

Fig Iohexol myelogram showing narrowing of the subarachnoid space at L4/5 and L3/4.

model that lumbar disc prolapse causes less stretch in the sacral roots than in their lumbar counterparts. The lower tension in the sacral roots may cause relative sparing from the exercise induced ischaemic neuritis which Blau and Logue² thought caused "neurogenic claudication", and this explains the rarity of sphincter movement in this condition.

Most causes of faecal incontinence are "idiopathic" and are not associated with more generalised neurological disturbance. Electromyography and histological studies suggest that injury to the distal nerve supply, perhaps due to stretch injury during straining at stool or childbirth, causes partial denervation of the muscles of the pelvic floor.⁹ It may be that in our patient an already compromised sphincter mechanism was rendered incompetent by an exercise induced alteration in sacral outflow.

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Argyll Robertson pupils in lymphocytic meningoradiculitis (Bannwarth's syndrome)

Sir: Lymphocytic meningoradiculitis or Bannwarth's syndrome is clinically characterised by severe radicular pains with sensory and motor impairment and cranial nerve palsies, especially unilateral or bilateral facial weakness.¹⁻⁴ The syndrome is probably a European variety of Lyme disease,⁵ which is caused by a spirochete, *Borrelia burgdorferi*,⁶ and transmitted by the bite of a tick. In Bannwarth's syndrome antibodies to the same⁷ or to closely related⁸ spirochetes are found. CSF findings such as elevated protein and pleocytosis¹⁻³ are consistent with aseptic meningitis. The disease is often self-limiting, but the course of the disease and the duration of pain may be shortened by high doses of penicillin given intravenously.⁹ In some cases it may be difficult to distinguish Bannwarth's syndrome from neurosyphilis. We present such a case.

A 65 year old man was admitted to the department of neurology because of burning pains in both legs and transient periods of low-grade fever for ten days. The patient had noticed slight weakness and numbness of both legs. He had twice experienced cramping abdominal pains. He did not recall the bite of a tick or any skin lesion. Three years before he had visited our outpatient clinic after a car accident. No neurological abnormalities were found at that time, and the pupils had been recorded as normal.

On admission the patient was extremely loquacious. He appeared well orientated, but his thoughts were incoherent. Neuropsychological testing, which was performed later, showed normal intelligence and intact memory. A cerebellar dysarthria was noted. There was no neck stiffness. Both pupils

were very small and irregular in shape and there was slight anisocoria. The light reaction was almost absent, whereas the response to near-effort was normal and brisk. In darkness the miosis remained unchanged. Visual acuity was normal. The straight leg raising test was positive in both legs. There was a slight weakness of dorsiflexors and extensors of both feet and cerebellar ataxia of both legs. All tendon jerks in the legs were absent. The patient was unable to walk because of severe ataxia.

CSF analysis showed 13 white cells/mm³ and a total protein of 1.01 g/l on admission and after one week 9 white cells, a protein of 1.60 g/l and a normal IgG index and agarose gel electrophoresis. Laboratory analysis showed negative *Treponema pallidum* haemagglutination assay (TPHA), fluorescent treponemal antibody absorption (FTA-BS), venereal disease research laboratory test (VDRL) and Reiter-CIE reaction on two occasions in both blood and CSF. Computed tomography of the brain, ECG and chest radiographs were all normal. Pharmacological tests showed no pupillary response following pilocarpine 0.125% or adrenaline 0.10%, but brisk dilatation after cocaine 4%.

Initially, a diagnosis of neurosyphilis was made and the patient received high doses of penicillin intravenously. After three days right-sided facial weakness developed, together with left-sided facial myokymia. A prominent cerebellar ataxia was now present in both arms as well as in the legs. At that time the results of the diagnostic tests for syphilis proved negative and a diagnosis of Bannwarth's syndrome was considered. This diagnosis was later confirmed by the presence of serum antibodies (IgG) against *Borrelia burgdorferi* in a titre of 1:128, determined by indirect immunofluorescence assay. During and after the penicillin treatment the patient completely recovered and the CSF findings normalised. Only the pupillary abnormalities remained.

