SHORT REPORTS

Myokymia, not myotonia

We wish to make a preliminary report of a case of myokymia with sustained muscular contraction and continuous motor unit activity at rest. We do so for two reasons. It is a new case in a very rare group of disorders. It is also essential to distinguish this type of disorder from the myotonias.

Case report

In June 1976 a 26-year-old man was admitted for investigation. He had enjoyed perfect health until 1971, when he experienced increased difficulty in playing the guitar. His manual dexterity deteriorated further while he was in Nigeria, where myotonia was diagnosed. He contracted infective hepatitis and returned to London in 1973. He now had great difficulty in relaxing his limb and facial muscles, but within 24 hours of treatment with carbamazepine felt almost total relief from these symptoms. In 1974 he was told he probably had dystrophica myotonica. By this time, within six hours of omitting a dose of carbamazepine, he experienced difficulty with breathing, walking, and speaking, and had to perform any movement several times before losing the sensation of stiffness. He had never experienced weakness, painful cramps, or sensory symptoms. There was no family history of a similar disorder, but, his wife being pregnant, he was worried by the genetic implications of his condition.

He was a fit man of normal build, posture, and muscle bulk. There was no sign of hyperthyroidism, no frontal balding, and his genitalia were normal. No excessive sweating was noted. Examined while he was taking carbamazepine, 200 mg twice daily, muscle power and tendon reflexes in the limbs were normal, but tone in all limbs was uniformly increased. There was a continuous, undulating movement in his muscles, especially the deltoids, quadriceps femoris, and the tongue. Within a few hours of discontinuing

medication, the spontaneous abnormality of muscle movement with the patient at rest became more obvious. It differed from fasciculation in that the contraction rippled along the muscle down the same route repeatedly, and occurred regularly within the same area of muscle; it was coarser than the contractions typical of fasciculation. He became slightly dysarthric, and was unable to relax his grip other than very slowly. There was no percussion myotonia either in the thenar eminence or in the tongue. The result of sensory examination was normal.

The results of routine investigations were normal, and in particular the following: plasma potassium, magnesium, and thyroxine; serum calcium; blood glucose, random and fasting; and cerebrospinal fluid. Off carbamazepine, his FEV₁ was 5.1 l and FVC 6.2 l. Serum creatine phosphokinase was 18 U/l (upper normal) but serum hydroxybutyrate dehydrogenase was 308 U/l (normal 140 U/l). Slit lamp examination showed no cataracts. Electromyography (EMG) with a concentric needle electrode showed continuous electrical activity in all muscle groups. This was suppressed after exercise for 30 to 60 seconds. The constituent potentials were typical normal motor unit potentials, quite distinct from myotonic discharges. From other EMG findings (fig) it appears that the spontaneous unit activity was of peripheral origin.

Comment

The EMG confirmed our clinical impression of myokymia rather than myotonia. The spontaneous, continuous motor unit activity in a muscle at rest is the one constant diagnostic feature of what clearly appears to be a heterogeneous group of disorders. Even the EMG findings vary slightly among the cases reported.^{1,2} Individual clinical features, as reviewed by Welch, Appenzeller, and Bicknell,² are even more variable. The prognosis of the group as a whole is good,² and the response of our patient to carbamazepine augers well for his future. He was extremely relieved to shed the diagnosis of dystrophica myotonica. His return to Africa meant the postponement of further investigations.

We thank Dr H E Webb for permission to report on this patient.

¹ Gardner-Medwin, D, and Walton, J N, *Lancet*, 1969, **1**, 127.

² Welch, L K, Appenzeller, O, and Bicknell, J M, *Neurology*, 1972, **22**, 161.

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Surface recorded EMG from 1st dorsal interosseous muscle of right hand with variable spontaneous activity. (a) Large unit potentials observed firing irregularly. Activity is of peripheral origin because: (b) maximal stimulation of ulnar nerve caused no silent period, and (c) bupivacaine block of the ulnar nerve, reducing voluntary power by 75%, showed no effect. Calibrations: 500 μ V and 25 ms.

Atypical infectious mononucleosis with bone marrow granulomas and pancytopenia

Bone marrow granulomas rarely complicate infectious mononucleosis. The nature of this case remained obscure until the patient's daughter had a febrile illness with typical glandular fever blood count and positive result to the Paul-Bunnell test.

Case report

A 39-year-old man presented with a 10-day history of malaise, severe headache, weight loss, and fever. His temperature was 40°C and he had no glandular enlargement or palpable spleen. Blood count: haemoglobin 13.9 g/dl; erythrocyte sedimentation rate (ESR) 16 mm in 1 h (Westergren); leucocytes $4.8 \times 10^9/l$ (4800/mm³), neutrophils 37%, lymphocytes 62%, monocytes 1%. Neutrophils showed a left shift, with a few Turk cells present.

The result of the Paul-Bunnell test was negative, as were tests on sputum and urine and the cerebrospinal fluid. Blood culture grew no organisms, the chest x-ray film was normal, while the throat swab grew β -haemolytic streptococci group F. Two days later his condition deteriorated, with temperature 41°C , tachycardia 130 beats/min, and triple rhythm. Electrocardiogram showed S-T elevation in anterior chest leads. Haemoglobin 9.9 g/dl; leucocytes $3.0 \times 10^9/\text{l}$ (3000/mm³), neutrophils 42%, lymphocytes 39%, monocytes 19%; platelets $47 \times 10^9/\text{l}$; ESR 110 in 1 h; aspartate aminotransferase 208 IU (normal 0-40 IU), creatine phosphokinase 67 IU (normal 0-50 IU), lactate dehydrogenase 2320 IU (normal 200-560 IU), and gamma-glutamyltranspeptidase 46 IU (normal 0-45 IU). Tests for Epstein-Barr (EB) virus and cytomegalovirus antibodies were negative. A bone marrow film showed moderate cellularity but no significant abnormality. The histology revealed histiocytic granulomas with poorly formed giant cells and some foci of necrosis (see figure). Many of the granulomas surrounded circular spaces which contained fat.

