probable diagnosis of osteomalacia. The extremely low plasma 25-hydroxy vitamin D (25-OHD) concentration confirmed the diagnosis. Calciferol replacement treatment 3000 units daily was started. Two months later a mean unilateral hearing improvement of 12 db was apparent.

CASE 2

A 49-year-old vagrant Irish ex-labourer was admitted in a state of confusion, dishevelment, and dehydration. One year previously he reported the onset of muscular weakness, progressive bilateral deafness, tinnitus, and episodic vertigo. Neurological examination showed gross wasting and weakness of the small hand, calf, and deltoid muscles. The tympanic membranes were normal; there was no spontaneous nystagmus. Audiometry showed a trough-shaped 55 db bilateral recruiting deafness, and caloric testing bilateral canal hypofunction.

Biochemical evaluation (table) showed a reduced 25-OHD concentration, which was consistent with osteomalacia, probably due to inadequate dietary vitamin D intake. Results of cochlear tomography were normal, but skull radiography showed pronounced basilar impression. The temporal relation of the auditory and muscular symptoms suggested a direct correlaton. He discharged himself from hospital before treatment could be started.

Results of preliminary biochemical evaluation and radiology in two patients

	Case 1	Case 2
	Plasma variables	
Venereal disease research laboratory test Treponemal haemagglutination test Fluorescent treponemal antibody absorption test	Negative	Negative
Calcium (corrected) (mmol/l)	2·16	2·56
Inorganic phosphate (mmol/l)	0·68	0·90
Alkaline phosphatase (U/l)	193	482
Urea (mmol/l)	4·0	2·7
Creatinine (μmol/l)	66	55
25-OHD (nmol/l)*	<5·3	15·3
	Radiology	
Skull	Normal	Pronounced vertebrobasilar impression
Cochlear tomography	Demineralisation	Normal
Skeletal survey	Normal	Normal

*Measured by competitive protein-binding assay.
Conversion: SI to traditional units—calcium, inorganic phosphate: 1 mmol/l = 2 mEq/l. Urea: 1 mmol/l ≈ 6 mg/100 ml. Creatinine: 1 μmol/l ≈ 11·3 μg/100 ml. 25-OHD: 1 nmol/l = 0·4 ng/ml.

Comment

It is generally accepted that serum abnormalities of calcium, inorganic phosphate, alkaline phosphatase, and 25-hydroxycholecalciferol or 25-OHD may enable early osteomalacia to be diagnosed in the absence of radiological features of demineralisation. In the series of Dunnigan *et al* only 29% of children with biochemical evidence of rickets showed radiological abnormalities.[2] Localised bone pain and muscle wasting are recognised symptoms of osteomalacia, but deafness has not been reported as a presenting feature.

Deficiency of vitamin D or its metabolic derivatives 25-hydroxycholecalciferol and 1,25-hydroxycholecalciferol may result directly in auditory dysfunction, or exert a secondary effect by disturbances of calcium metabolism. Calcium ions play an important part in membrane permeability. Active transport mechanisms that include ATPase activity, which maintain the differential biochemical integrity of the inner ear fluids vital for normal cochlear function, may be calcium dependent. In addition, ionised calcium is necessary for normal nerve function and deficiency may adversely affect transmission of the nerve action potentials generated by the cochlea. Retrocochlear deafness in hereditary vitamin-D-resistant rickets, due to secondary hyperparathyroidism causing osteosclerotic narrowing of the internal auditory canals, has been described.[3]

Basilar impression is a bony, vascular, and neural malformation in the craniocervical region that may be associated with deafness and episodic vertigo.[4] The acquired type is rare but may occur secondary to osteomalacia.[5] In case 2 we are not certain whether a direct relation between the two conditions existed as no other skeletal radiological abnormalities were present. Finally, demineralisation of the otic capsule may lead to secondary degenerative changes in the spiral ligament, stria vascularis, and cochlear hair cells. Whatever the mechanism, the threshold improvement in case 1 after treatment suggests a previously unrecognised causal correlation between vitamin D deficiency and cochlear deafness.

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Department of Otolaryngology, The London Hospital, London E1 1BB
G B BROOKES, FRCS, senior registrar
A W MORRISON, FRCS, DLO, senior consultant surgeon

Valproic acid in Sydenham's chorea

Valproic acid is an effective anticonvulsant that may act by facilitating the gamma-aminobutyric acid (GABA) neurotransmitter system.[1] Patients with Huntington's chorea who have low brain concentrations of GABA do not respond to treatment with valproic acid,[2 3] but one patient with familial paroxysmal choreoathetosis improved when taking the drug.[4] I present the first report of a patient with Sydenham's chorea treated with valproic acid.

Case report

A 19-year-old previously healthy woman developed twitching of the right hand that progressed to all parts of the body over the next two months. Stress or fatigue exacerbated the movements, which disappeared during sleep. She came to hospital when she began to have slurred speech and difficulty in walking. She had no neurological symptoms apart from emotional lability noted by her mother. Although she had experienced frequent upper respiratory infections, she could not recall any specific illness before the onset of her movement disorder. A non-pruritic, evanescent, red rash had been present on her trunk and proximal limbs for two months, and during the first month of her illness her right shoulder and right thumb ached. At no time did she have fever, joint swelling, chest pain, or shortness of breath. There was no family history of chorea.

On examination the most prominent finding was pronounced choreoathetosis of the arms, legs, trunk, face, eyes, and tongue. Muscle tone was reduced. She had a macular, erythematous, irregular rash on the trunk and upper arms during the first day in hospital. There was a moderate aortic systolic ejection murmur and softer mitral systolic murmur at the apex. She did not have fever, arthritis, or skin nodules. Antistreptolysin titre was 166 Todd units. The following investigations were normal: erythrocyte sedimentation rate; C-reactive protein concentration; titres of antihyaluronidase, antinuclear antibody, and anti-DNA; lupus erythematosus test; serum protein electrophoresis; throat swab; concentrations of triiodothyronine, thyroxine, and electrolytes; cerebrospinal fluid; electroencephalography; computed tomography; electrocardiography; echocardiography; and chest *x*-ray examination.

The oral contraceptive pill that she had been taking for one year was stopped and she was given diazepam and penicillin. Her chorea continued to worsen until her speech was unintelligible and she had difficulty in swallowing and could not walk unassisted. Valproic acid 250 mg twice daily was started; within six hours the chorea had begun to lessen and after 48 hours she could walk and swallow normally. Speech also improved but mild chorea persisted. After four days the valproic acid was stopped. By the next day her chorea and dysarthria had worsened. When the drug was restarted again at the same dose three days later speech returned to normal and the chorea lessened to affect only the arms and abdominal muscles. Chorea gradually disappeared over the next month and the drug was stopped.

Comment

When the presenting symptom is chorea, as in this patient, laboratory tests often do not confirm the diagnosis of rheumatic fever. Other symptoms and signs and the absence of other causes support the diagnosis of Sydenham's chorea, although the oral contraceptive pill used by this patient may have influenced the severity of the disease.

The rapid response to low-dose valproic acid and the subsequent deterioration when the drug was stopped support the effectiveness of the treatment. Valproic acid increases GABA throughout the body,[5] but its effectiveness in chorea is probably centrally mediated. Although

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.

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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 cephalexin 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 cephalexin 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 distribution[1] 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 infections[1-5] 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.

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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, BAPPSC, 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 paralysis[3] (particularly