This patient demonstrates that lymphocytic meningoradiculitis or Bannwarth's syndrome may closely resemble neurosyphilis. The latter diagnosis was entertained on the basis of the eye signs, when the patient was first seen on admission. When all diagnostic tests for *Treponema pallidum* proved negative, the diagnosis of Bannwarth's syndrome was considered and subsequently confirmed by the demonstration of a high antibody titre against *Borrelia burgdorferi*.

In retrospect, most of the findings in our patient, such as burning radicular pains, leg weakness, facial paresis,¹⁻⁴ facial

myokymia¹⁰ as well as the CSF findings⁹ are compatible with the diagnosis of Bannwarth's syndrome. Unusual findings were the prominent cerebellar ataxia, which has rarely been described in Lyme disease¹¹ and Bannwarth's syndrome,¹² and the small number of white cells in the CSF, which is uncommon in the acute stage of the disease.¹³ The most strikingly unusual finding, however, was the Argyll Robertson syndrome. All criteria for this syndrome were fulfilled namely small and irregular pupils, a more extensive reaction to near-effort than to light, and intact visual function.¹⁴ Since these abnormalities were not noted when the patient visited our out-patient clinic after a car accident some years before, it seems very likely that the Argyll Robertson pupils are indeed related to the present disease.

The abnormalities of the pupils must be differentiated from Adie's syndrome,¹⁴ which may appear later in life, sometimes following a viral infection.¹⁵ However, this abnormality is unilateral in 90% of all cases, the pupil is mydriatic instead of miotic, at least in the acute stage, and the narrowing and redilation during and after convergence is tonic instead of brisk as in our patient.¹⁴ Furthermore, the patient showed no supersensitivity reaction to pilocarpine, which is present in 80% of patients with Adie's syndrome.¹⁶ Other diseases which may cause Argyll-Robertson-like pupils such as diabetes mellitus and von Economo's encephalitis were all easily excluded.

The site of the causative lesion in the Argyll Robertson syndrome is the subject of longstanding controversy. Pupillary light-near dissociation has been documented in a number of peripheral neuropathies,¹⁸ in which the abnormalities were attributed to para-sympathetic denervation of the pupillary sphincter.¹⁸ Most of these cases, however, lacked typical features of the Argyll Robertson syndrome, such as miosis or irregular pupils. At present, most evidence supports a central rather than a peripheral origin of the Argyll Robertson syndrome.^{17 19 20} Loewenfeld concluded that the site of the lesion is probably rostral to the oculomotor nucleus.¹⁷ This lesion destroys the light-reflex pathway but spares the accommodative fibres, the pupillary constriction nucleus (Edinger-Westphal), the efferent fibres to the iris, and the descending corticotectal inhibitory fibres, which cause the miosis by uninhibited pupilloconstrictor tone.¹⁷ Since both neurosyphilis and Bannwarth's syndrome are caused by spirochetes, the presence of Argyll Robertson pupils in our patient might imply that the rostral mid-brain is a predilection site for all spirochetal

infections. Unfortunately, how such a lesion is produced is poorly understood at the present time.²¹

In conclusion, our findings indicate that Bannwarth's syndrome should be considered in seronegative patients with the eye signs of neurosyphilis.