Histiocytic granulomas. (Haematoxylin and eosin $\times 60$.)

Fever continued without remission, and he was given minocycline, 200 mg daily, for suspected brucellosis without effect. After six days this was changed to co-trimoxazole. Nevertheless, brucella agglutinations, complement fixation, and antiglobulin tests were negative. His weight had fallen 17 kg, and this continued, so that antituberculous treatment was started because the bone marrow histology was thought to suggest tuberculosis. At the same time information was received that the patient's daughter had glandular fever. So Paul-Bunnell, differential absorption, and EB-virus antibodies tests were all repeated in the patient and were found to have become strongly positive. Sheep erythrocytes agglutination titre 1/512, after absorption with guinea-pig kidney titre 1/512, after ox cells 1/14. EB-virus antibody titre IgG 1/128, IgM positive. At that time the haemoglobin was 9.6 g/dl; leucocytes $2.9 \times 10^9/\text{l}$ (2900/mm³), neutrophils 51%, lymphocytes 35%, monocytes 11%, basophils 2%, eosinophils 1%, platelets normal.

Anti-tuberculous treatment was stopped, and 24 days after admission prednisolone, 60 mg daily, was begun, the dose later being steadily reduced. One week later haemoglobin was 11 g/dl, leucocytes and platelets normal. The abnormal enzyme values and electrocardiogram returned to normal, and prednisolone was stopped after nine weeks. One year later he remained in good health. The results of tests on the bone marrow, blood count, and serum enzymes and electrocardiogram were all normal; the EB-virus antibody titre was IgG 1/128, IgM negative.

Comment

The granulomas of brucellosis, sarcoidosis, and tuberculosis are similar to those of infectious mononucleosis, though the lesions of the first three are larger, more nodular, and more circumscribed than those of infectious mononucleosis—which are smaller and usually lack giant cells.¹ Pancytopenia only rarely complicates infectious mononucleosis,² and a report which illustrated its association with a lack of clinical manifestations has postulated that there is also a

focal granulomatous process in the marrow rather than a simple hyperplasia.³

Bone marrow examination is rarely needed in infectious mononucleosis. The main recorded reasons² for it are anaemia, or low platelet or granulocyte counts, usually associated with a definite generalised hyperplasia of erythroid, myeloid, megakaryocytic, and reticulum cell elements. Granulomas have been found in about one-third of these cases.

Serious suppression of marrow function is also rare, though aplastic anaemia has been reported.⁴ Granulomas usually arise in response to an antigenic stimulus.⁵ As there are many antibodies in infectious mononucleosis, granulomas may be more common than suspected,² although often asymptomatic. This patient, who lacked any of the normal clinical manifestations of infectious mononucleosis, showed a significant rise in heterophil and EB-virus antibodies during his illness. He not only had a myocarditis and pancytopenia, but illustrated another variation in the type of granulomas that are found. Unlike described lesions, these showed histiocytic cells with occasional giant cells surrounding a central circular area of fat.

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² Boyd, J F, and Reid, D, *Journal of Clinical Pathology*, 1968, **21**, 683.

³ Read, J T, and Helwig, F C, *Archives of Internal Medicine*, 1945, **75**, 376.

⁴ Worledge, S M, and Dacie, J V, *Infectious Mononucleosis*. Oxford, Blackwell Scientific Publications, 1969.

⁵ Rothwell, D J, *Archives of Pathology*, 1975, **99**, 508.

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Axillary hyperhidrosis: failure of subcutaneous curettage

Axillary hyperhidrosis (excessive perspiration) is a condition which is being encountered more and more frequently in general practice, dermatological clinics, and surgical departments. This is probably due to the stricter hygienic and social conditions of modern life and also to the increasing awareness of patients that effective treatment is now available.

The vast majority of patients have primary or idiopathic hyperhidrosis.¹ Secondary hyperhidrosis due to hyperthyroidism, obesity, anxiety states, the menopause, or disseminated malignant disease, and paroxysmal sweating due to pheochromocytoma are usually easy enough to differentiate because of their obvious accompanying manifestations. The primary type affects males and females equally, but girls and women are far more likely to complain of the axillary sweating.

Surgical treatment

Axillary sweating originates mostly from the eccrine glands located in a comparatively small area which roughly corresponds to the hair-bearing zone of the axilla. This can be mapped out easily by the application of a starch-iodine powder, which turns blue in contact with moisture. Surgical treatment of this condition comprises ablation of the sweat glands from this region. Skoog and Thyresson² first reported a technique of excision of these sweat glands in 1962. Four skin flaps are raised and the glands are snipped away from the underlying surface of the flaps by fine scissors. Hurley and Shelley³ reported good results after excision of the involved ellipse of axillary skin with primary wound closure. The results are satisfactory but undoubtedly a rather wide and ugly scar may result in the axilla, particularly in the skin excision technique. A follow-up of our first 50 patients treated by axillary skin excision (7 male and 43 female), reviewed after a year, showed that 47 were pleased with the results. Only three noted no improvement. Never-