chorea occurs when the function of the corpus striatum is disrupted, the specific abnormality in Sydenham's chorea is unknown. The response to valproic acid suggests that the defect affects the GABA neurotransmitter system. Possibly the difference in effectiveness of valproic acid between Sydenham's chorea and Huntington's chorea reflects the degree of severity of the pathologic process or, alternatively, indicates a difference in the pathogenesis of the two diseases.

- ¹ Browne TR. Valproic acid. *N Engl J Med* 1980;**302**:661-6.
- ² Perry TL, Hansen S, Kloster M. Huntington's chorea: deficiency of gamma-aminobutyric acid in brain. *N Engl J Med* 1973;**288**:337-42.
- ³ Lenman JAR, Ferguson IT, Fleming AM, et al. Sodium valproate in chorea. *Br Med J* 1976;ii:1107-8.
- ⁴ Suber DA, Riley TL. Valproic acid and normal CT scan in kinesiogenic familial paroxysmal choreoathetosis. *Arch Neurol* 1980;**37**:327.
- ⁵ Loscher W, Schmidt D. Increase of human plasma GABA by sodium valproate. *Epilepsia* 1980;**21**:611-5.

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Oshawa General Hospital, Oshawa, Ontario, Canada

R S McLACHLAN, MD, FRCP(C), neurologist

Vibrio alginolyticus peritonitis associated with ambulatory peritoneal dialysis

Vibrio alginolyticus is an unusual human pathogen. It is related to *V parahaemolyticus* and its natural habitat is the sea. Human infections reported to date have been mostly wound and ear infections in swimmers and others with marine contact. We believe that the following is the first reported case of *V alginolyticus* peritonitis.

Case report

A 20-year-old man with end-stage renal failure, who had been receiving treatment for six months with continuous ambulatory peritoneal dialysis, presented with signs and symptoms of peritonitis. Several days previously he had been scuba-diving off the South Australian coast and had changed his peritoneal dialysis fluid on the beach without taking adequate precautions against infection.

He presented with severe abdominal pain, appreciable fever, and signs of peritonism. Samples of macroscopically turbid peritoneal fluid were taken immediately from the Tenckhoff catheter for microscopy and culture. Gram staining showed pus cells but no organisms. Treatment was started with continuous peritoneal lavage with flucloxacillin (50 mg/l) added to the dialysis fluid. His symptoms improved but oxidase-positive, Gram-negative bacilli resistant to flucloxacillin (420 colonies/ml) were cultured from the first sample of dialysis fluid collected. The treatment was changed to gentamicin 4 mg/l as *Pseudomonas* was considered to be the likely infecting organism.

Further identification was carried out using routine biochemical tests. The initial reactions suggested that the organism was possibly a member of the *Bacillus* genus. Since Gram staining showed large Gram-negative rods, however, the organism was resubmitted for appareil procédés d'identification (API) and conventional tests. As the API system indicated that the organism was a vibrio (? *V cholerae*) his history was reviewed and marine vibrio suspected. Further identification procedures showed the organism to be *V alginolyticus*, sensitive to aminoglycosides, co-trimoxazole, cephalothin, and cephalixin but resistant to ampicillin.

He made a satisfactory recovery over this period, and after four days cultures of the peritoneal fluid were negative and the gentamicin was stopped. He was discharged from hospital taking oral cephalixin and remained in good health.

Comment

Peritonitis is the most important complication of ambulatory peritoneal dialysis. The infective organism is usually from the patient's own flora and infection occurs after breakdown of the aseptic technique. In this case the infecting organism, *V alginolyticus*, was of marine origin. It has a worldwide distribution¹ and can be isolated from fish and crustacea.² It differs from *V parahaemolyticus* in that it ferments sucrose, is Voges-Proskauer positive, will grow in both 8% and 10% sodium chloride, and swarms on blood agar. An unusual cause of human infection, *V alginolyticus* has usually been associated with skin and ear infections¹⁻⁵ acquired in a marine setting. To our knowledge *V alginolyticus* peritonitis has not been reported previously.

Patients with indwelling Tenckhoff catheters, and especially those receiving ambulatory peritoneal dialysis, should be advised of the infective risk of swimming in salt water. When such patients with marine contact develop peritonitis the possibility that the causative organism may be a marine vibrio, such as *V alginolyticus*, should be considered.

- ¹ Schmidt U, Chmel H, Cobbs C. *Vibrio alginolyticus* infections in humans. *J Clin Microbiol* 1979;**10**:666-8.
- ² Procv P. *Vibrio alginolyticus* in Western Australia. *Med J Aust* 1978;**2**:296.
- ³ Pien F, Lee K, Higa H. *Vibrio alginolyticus* infections in Hawaii. *J Clin Microbiol* 1977;**5**:670-2.
- ⁴ Rubin SJ, Tilton RC. Isolation of *Vibrio alginolyticus* from wound infections. *J Clin Microbiol* 1975;**2**:556-68.
- ⁵ Ryan WJ. Marine vibrios associated with superficial septic lesions. *J Clin Pathol* 1976;**29**:1014-5.

