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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 electrophysiological studies provide interesting evidence of the site of the visual dysfunction.

A 32 year old white male developed symptoms of severe frontal headaches, right 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 papillary 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 α2 globulin; THA/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/lgG/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 dysfuction 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, Coxieilla burnetti, 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 papillary 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.

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 infections with neurological complications is well established.12 Audry-Chaboud1 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...
optic atrophy and a central scotoma in the right eye. Both received steroid therapy, detailed as ACTH for the female and unspecified for the male. Four months after presentation our patient had a residual arcuate scotoma in his left eye, but no optic atrophy. The two differences between our patient and those previously reported were that our patient had a coxsackie B5 infection as opposed to coxsackie B4 in the French patients; secondly our received high dose prednisolone therapy as opposed to ACTH. It may be that the differences in treatment influenced the final visual outcome.

At presentation the visual loss in the left eye was ascribed to optic nerve disease but the electrodiagnostic tests suggest this was more likely to be retinal in origin. The flash ERG showed no definite abnormality in the left eye whereas the PERG was grossly abnormal. It has recently been reported that if the PERG is abnormal in optic nerve disease the abnormality is likely to be confined to the N95 component. The left PERG showed a severe P50 component abnormality in keeping with macular dysfunction, demonstrating the ability of the PERG to determine whether a PVEP delay reflects optic nerve or more distal retinal dysfunction.

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References


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Some historical observations on involuntary movements of the face

Part I

In June 1920, Pierre Marie and Gabrielle Levy1 described 49 patients with transient or permanent movement disorders related to epidemic encephalitis and possibly influenza seen since 1918. Their paper was called "Le Syndrome Excito-Moteur de L’Encéphalite Epidémique". Four patients were described in detail and the movements were categorized as:

1 Choreic, local or generalised, of coarse or small amplitude.
2 Bradykinetic oscillations, coarse, slow, rhythmic movements particularly of the proximal limbs.
3 Myoclonus of either the trunk or the limbs.
4 Parkinsonism, with or without tremor.
5 Isolated tremor, rare and present almost exclusively in the face.
6 Localised facial movements, either tongue-face-masticatory or ocular, sometimes with concomitant facial pain similar to tic doloureux.

Included in the facial movements was trismus as well as difficulty in opening and closing the mouth and involuntary movements of the jaw in several directions. These movements were sometimes accompanied by pain in the face and irritation and grinding of the teeth and a repetitive sucking movement.

There was also muscle spasm around the eyes and mouth and excessive persistent salivation. Speech was sometimes explosive without a real dysarthria and the tongue was often fasciculating.

In the same year Marie and Levy reported three other cases of postencephalitic dystonias. Two of these had involuntary movements shown exclusively as lingual-facial-masticatory disorders. The third had the same movements plus tic doloureux.

A sensation of the throat being obstructed, paroxysmal or constant, was also reported. It was accompanied by disturbances of phonation, shortness of breath or dysphagia, all related to spasm of the muscles of the larynx and pharynx. Abnormal yawning was commonly associated with the development of Parkinsonism and hiccoughs with the myoclonic abnormalities. A sensation of suffocation with an increase in respiratory rate occurred and occasionally a prolonged inspiratory phase as if the patient was sobbing.

RT ROSS

References

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