LETTERS TO THE EDITOR

Isolated intracranial hypertension presenting with trigeminal neuropathy

Isolated intracranial hypertension (pseudotumour cerebri) is an idiopathic condition characterised by raised intracranial pressure in the absence of a cerebral mass lesion or hydrocephalus. By definition symptoms and signs are restricted to those of raised pressure including papilloedema and abducens nerve palsies. Other cranial nerve palsies in association with this disease are rare and we report a case presenting with headache and unilateral trigeminal sensory disturbance.

In December 1991, a 20-year-old woman presented to the casualty department with headache and numbness initially affecting the right side of the lips before spreading to involve the right side of the face. She had restarted the oral contraceptive pill two months previously after a one year break. There was no other relevant history.

On examination she was obese with impaired sensation to light touch and pinprick affecting all three branches of the right trigeminal nerve together with an ipsilateral attenuated corneal reflex. Trigeminal motor function was intact and there were no other neurological signs; in particular, fundoscopic examination was normal.

Her symptoms persisted and on review seven days later, examination of the optic fundi revealed bilateral haemorrhagic papilloedema with enlarged blind spots but normal visual acuity; the trigeminal sensory signs remained unchanged. She was admitted for further investigations. A contrast-enhanced CT brain scan was normal and lumbar puncture revealed an opening pressure of 390 mm CSF. The cerebrospinal fluid was acellular with a protein content of 0.3 g/l. Her symptoms improved rapidly after the lumbar puncture and within 48 hours facial sensation had returned to normal and the headache had resolved completely. The contraceptive pill was stopped and she was discharged on acetazolamide. At review a week later examination confirmed normal trigeminal sensory function and resolving papilloedema. The lumbar puncture was repeated and the opening pressure was 190 mm of CSF with normal CSF constituents. Six weeks later her optic discs appeared normal and the acetazolamide was gradually withdrawn. The patient has since been reviewed regularly and has remained asymptomatic for 12 months.

Suggested diagnostic criteria for isolated intracranial hypertension comprise a raised CSF pressure of normal constituents, a normal cranial CT image, and symptoms and signs of raised intracranial pressure alone.¹ The patient in this case satisfied the first three criteria and no other explanation for the trigeminal sensory loss was discovered on examination or investigation. The close temporal relation between the reduction of CSF pressure and a resolution of symptoms and signs suggests that the trigeminal sensory loss may have been a pressure related phenomenon.

Trigeminal palsies may occur as false localising signs secondary to brain tumours.² The postulated mechanisms for this occurrence are direct compression of the trigeminal root by cerebral tissue, traction of the nerve by caudal displacement of the brainstem, or vascular disturbance secondary to the first two insults. Whereas abducens nerve palsies are well recognised in association with isolated intracranial hypertension (9%-36% of cases),³ involvement of other cranial nerves has been described infrequently. Oculomotor,⁴ trochlear,⁵ and facial nerve⁶ palsies have all been reported, most recently in this journal.⁷ To our knowledge only one case report of an isolated trigeminal lesion in association with isolated intracranial hypertension exists.³ This patient presented with a six year history of intermittent symptoms culminating in 12 months of recurrent facial pain. The late development of headache and papilloedema provided the diagnostic clue with resolution of symptoms and signs on appropriate treatment. The current case is a more acute presentation of trigeminal disturbance with numbness rather than pain. Numbness of the face was also reported by Zachariah *et al*⁸ in a patient with isolated intracranial hypertension but the presence of an ipsilateral hemiparesis suggested a higher level of cerebral involvement.

Our report reinforces the fact that patients with isolated intracranial hypertension may present with disturbance of trigeminal function, albeit rarely. We concur with Davie *et al*⁷ that the rapid resolution of symptoms and signs after lumbar puncture in these cases suggests that the signs are falsely localising in nature and as such are compatible with the diagnosis. Recognition of this association may avoid unnecessary investigation in such patients.

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- 1 Ahlskog JE, O'Neill BP. Pseudotumour cerebri. *Ann Intern Med* 1982;97:249-56.
- 2 O'Connell JEA. Trigeminal false localising signs and their causation. *Brain* 1978;101:119-42.
- 3 Hart RG, Carter JE. Pseudotumour cerebri and facial pain. *Arch Neurol* 1982;39:440-1.
- 4 McCammon A, Kaufman HH, Sears ES. Transient oculomotor paralysis in pseudotumour cerebri. *Neurology* 1981;31:182-4.
- 5 Helpert JL, Gordon WH. Trochlear nerve palsy as a false localising sign. *Ann Ophthalmol* 1981;13:53-6.
- 6 Kiywak KJ, Levine SE. Benign intracranial hypertension and facial diplegia. *Arch Neurol* 1984;41:787-8.
- 7 Davie C, Kennedy P, Katifi HA. Seventh nerve palsy as a false localising sign. *J Neurol Neurosurg Psychiatry* 1992;55:510-1.
- 8 Zachariah SB, Jimenez L, Zachariah B, Prockop LD. Pseudotumour cerebri with focal neurological deficit. *J Neurol Neurosurg Psychiatry* 1990;53:360-1.

