Pleuropulmonary and retroperitoneal fibrosis associated with bromocriptine treatment

Sir: Both retroperitoneal fibrosis and pleuropulmonary fibrosis have recently been reported following treatment with bromocriptine. We describe the first case in which bromocriptine treatment was associated with both conditions, which may be manifestations of the same disease process.

Mr W, born in 1932, developed Parkinson's disease in 1976. He presented in 1985 with a 2 month history of dyspnoea. He had lost about 13 kg in weight in the preceding year and was complaining of pain in the low back, radiating to the genitalia. The only abnormal finding on examination was dullness to percussion with reduced air entry in the lower zone of the left lung. Chest radiographs showed left lower lobe collapse with an overlying pleural reaction. The lack of mediastinal shift raised the possibility that the changes were longstanding. Plasma urea and creatinine and haemoglobin levels were normal but his sedimentation rate (ESR) was 43 mm/h. Bronchoscopy with brush biopsy showed no evidence of tumour. In September 1986 he still had dyspnoea, and back pain and said that he could only walk 50 yards without severe fatigue. He also reported nocturnal urinary frequency and intermittent vomiting. The chest signs and chest radiograph (fig 1) had changed little since the previous year. Blood tests revealed a normochromic normocytic anaemia (9.3 g/dl), ESR 74 mm/h, urea 18.4 mmol/l and creatinine 331 μmol/l. Intravenous urography (IVU) (fig 2) and abdominal CT showed bilateral hydronephrosis with occlusion of the distal ureters. At laparotomy dense retroperitoneal fibrosis (later confirmed histologically) was found extending from the hilum of the right kidney down to the bladder. Bilateral ureterolysis was performed with immediate improvement in renal function.

The patient had received a levodopa formulation in combination with an anticholinergic drug continuously since 1978. Selegiline was added in 1984. He was also given baclofen from 1984–6 in an attempt to improve his dystonia. Bromocriptine was commenced in 1983 and maintained at 30–40 mg daily in divided doses. It was discontinued after the diagnosis of retroperitoneal fibrosis.

We recognise that fibrotic reactions of the kind observed in our patient cannot be unequivocally attributed to bromocriptine. However, the other drugs which he received have never been implicated. Retroperitoneal fibrosis and pleuropulmonary fibrosis have been reported separately (although never in combination) as complications of bromocriptine treatment. To date, all affected patients including our own have received total daily doses of 30 mg or more, for treatment of Parkinson's disease. Bromocriptine is an ergot derivative like...
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methysergide, but is not a serotonin antagonist. An autoimmune basis has been proposed for ergot-induced retroperitoneal fibrosis\(^1\) 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 pleuropertitoneal fibrosis and retroperitoneal fibrosis should be sought. This case and others\(^1-4\) 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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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 dystonic 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 posture 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 ramiesectomy 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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Parkinsonism in neurobrucellosis

Sirs: 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 meningoymeloradiculitis 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-
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