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Departments of Microbiology and Nephrology, The Royal Melbourne Hospital, Melbourne, Victoria, Australia 3050

ROD TAYLOR, MB, BS, nephrology resident
MALCOLM McDONALD, FRACP, microbiology registrar
GRAEME RUSS, FRACP, nephrology registrar
MARA CARSON, RT, microbiology technologist
ELIZABETH LUKACZYNSKI, BAPPS, microbiology technologist

Respiratory and bulbar paralysis with relapsing hyperthyroidism

Bulbar paresis occurring in patients with hyperthyroidism is a well-recognised phenomenon.^{1,2} Reports have cited only difficulty in swallowing and aspiration pneumonia as serious complications of the bulbar weakness. We report a case of hyperthyroid bulbar myopathy that required artificial ventilation to support life.

Case report

A 49-year-old Chinese woman presented with intermittent diplopia and difficulty in swallowing of three weeks' duration. Swallowing fluids often resulted in coughing and spluttering. She noticed that she could no longer whistle. One week after the onset of the symptoms ethinyloestradiol (50 µg) daily had been prescribed. In 1958 she had undergone partial thyroidectomy for hyperthyroidism in Hong Kong. This had been repeated in 1968 because of a recurrence of hyperthyroidism.

Examination showed a fit woman with a resting pulse of 96 beats/min and normal blood pressure. There was no thyromegaly or thyroid bruit. Diplopia with weakness of the right inferior rectus muscle was present intermittently. No exophthalmos or lid lag was noted. Definite bilateral facial weakness, poor palatal movement, and bilateral tongue weakness were evident. Mild shoulder girdle weakness was present. Tendon reflexes and sensory testing were entirely normal. Routine haematology, electrolyte concentrations, and liver function tests, including creatine kinase activity, were normal. Electromyography and nerve conduction studies were normal, and an edrophonium test (10 mg) was negative. Barium swallow showed no peristalsis. Direct laryngoscopy showed no movement of the cords. Thyroid function tests showed: thyroxine concentration 237 nmol/l (18.4 µg/100 ml) (normal 70-140 nmol/l; 5.4-10.9 µg/100 ml); unbound thyroxine binding globulin 95% (85-110% of normal pool); and triiodothyronine 2.8 nmol/l (182 ng/100 ml) (normal 1.1-2.6 nmol/l; 72-169 ng/100 ml). A thyroid pertechnetate scan showed increased uptake in the right and pyramidal lobes. The antithyroid (microsomal) antibody titre was 1/6400 and thyroglobulin antibody titre 1/320.

Soon after admission to hospital she required endotracheal intubation and artificial respiration as her vital capacity had fallen below 600 ml, she could no longer swallow, and arterial blood-gas analysis showed oxygen pressure 8.1 kPa (61 mm Hg), carbon dioxide pressure 7.2 kPa (54 mm Hg), and pH 7.33. Ventilation was required for six weeks. At the end of this period bulbar and respiratory muscle power had almost recovered. During her admission treatment consisted of carbimazole 20 mg daily and prednisolone 100 mg on alternate days. Thyroid function (biochemically) had returned to normal two weeks before she could be weaned off the ventilator. At follow-up three months later she was well with no symptoms.

Comment

In patients with hyperthyroidism weakness may be due to a thyrotoxic myopathy, hypokalaemic periodic paralysis³ (particularly

in Orientals), or myasthenia gravis.⁴ Initially, bulbar muscle dysfunction was believed to complicate a pre-existent, proximal skeletal muscle weakness. Kammer and Hamilton² and other workers showed that hyperthyroid patients without evidence of chronic myopathy sometimes presented with dysphagia. Such patients complained of dysphagia, nasal regurgitation of solids and liquids, voice changes, diplopia, facial paresis, and diminished tongue strength. Nasogastric feeding and intensive physiotherapy to protect the airway was sometimes required. This patient presented acutely and had all of these symptoms but, unlike similar patients, required artificial respiration for six weeks because of complete paralysis of laryngeal and intercostal muscles and inability to swallow.

This case shows that hyperthyroid patients without chronic myopathy may present with a disabling non-myasthenic bulbar myopathy that may require intubation or tracheostomy because of the acute bulbar paralysis.

¹ Joasoo A, Murray IPC, Steinbeck AW. Involvement of bulbar muscles in thyrotoxic myopathy. *Aust Ann Med* 1970;4:338-40.

² Kammer GM, Hamilton CR. Acute bulbar muscle dysfunction and hyperthyroidism. A study of four cases and review of the literature. *Am J Med* 1974;56:464-70.

³ Ali K. Hypokalaemic periodic paralysis complicating thyrotoxicosis. *Br Med J* 1975;iv:503-4.

⁴ Millikan CH, Haines SF. The thyroid gland in relation to neuro-muscular disease. *Arch Intern Med* 1953;92:5.