Spinal intrathecal baclofen suppresses central pain after a stroke

Central pain due to cerebral stroke is one of the most difficult of all pain syndromes to ameliorate. Medical treatment is usually unsatisfactory and surgical intervention in

the thalamus or the midbrain may be indicated. We had experience of a patient who was given lumbar intrathecal baclofen for spasticity associated with dysaesthetic pain in his extremities, after a stroke. The pain was suppressed appreciably with a small dose (25 µg) of intrathecal baclofen, which did not affect the spasticity. Encouraged by this experience, we investigated the effect of intrathecal baclofen in five patients with central pain after a stroke.

The patients were admitted to the neurosurgical ward for this investigation. All the patients had been treated medically without significant pain relief. Intravenous morphine was also ineffective. No patient had been taking oral baclofen. Intrathecal baclofen was given once a day through a lumbar puncture at the L3-4 level. The patients were asked to report their subjective pain hourly with a 10-grade score, in which 0 = no pain and 10 = pain at the pre-treatment level. The injection was repeated three to five times during a week. Normal saline was used once to exclude a placebo effect. The placebo was given between baclofen injections. The baclofen solution for intrathecal use was supplied by Ciba Geigy Corporation (Basel, Switzerland). The original solution (0.5 mg/ml) was diluted 10 times with normal saline for bolus injection. The experimental nature of this investigation and its possible risk were explained to the patients and the family and informed consent to the use of baclofen for central pain was obtained. The procedure was approved by the ethics committee of the Tokyo Women's Medical College.

Case 1

A 60-year-old man had had severe constant dysaesthetic pain in his left upper and lower limbs for five years. The cause was a small haemorrhage in the right posterior thalamus. Allodynia to light touch and anaesthesia to pinprick were noted on the left side of his body. A bolus of intrathecal baclofen (50 µg) was given and he was then allowed to walk around as usual. After one hour he reported considerable reduction (2/10) of the leg pain and after four hours the arm pain was relieved (3/10). The allodynia was also relieved but anaesthesia to pinprick was not affected. The pain relief lasted for about 24 hours. We repeated the same procedure twice and obtained a consistent response. Placebo gave no pain relief. He reported a transient headache after the injection.

Case 2

A 57-year-old woman had had intractable pain for 20 years after a small haemorrhage in the left pons. Her pain was in the right arm and leg. It was constant pain of dysaesthetic and burning nature. Anaesthesia to pinprick was noted in the painful area but allodynia was not observed. We gave 50 µg of intrathecal baclofen, which resulted in good pain reduction (4/10). The pain relief started from the leg and then progressed to the upper arm in three hours. There was no objective change in sensation. The effect continued for about 12 hours. We gave baclofen five times and increased the dose gradually, up to 150 µg, which resulted in pain relief of longer duration. Her gait became unsteady with this dose. Placebo was not effective.

Case 3

A 57-year-old woman had a severe burning pain in her right extremities that developed

two months after a small infarction in the left thalamus. She stated that the pain was constant, burning, and unbearable. There was pronounced allodynia to light touch and cold stimuli. Pinprick showed hyperaesthesia. At first we gave 50 µg of baclofen intrathecally, with no pain relief. The dose was gradually increased to 150 µg but she reported no pain relief. There was transient urinary retention.

Case 4

A 62-year-old man had central pain due to cerebral haemorrhage in the left corona radiata. His pain was in the distal parts of the right upper and lower extremities. There was no allodynia. Hyperaesthesia to pinprick was noted in the right half of his body. A bolus of 50 µg of intrathecal baclofen resulted in considerable pain reduction in an hour (1-2/10), that developed in the upper and lower limbs at the same time. The effect continued for about 12 hours. This response was confirmed three times with repeat intrathecal injections of the same dose. There was no sensory change with the injection.

Case 5

A 47-year-old woman had had disabling central pain for the past two years. The pain was in the right half of her body including the face. This pain started two months after a hypertensive haemorrhage in the left putamen. Although there was no allodynia, hyperaesthesia to pinprick stimuli was noted in the right half of the body. About one hour after intrathecal baclofen (50 µg), pain relief developed in the face first, and then in the arm and leg. She reported pain scores of 4-6/10. We gave the injection five times and the pain relief was consistent. Relief was always associated with suppression of hyperaesthesia. Placebo did not relieve the pain.

All the patients except one (case 3) reported that no other treatment had been as effective as intrathecal baclofen. Although the present study was not double-blind, we do not consider that the result is merely a placebo effect or is due to observers' bias. The pain relief was always associated with baclofen and placebo was ineffective. Although the patients were not informed of the possible course of pain relief, it was constant for each individual patient. After the study period of intrathecal baclofen, we gave oral baclofen (30-60 mg/day) to all patients; no pain relief was achieved.

Baclofen, an agonist of γ -aminobutyric acid (GABA) receptors, has antispastic effects and its intrathecal use relieves severe spasticity.² Furthermore, intrathecal baclofen is known to have an antinociceptive effect in experimental animals, which is not reversed by naloxone.^{4,5} Intrathecal baclofen also suppresses allodynia induced by prostaglandin $F_{2\alpha}$ in mice.³ Recently Herman *et al*¹ reported that intrathecal baclofen relieves central pain in patients with spinal lesions. In their report, even a patient with a C3 lesion showed suppression of leg pain with lumbar intrathecal baclofen.

