Letters

methysergide, but is not a serotonin antagonist. An autoimmune basis has been proposed for ergot-induced retroperitoneal fibrosis⁵ and a common autoimmune mechanism has been postulated for retroperitoneal fibrosis and for fibrosis in other sites such as the mediastinum.⁶

All patients receiving bromocriptine for Parkinson's disease require regular monitoring of neurological, psychiatric, cardiovascular and other effects. During follow-up the clinical features of pleuroperitoneal fibrosis and retroperitoneal fibrosis should be sought. This case and others¹⁻⁴ show that a useful screening test for both conditions is the ESR. Chest radiography ray and plasma creatinine measurement may also be indicated.

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Accepted 8 June 1987

Dystonia complicated by respiratory obstruction

Sir: It is not often that dystonia is complicated by respiratory obstruction requiring emergency medical care. We have recently reported two patients who developed dys-

tonic stridor during the course of Parkinson's disease; an urgent tracheostomy was performed in one case.¹ The present case is a 29 year old lady with idiopathic generalised dystonia who required endotracheal intubation and then a permanent tracheostomy for severe laryngeal adductor dystonic spasm.

The patient had been dysphonic from the very onset of her disease at the age of 13 years when she presented with dystonic posturing of the left arm and leg. Initially, she had a quiet dysphonia but from about the age of 26 her speech became unintelligible. At about age 18, she developed retrocollis, left lateral torticollis and axial dystonia with twisting of the trunk to the left. Various drug therapies were tried; including levodopa, which caused confusion and floppiness, benzhexol, which caused hallucinations, tetrabenazine and carbamazepine which were ineffective. Her retrocollis progressed and though she remained ambulant, a posterior cervical ramisection was done at age 27 without success. Regular botulinum toxin injections into orbicularis oculi were commenced a year previously for blepharospasm.

For 2 years, she had experienced episodes of severe generalised muscular spasm, each characterised by opisthotonus, difficulty in breathing and profound diaphoresis. These episodes had become progressively worse having increased from once per month to two or three times daily and from 20 minutes to 3 hours in duration. Recently, she was admitted as an emergency with inspiratory stridor. Parenteral diazepam produced some benefit but respiratory distress promptly recurred. At laryngoscopy, adductor spasm was seen and a nasotracheal tube passed immediately. After discussion with the patient and her mother, a tracheostomy was performed the next day. This proved very effective and there were no further episodes of respiratory distress.

Tracheostomy is apparently very rarely performed for laryngeal adductor dystonia, which may occur (as in this case) in the setting of generalised dystonia, or as an isolated phenomenon.² A more destructive operation, such as ablation of one vocal cord or rendering it paralysed by nerve section may also be considered.^{3,4} There is limited experience with botulinum toxin injection into the laryngeal muscles but this is an attractive option under investigation.⁵

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Accepted 1 June 1987

Parkinsonism in neurobrucellosis

Sir: Since the first description by Hughes in 1897,¹ neurobrucellosis has been the subject of excellent reviews,^{2,3} but in some countries where it is infrequent it still poses a diagnostic and therapeutic problem because of its wide clinical spectrum and the lack of extraneurological findings. Meningitis, meningoencephalitis, meningomyelitis and meningomyelodradiculitis are the most frequently reported forms of nervous system affection.

We report an exceptional neurologic manifestation of brucellosis. A 68 year old woman, living and working under conditions favouring brucella infection, sought medical attention for nocturnal fever, arthromyalgia and profuse sweating 5 months previous to her admission. Brucellosis was suspected and she was treated with streptomycin 1 g IM daily and doxycycline by mouth 100 mg bid for 30 days, with complete recovery. Two months later, an increasing slow and unsteady gait with tremor in both hands appeared. A clinician diagnosed Parkinson's disease and tried levodopa treatment without improvement; bromocriptine did not help. Then the patient was transferred to our hospital. On admission, general physical examination was negative, tests of higher cortical functions, plantar responses and cranial nerves were normal, deep tendon reflexes were brisk (degree 4/5) and bilateral disabling hypokinesia with cogwheel rigidity and rest-

ing tremor was observed. Laboratory investigations were normal except for brucella agglutination titres of 1/80 and non-agglutinating antibodies titre of 1/1280 in the serum. CSF protein, glucose, cells were normal and cultures were negative. Brucella agglutination titres were only slightly positive (1/8). CT scan of the head disclosed no abnormality. *Brucella melitensis* was isolated from two blood cultures performed when the patient had no fever. After levodopa and bromocriptine withdrawal, treatment with doxycycline by mouth 100 mg bid, streptomycin 1 g IM daily and rifampin 600 mg daily was introduced. One month later, the patient was able to walk without assistance, but still had a mild hypokinesia and resting tremor. Doxycycline and rifampin were continued for 2 months, and the patient was discharged free of symptoms.

