Letters to the Editor

Non-epileptic attack disorder, psychoseizures and schizophrenia

Sir,

I read with great interest the recent paper on 'non-epileptic attack disorder' by Binnie.¹ Over the past years I have been interested in patients presenting with acute psychosis and labelled as schizophrenics, when in fact they may be suffering from non-epileptic attack disorders. These patients, at least in my own practice, are often post-pubertal adolescents who frequently have a vague history of a preceding viral illness or head injury. They present with the episodes of depression, paranoia, associated with outbursts of irrational behaviour and aggression, often with auditory perceptual changes such as hyperacusis, hypoacusis or frank auditory hallucinations.

Subtle examinations of these patients even before treatment often demonstrate minimal extrapyramidal dysfunction, which may be enhanced with neuroleptic treatment. Although most patients do not demonstrate any abnormality on routine electroencephalogram studies, they do present with non-epileptiform psychogenic seizures, or psychoseizures,² and recent positron emission tomography scan studies seem to confirm abnormal neuronal outbursts in these patients. One patient who was most refractory to most neuroleptics, responded well when treated with valproic acid.

It therefore seems to me that many so-called schizophrenics are actually suffering from a non-epileptic seizure disorder or psychoseizures, and that the ongoing work by Binnie and his colleagues will hopefully solve some of these questions.

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Androgen-producing adenoma masked by obesity

Sir,

Cushing's disease is uncommon in hospital practice while obesity is common. It is therefore important to exclude Cushing's disease and other causes of excess androgen production as a cause of hirsutism in obese individuals.

Recently a patient with simple obesity in whom Cushing's disease was initially excluded but who had an adrenal adenoma, was admitted to our hospital.

The 72 year old woman was admitted with an acute myocardial infarction. She was markedly hirsute with

male pattern hair. She required to shave daily. A review of the case sheet demonstrated that marked hirsutism had originally been noted 11 years previously and during three subsequent admissions to hospital.

Hirsutism was originally noted when she presented with vitreous haemorrhages, was found to be hypertensive, with a blood pressure of 190/100 mmHg and was referred to an endocrine unit.

Clinical features included male distribution of hair over her back, face, breasts and abdomen. Centripetal obesity and purple striae were also noted. A normal diurnal serum cortisol rhythm was present with a concentration of 727 nmol/l at 9.00 a.m. and 84 nmol/l at 12 midnight. An overnight dexamethasone suppression test response was also normal as were her skull and chest X-rays. A glucose tolerance test demonstrated impaired glucose tolerance. A diagnosis of hirsutism, secondary to simple obesity, was made.

On the current admission her serum testosterone concentration was 10.8 nmol/l (reference range <3.5 nmol/l), androstenedione concentration was 50 nmol/l (reference range 2–11 nmol/l) and dehydroepiandrosterone sulphate concentration was raised at 38 µmol/l (reference range 2–10 µmol/l). Computerised tomography of the adrenal glands demonstrated a 3 cm solid left adrenal mass. She declined surgery.

This woman illustrates the relatively benign course that may be followed by androgen-producing adenomas, but nonetheless she had suffered psychologically and the excess androgen production may have contributed to the development of her coronary arteriosclerosis.

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Tertiary hyperparathyroidism in nutritional osteomalacia

Sir,

Tertiary hyperparathyroidism is a rare complication of severe nutritional osteomalacia.¹ We report here an instance of this complication of long-standing osteomalacia.

A 30 year old single Kuwaiti woman presented with bone pains involving the pelvic girdle and rib cage, and progressive proximal muscle weakness of 2 years' duration. There was no history of diarrhoea, weight loss or anticonvulsant use, but the nutritional history assessed by a 24 hour recall showed an average calcium intake of 230 mg/day, phosphate intake of 400 mg/day, and vitamin D intake of 100 IU/day. From age 14 she had

been using an 'abaya' (black veil) and had very little sunlight exposure. Physical examination was normal except for severe proximal muscle weakness and tenderness over the pelvic girdle and thoracic cage. Biochemical evaluation revealed a mild hyperchloraemic metabolic acidosis (pH 7.3, Cl 107 mmol/l), severe hypophosphataemia (0.6 mmol/l), markedly elevated alkaline phosphatase (655 IU/l) and normal serum calcium (2.4 mmol/l). X-rays of the pelvis showed multiple pseudofractures, X-ray skull/lateral view showed a salt and pepper appearance, and hand X-rays showed subperiosteal bone resorption. A urine amino-acid study showed marked generalized aminoaciduria.

