high intensity signal area in the cervical and thoracic segment of the spinal cord.

usually signifies a spinal hourglass neurinoma (dumb-bell tumour). The clinical features of our case, which were mainly composed of spasticity and posterior ataxia, differed from the clinical features of ordinary spinal neurinoma in the absence of radicular pain and radicular motor disturbance. It was difficult to decide from the findings of

myelography if our patient had an intradural or an extradural tumour, but MRI made an intradural spinal lipoma very likely. This was confirmed at operation which also showed small scattered extradural lipomatous tumours.

Spinal lipoma with dumb-bell extension is rare. Maier,¹ in 1962, reported a case of extradural and intrathoracic lipoma causing spinal cord compression in a 17 month old girl. The clinical and radiological features of the case were compatible with the diagnosis of neuroblastoma but she had a small subcutaneous lipoma in the left paraspinal area of the thorax. Rao, *et al*² reported a case of extradural spinal lipoma associated with intrathoracic lipoma in a 25 year old black female. She had multiple developmental anomalies and during childhood had undergone removal of a large lipoma over the upper back. The presence of an extradural spinal lipoma was verified by myelography and confirmed during operation. Quinn, *et al*³ in 1983, reported an extradural spinal lipoma with mediastinal extension. The case was a 12 year old boy with progressive weakness of the left arm over a period of four months. Myelography and laminectomy showed an extradural spinal tumour. The pathological diagnosis was spinal lipoma.

On reviewing the literature, we found there were no case reports of intradural spinal lipoma in the absence of spina bifida, which extends to the intervertebral foramen. If an hourglass tumour has atypical clinical features, the possibility of spinal lipomas should be considered, even in the absence of spina bifida.

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Coxsackie B5 papillitis

Sir: We describe the clinical and electrophysiological findings in a patient with papillitis associated with coxsackie B5 viral infection. Reports of proven coxsackie B papillitis are rare and in this case the electrodiagnostic studies provide interesting evidence of the site of the visual dysfunction.

A 32 year old white male developed symptoms of severe frontal headaches, night sweats, joint aches, photophobia and neck stiffness which resolved spontaneously. Two and a half weeks later the headaches and fever returned accompanied by pain on ocular movement. These symptoms persisted for a week and one week later he presented with progressive central blurring of vision in his left eye. There was nothing of note in his past medical or family history and there was no history of drug usage other than paracetamol for the headache.

On examination visual acuities were 6/5 right, and hand movements left. The left visual field showed a large central scotoma, the right an enlarged blind spot. A left relative afferent pupillary defect was present. There was flare with cells in his left anterior chamber and cells in the anterior vitreous of both eyes, but more in the left eye than the right. Both optic discs were considerably swollen with peripapillary nerve fibre layer haemorrhages; a macular star was present in the left eye (fig a, b). Ocular movements were full but there was discomfort on lateral gaze of each eye. Neurological examination revealed no focal abnormality and systemic examination was unremarkable. He was apyrexial.

Investigations showed normal urea and

electrolytes, normal liver function and plasma proteins, apart from an increase in α_2 globulin; TPHA/VDRL were negative, ESR 14 mm/h, Hb 14.9 g/l, WBC 12.3, platelets 223. A lumbar puncture showed a CSF pressure of 160 mm, 18 lymphocytes, protein 0.45 g/l (no increase in globulin) and glucose of 3.8 mmol/l (blood glucose 5.4 mmol/l). Serum angiotensin converting enzyme was normal as was a complement profile. Immune complexes were slightly raised by PEG precipitation assay at 6.0 mg/IgG/dl (normal 4.9). Chest radiograph and CT of the brain were normal. Fluorescein angiography confirmed bilateral swollen discs with adjacent peripapillary serous retinal detachment. EEG was normal.

Electrodiagnostic testing comprised EOG, ERG, pattern ERG (PERG) and both flash and pattern VEPs (FVEP, PVEP). PVEP and FVEP from the right eye fell within the normal range for a patient of this age. Left eye PVEP was grossly delayed and of markedly subnormal amplitude; FVEP also showed an amplitude reduction and latency increase compared with the right eye. Right eye PERG positive P50 component was of marginally subnormal amplitude, that from the left eye being of severely reduced amplitude with additional latency changes. ERGs from both eyes, although of somewhat low amplitude, showed no unequivocal abnormality. EOG findings suggested bilateral dysfunction in the region of the pigment epithelial/photoreceptor complex. The PERG abnormalities are consistent with bilateral macular photoreceptor involvement, severe on the left, mild on the right.

A Paul Bunnell test was negative as were serological studies for mumps S and V antigen, herpes simplex and zoster, cytomegalovirus, *Coxiella burnetii*, *Mycoplasma pneumoniae*, influenza A and B, HIV, *Chlamydia trachomatis* and psittaci, *Candida albicans*, and *Cryptococcus neoformans*. Coxsackie B virus specific IgM was detected on two separate occasions separated by eight days, diagnostic of active infection. The IgM was specific for coxsackie virus B5.

The patient was treated with oral prednisolone reducing from 80 mg per day over 10 weeks. Over the next two months he made an excellent recovery with visual acuities returning to right 6/5, left 6/9 and resolution of the afferent pupillary defect. The right visual field was full, but a large inferior arcuate scotoma persisted in the left eye. Both optic discs showed resolution of the oedema; there was a residual left macular star. The steroid therapy was tailed off and stopped and the patient failed to attend for further follow up.

FIG Fundus photographs taken 12 days after presentation show bilaterally swollen optic discs, worse in the left eye (A) than right (B). A macular star is present in each eye but more prominent in the left and in the left eye the retinal features are blurred by vitreous cellular infiltrate.

The combination of a previous systemic illness, aseptic meningitis and subsequent visual loss in the left eye associated with pain on ocular movement and signs of intraocular inflammation suggested a viral papillitis and this is supported by evidence of active infection with coxsackie B5.

The association of enterovirus infection with neurological complications is well established.^{1,2} Audry-Chaboud³ reported six cases of neuropapillitis linked to suspected coxsackie B virus infection although this was only confirmed in two cases by demonstration of specific IgM antibody titres to coxsackie B4 virus; (serological studies in the remaining cases were not conclusive). In both proven cases there was residual optic nerve damage. One, a 48 year old male, was left with bilateral optic atrophy and the other, a 25 year old female, was left with