The mechanism of suppression of central pain after a stroke with lumbar intrathecal baclofen is difficult to explain. In the first two patients, pain relief started from the leg. Therefore we considered that the effect was

segmental and due to a direct effect of baclofen on the GABA receptors in the spinal cord. This supports the hypothesis that the loss of dorsal horn interneurons containing GABA and glycine in humans leads to an allodynic or hyperaesthetic state,⁵ and it should be studied further if such loss of spinal interneurons occurs in central pain syndrome of supraspinal origin. In two patients, however, relief developed in the arms and legs at the same time and even facial pain was relieved. This is not explained by the spinal segmental effect of baclofen. Therefore, we also consider that there might be a GABA-receptor related pain suppression system that ascends from the spinal cord to a higher level.

This preliminary study indicates that a controlled clinical trial of continuous baclofen infusion is feasible for patients with central pain of supraspinal origin.

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- Herman RM, D'Luzansky SD, Ippolito R. Intrathecal baclofen suppressed central pain in patients with spinal lesions. *Clin J Pain* 1992;8:338-45.
- Lazorthes Y, Sallerin-Caute B, Verdier JC, Bestide R, Carillo JP. Chronic intrathecal baclofen administration for control of severe spasticity. *J Neurosurg* 1990;72:393-402.
- Minami T, Uda R, Horiguchi S, Ito S, Hyodo M, Hayaishi O. Effects of clonidine and baclofen on prostaglandin $F_{2\alpha}$ induced allodynia in conscious mice. *Pain Res* 1992;7:129-34.
- Wilson PR, Yaksh TL. Baclofen is antinociceptive in the spinal intrathecal space of animals. *Eur J Pharmacol* 1978;51:323-30.
- Yaksh TL, Reddy SVR. Studies in the primate on the analgesic effects associated with intrathecal actions of opiates, α -adrenergic agonists and baclofen. *Anesthesiology* 1987;54:451-67.

Effect of immobilisation on position and movement sense of the knee

It has been suggested that joint position and movement sense can be improved by practice or by specialised therapeutic techniques such as weight-bearing exercises.¹ Also, motor imbalance after immobilisation may be attributed to proprioceptive deconditioning. To investigate these theories we used established measurement techniques² to test normal knees in 18 patients after a minimum two weeks of immobilisation. Ten patients had been immobilised in a full length leg plaster after fractures of the ankle region and eight were on strict bed rest due to injuries of the untested leg. Results were compared with those of 30 controls with no history of immobilisation.

Movement sense was tested by determining the threshold of perception of slow joint movement at a velocity of 0.5° per second. A motor extended or flexed the knee (starting position 35° flexion) via a system of pulleys connected to the seated and blindfolded subject by a canvas sling wrapped around an inflatable boot. Four randomised tests were carried out in flexion and extension ranges. Subjects signalled

appreciation of knee joint movement by depressing a hand-held switch. The range of movement traversed before detection (threshold angle) was extrapolated from the output of potentiometers placed in parallel with the main pulley circuit.

Position sense was tested by measuring the margin of error in the reproduction of previously held knee joint positions. Tests were carried out within the range 20°-50° of knee flexion—the normal arc of movement during walking. Three conditions operated—namely, active reproduction of an active movement, active reproduction of a passive movement, and passive reproduction of a passive movement. Tests involving active movement were not carried out on the plaster group because of potential effects of muscle weakness and lack of coordination on results. Achievement of target positions was signalled by depressing a hand-held switch.

Results were recorded directly onto computer disc and analysed with two-tailed *t* tests and one way analysis of variance. Tests of movement sense did not show any significant differences either between or within groups (figure). Movement was detected within a mean of 2°.

There were no significant differences in results for position sense either between or within groups. Subjects were accurate to within a mean of 4°. There was a trend, which reached significance in controls ($n = 13$, $p < 0.001$), for greater inaccuracy in active reproduction of a passive movement compared with active reproduction of an active movement and passive reproduction of a passive movement.

In conclusion, results for controls were comparable with those of other studies on the knee joint.^{2,3} No significant differences were detected in position or movement sense between the knees of subjects experiencing altered mobility and weight-bearing conditions and normal controls. The applicability of the results of these tests to normal functional movement is uncertain and warrants further investigation. Results suggest, however, that functional deficits in non-neurological subjects after a period of altered weight-bearing and mobility of the knee joint may be due to factors other than adaptation of position and movement sense mechanisms. Position and movement sense seem to be resistant to changing physical states. The rationale of therapeutic techniques that purport to improve position and movement sense in neurologically intact patients should be reconsidered.

Geometric mean threshold angles in extension range. Right and left = right and left legs of the control group ($n = 30$); immob and other = the previously immobilised leg and the other leg in the immobilised plaster group ($n = 10$); tested = the unaffected leg in the bed rest group ($n = 8$).