According to Roger and Poursine's classification, our patient suffered a late onset neurobrucellosis presenting as Parkinsonism. Though considered rare, extrapyramidal forms of neurobrucellosis have been well recorded, especially in Italian publications⁴; amongst these were two cases of subacute hemiparkinsonism.^{4,5} Unlike ours, both may be included in the early form of Roger and Poursine's, their symptoms having appeared during the course of brucellosis. In both cases, right hemiparkinsonism persisted because treatment was limited by streptomycin toxicity and the lack of new antibiotics at the time. The exact pathogenesis of Parkinsonism in brucellosis is still unknown; there is a lack of pathological studies. Gianelli⁴ suggest a focal encephalitis with circumscribed symptoms, related to concurrent treatment (there being normal CSF), probably due to an idiosyncratic immunological reaction. In our opinion, Parkinsonism unresponsive to levodopa and bromocriptine suggests a post-synaptic dopamine receptor lesion, such as corpus striatum vascular inflammation similar to that observed in syphilitic Parkinsonism.⁶ Whether this reaction is immunological and not due to direct invasion by the organism, cannot be determined only on the basis of a negative CSF culture and agglutination titre, because serine pre-treatment CSF cultures obviously are not performed. Besides, CSF agglutination titres in chronic cases are usually low, and non-agglutinating antibodies or complement-fixation titres need to be included in adequate serologic study.² Finally, we believe that previous treatment is the reason for the lack of CSF abnormality, as found in modern or partially treated neurosyphilis,⁷ especially in chronic and

encephalitic forms. All these considerations lead us to recommend serine blood cultures when suspecting brucellosis in patients with unusual neurological manifestations and normal or inconvincing CSF changes, even if fever or other general symptoms are absent. Treatment with two, three or even four drugs, including those with high CSF penetration (rifampin, trimethoprim-sulfamethoxazole), seems to be desirable⁸ but there are neither comparative studies nor large series to verify the best drug combination.

We conclude by emphasising the importance of this sometimes underestimated disease, with a marked CNS affinity, which is still far from being eradicated.

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Accepted 13 July 1987

Pure word deafness and unilateral right temporo-parietal lesion: a case report

Sir: The syndrome of pure word deafness

has up until now only been reported either in patients with bilateral temporal lesions or with a unilateral lesion affecting the middle third of the superior temporal gyrus of the left temporal lobe.^{1,2} We have recently seen a patient with an agnosia predominantly for spoken words who had a unilateral right hemisphere temporo-parietal lesion demonstrated by computed tomography (CT).

The patient, a 61 year old man who lived alone, noticed one morning when he turned on his radio that he was no longer able to recognise sounds; he could not make sense of what voices were saying and could not recognise any musical tunes. He was a keen CB radio operator and reported that "it was as though the audio was quite correct and loud but I could not make out who was speaking. It was as though some of the frequencies had been chopped out". Although spoken conversation with the patient was almost impossible because of his problem interpreting speech, he was able to continue his work as a commercial artist without any apparent problems. A friend eventually persuaded him to seek medical advice. Although there was no history of any previous illness, hypertension or cardiovascular disease, an electrocardiogram showed changes of a previous, apparently asymptomatic, myocardial infarction. On examination the blood pressure was 110/70 mm Hg and the abnormalities found were confined to the nervous system. The patient's spontaneous speech was fluent, appropriate and comprehensible, though with occasional paraphasic errors. He was able to give a reasonable account of his illness. In contrast, his comprehension of the spoken word was restricted to simple direct questions about his age, name and occupation, though comprehension improved if he was aware of the context of the question. He sometimes understood other direct questions such as "how are you?", but he could not reliably obey simple commands such as "sit down" or "stand up". Repetition of spoken material was poor and limited to three digits and two simple words. He could understand the written word normally, responding rapidly and accurately to complex written commands. Object naming and reading out loud were slightly impaired. He had occasional difficulty recognising non-verbal sounds such as rattling keys and tearing paper. He reported that he was no longer able to appreciate music or recognise familiar melodies. Visuospatial and constructional abilities were normal, he was able to perform the block design test of the WAIS without difficulty and perhaps more importantly, his employers had not noticed any impairment.