Serum parathyroid hormone (PTH) was 237 pg/ml (normal range: 10–55), 25 (OH) vitamin D₃ 16 nmol/l (25–104), and 1,25 (OH)₂D₃ 42 pmol/l (16–42). A Thallium–technetium subtraction scan showed a left inferior parathyroid adenoma and an ultrasound of the neck revealed that the adenoma was about 2.5 cm in diameter. A diagnosis of tertiary hyperparathyroidism due to long-standing nutritional osteomalacia was made. Since therapy with long-acting vitamin D₃ in this setting carries a significant risk of hypercalcaemia,² the patient was treated with 1- α (OH) vitamin D₃, calcium carbonate and neutral phosphate. Five months later, the patient showed a 50% improvement in bone pains and proximal weakness, the alkaline phosphatase decreased to 406 IU/l and PTH decreased to 105 pg/ml, but her serum calcium rose to 2.8 mmol/l and serum phosphate was still low at 0.7 mmol/l. She underwent a neck exploration and a 2.5 cm $\times$ 3 cm left inferior parathyroid adenoma was removed. Postoperatively serum calcium declined to 1.8 mmol/l and phosphate increased to 0.98 mmol/l. The dose of 1- α (OH) vitamin D₃ was increased to 2 μ g/day and 4 months after surgery, the patient is asymptomatic with normal serum calcium and phosphate values, and undetectable serum PTH.

Tertiary hyperparathyroidism is a rare complication of nutritional osteomalacia and may be due to an increase in parathyroid cell mass beyond a critical level as in the experimental model of Gittes and Radde.³ When this complication occurs, it may be prudent to use short-acting vitamin D metabolites, such as 1- α (OH) vitamin D₃ or 1,25 (OH)₂D₃ rather than native vitamin D₃ in order to avert protracted hypercalcaemia.

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Purpura fulminans following a dog bite

Sir,

Prevention of infection post-splenectomy has focused on the risks of infection due to the encapsulated organisms, *Neisseria meningitidis*, *S. pneumoniae* and *H. influenzae*. Currently, advice includes recommending lifelong penicillin prophylaxis, appropriate vaccinations, and the need to seek urgent medical attention, at the first signs of respiratory infections¹ or other febrile conditions. There are, however, other less common infections that may be equally devastating and to which splenectomized patients are peculiarly susceptible.

I would like to report a case of a fulminating septicæmia following a trivial dog bite in an asplenic patient. He had undergone a splenectomy for Hodgkin's disease 10 years previously, and had never taken penicillin prophylaxis.

A 37 year old engineer presented with shock, purpura fulminans and disseminated intravascular coagulation, 3 days after a playful nip on the cheek by his pet dog. A striking malar purpura and incipient gangrene of the tip of his nose was noted. He was treated initially with benzyl penicillin, cefotaxime, flucloxacillin and metronidazole, and was transferred to our intensive care unit for haemofiltration and ventilation. Gram-negative rods were seen within the neutrophils on the peripheral blood film, and a provisional diagnosis of *Capnocytophaga canimorsus* (Dysgonic Fermenter type 2) septicæmia was made. Ciprofloxacin was substituted for the cefotaxime and flucloxacillin, the first reported use of ciprofloxacin in this condition.

There followed a stormy clinical course, complicated by multisystem organ failure. Epileptiform seizures due to accumulation of penicillin resulted in the continuation of ciprofloxacin alone. Blood cultures yielded *C. canimorsus* 11 days after admission.

Despite no microbiological confirmation of meningitis at any time, there was a relentless and unexplained decrease in cerebral function, and after cessation of all sedation for 10 days he still remained comatose. Thirty-five days after admission, the consensus of neurological opinion was that at best a 'severe, near vegetative brain dysfunction' was likely. Withdrawal of all active measures was considered, but not implemented.

On day 37, a slight movement of his hand was noted, and over the next 48 hours he unexpectedly regained consciousness. He was finally discharged home cerebrally intact 2 months after admission. He was given Pneumovax, and advised to take prophylactic antibiotics for life.

Clinicians and patients should be aware of the potential severity of seemingly trivial animal bites in the asplenic patient, particularly if not on antimicrobial prophylaxis. Of the 60 *C. canimorsus* septicæmias reported to date, 86% followed animal bites or contact. One third involved asplenic patients, none of whom were taking prophylactic antibiotics on admission.^{2,3} The overall mortality in asplenic patients was 41%.³

Capnocytophaga canimorsus (DF2) is an oral commensal of many domestic animals, present in 24% of dogs.⁴ The organism is sensitive to penicillin, ciprofloxacin and erythromycin, but is resistant to gentamicin – an important point, since the antibiotic treatment of Gram-negative septic shock often includes gentamicin. In order