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Department of Neurology, Royal Perth Hospital, Perth, Western Australia 6001, Australia

J EDELMAN, MB, BS, medical registrar

E G STEWART-WYNNE, MRCP, FCP(SA), neurologist

Pronounced cerebellar features in legionnaires' disease

Confusion and disorientation are common in legionnaires' disease, and isolated localising neurological signs occur occasionally. We report a case of *Legionella pneumophila* pneumonia associated with pronounced cerebellar disease.

Case report

A 47-year-old man presented with a 48-hour history of malaise, generalised weakness, and shivering attacks, associated with production of mucopurulent sputum. In the 24 hours before admission he had had several episodes of vomiting and diarrhoea and his gait had become so unsteady that he was barely able to stand. On waking on the day of admission he had pronounced slurring of his speech.

Since undergoing a splenectomy for hereditary spherocytosis 29 years previously he had had several episodes of pulmonary embolism and deep vein thrombosis and remained permanently on anticoagulants, but his therapeutic compliance was poor. He smoked 40 cigarettes and drank at least eight pints of beer each day, but had continued regular work.

On admission he appeared distressed (respiratory rate 30/min) and feverish (38.6°C) with a pulse rate of 100/min. There were crepitations and rhonchi throughout both lung fields. Despite pronounced dysarthria he was orientated in time, place, and person, and he co-operated during history and examination. Examination of cranial nerves showed no abnormality but nystagmus was present. Co-ordination was impaired on finger-nose testing, and he had associated dysmetria and dysidiadochokinesia. Co-ordination on heel-toe testing was similarly impaired, with slight weakness in the left leg. Plantar responses were flexor. There was shadowing in the lingula area on a chest x-ray film. Investigations showed a haemoglobin concentration of 16.7 g/dl, white cell count of $24 \times 10^9/l$ (mainly neutrophils), urea concentration of 9.6 mmol/l (57.6 mg/100 ml), and serum sodium concentration of 127 mmol(mEq)/l. Despite improving initially with intravenous ampicillin he had a sudden cardiorespiratory arrest on the fifth day of his illness. Legionnaires' disease was considered at this stage, and intravenous erythromycin was given for two weeks followed by amikacin, metronidazole, and cloxacillin because of persistent fever and raised white cell count. Ventilation had to be continued for six weeks, during which time he developed the clinical and neurological signs of adult respiratory distress syndrome (figure). Hepatic and renal function deteriorated during this period but returned to normal with conservative treatment. Dieterle silver stain consistently failed

to show *L. pneumophila* in his sputum, transtracheal aspirate, and trans-bronchial biopsy specimens. Examination of the cerebrospinal fluid showed no abnormality on two separate occasions. There was a rise in *Legionella* fluorescent antibody titre from 1/16 at five days to 1/512 at four weeks.

Limitation of upward gaze was the only residual neurological sign after recovery and this improved slowly. Two months after artificial ventilation was discontinued he still had restricted lung function and carbon monoxide transfer was 40% of that expected.

Chest x-ray film showing bilateral pulmonary disease leading to the development of adult respiratory distress syndrome in a patient with legionnaires' disease.

Comment

Many of the clinical features of legionnaires' disease were present in this case.¹ Although isolated ataxia has been described there has been only one well-documented account of profound cerebellar disease.² In all but one case in which neurological signs have been reported the cerebrospinal fluid was normal.²⁻³ Alcohol was considered as a cause of this man's cerebellar signs, but he regularly took Brewer's yeast tablets (0.2 mg thiamine daily), there was no evidence of an encephalopathy or delirium tremens, and the onset of acute cerebellar signs began 48 hours after his initial symptoms and were thus directly related to the onset of legionnaires' disease.

There were other interesting features in this case. Firstly, legionnaires' disease occurs more often in compromised hosts or in patients with underlying pulmonary disease, and splenectomy in this man may have further increased his susceptibility.⁴⁻⁵ Secondly, recovery occurred only after six weeks' artificial ventilation, and this is unusual though a permanent diffusion defect remains. Legionnaires' disease should be considered in all cases of pneumonia associated with cerebellar signs.

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¹ Swartz MN. Clinical aspects of legionnaires' disease. *Ann Intern Med* 1979;90:492-5.

² Shetty KR, Cilyo CL, Starr BD, Harter DH. Legionnaires' disease with profound cerebellar involvement. *Arch Neurol* 1980;37:379-80.

³ Friedman HM. Legionnaires' disease in non-legionnaires: a report of five cases. *Ann Intern Med* 1978;88:294-302.

⁴ Saravolatz LD, Burch KH, Fisher E, et al. The compromised host and legionnaires' disease. *Ann Intern Med* 1979;90:533-7.

⁵ Gump D, Frank R, Winn WC, Foster RS, Broome CV, Cherry WB. Legionnaires' disease in patients with associated serious disease. *Ann Intern Med* 1979;90:538-42.

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University of Liverpool, Liverpool L69 3BX

MICHAEL R MASKILL, MB, MRCP, lecturer and honorary senior registrar in clinical pharmacology

Royal Liverpool Hospital, Liverpool

E CLARE JORDAN, MB, FFARCS, anaesthetic registrar