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A 27 year old man was hospitalised because of disturbed consciousness on 11 February 1985. He had had sore throat and a fever of 38°C for a week. On admission he was confused but followed simple commands. Bilateral abductor palsies, spasms of both masseters and the left orbicularis oris, and trismus were present. The deep tendon reflexes were bilaterally brisk with extensor plantar responses. He showed marked ataxia of the extremities. Meningeal signs were absent. On the second day, his extremities were markedly rigid, and dystonic postures, such as opisthotonus, were frequently observed. In addition to the bilateral impairment of upward and lateral gaze, conjugate downward spasmodic movements of the eyes (oculogyric crises) appeared. They consisted of slow tonic downward deviation of the eyes and dilatation of the pupils, each crisis lasting for several minutes (fig). They occurred spontaneously but could also be very easily induced by applying the noxious stimuli such as pinching. They were sometimes but not always accompanied by the exacerbation of the dystonic rigidity of the trunk and extremities. Adenine arabinoside 600 mg/day was started and continued for 10 days. Over the next three days he was stuporous, though trismus, masticatory and facial spasms, and dystonic rigidity subsided gradually. The initial hyperreflexia was replaced by areflexia. Tracheostomy was performed but artificial respiration was not required. On the seventh day, as his consciousness began to improve, he showed marked bulbar palsy and flaccid tetraplegia, though he communicated by blinking (locked-in syndrome).

Although the oculogyric crises also began to subside, they still occurred even at this stage; they finally disappeared on the eighth day. Rapid recovery followed, and his condition returned to normal by mid March. One year later he is well with no signs or symptoms of Parkinsonism.

Cerebrospinal fluid (CSF) obtained on the second day showed: xanthochromia; lymphocytosis, 60/mm³; total protein, 61 mg/dl; IgG, 5.2 mg/dl; and glucose, 86 mg/dl. Electroencephalogram showed marked slowing of the background activity. Brainstem auditory evoked potentials showed normal waves 1, 2, and 3, but waves 4 and 5 were absent bilaterally, suggesting a lesion in the upper pons. Computed tomographic scan and magnetic resonance imaging of the brain were unremarkable. Herpes simplex virus type 1 IgG immunofluorescence antibody titre in the CSF and serum were elevated to 1:2 and 1:160 respectively, both of which returned to normal later. Other viruses showed no remarkable elevation of the antibody titres. No virus was isolated from the throat and CSF.

Although the attack of dystonia and downgaze in the acute phase of this case might mimic what is sometimes called a pontine fit, the downgaze attack should be more accurately called the oculogyric crisis for two reasons: (1) the clinical feature was typical,¹ and (2) it was more prominent than dystonia and frequently occurred alone, independently of dystonia. The oculogyric crisis and dystonia are closely related with each other: both are known to be seen frequently in postencephalitic Parkinsonism

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Oculogyric crisis in acute herpetic brainstem encephalitis

Sir: So far oculogyric crisis has not been reported in acute herpetic brainstem encephalitis, although recently some 10 cases have been reported with its typical clinical features fully documented. We report a patient who showed this unique neurological symptom only at the onset of the disorder.

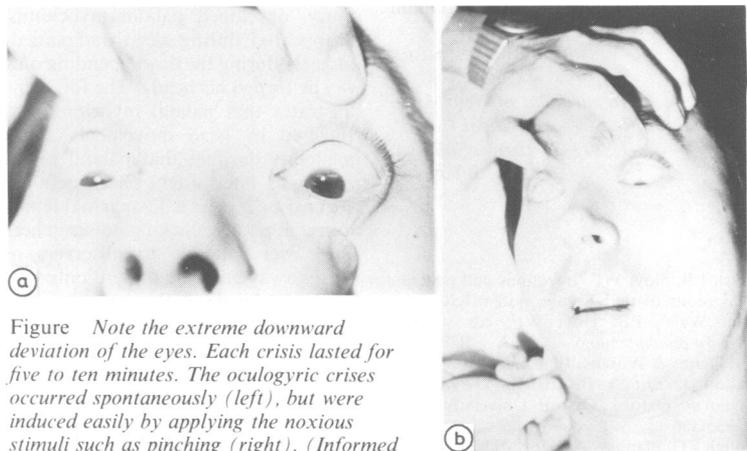


Figure Note the extreme downward deviation of the eyes. Each crisis lasted for five to ten minutes. The oculogyric crises occurred spontaneously (left), but were induced easily by applying the noxious stimuli such as pinching (right). (Informed consent was obtained from the patient for this photograph.)



Argyll Robertson pupils in lymphocytic meningoradiculitis (Bannwarth's syndrome)

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