within a week. In July 1986, he developed tender induration in both calves and the right thigh. Again, spontaneous remission was swift. At no stage has there been any evidence of a diffuse myopathy or systemic illness. Throughout the course, the CK has been markedly elevated (see table), only once falling into the normal range.

Polymyositis typically presents as a symmetrical proximal myopathy. The clinical diagnosis is confirmed by muscle biopsy showing lymphocytic inflammatory cell infiltrate and fibre necrosis. This is supported by elevated blood levels of CK and other sarco-plasmic enzymes and characteristic EMG findings of myopathy—spontaneous fibrillation, positive sharp waves and polyphasic motor unit potentials.

Cumming *et al*¹ first described the focal presentation of this disease, and for it coined the term "localised nodular myositis". Since then there have been rare cases,²⁻⁵ the majority of which rapidly progressed to the full clinical syndrome, despite steroid therapy. We describe a patient with three definite episodes of localised nodular myositis, including biochemical, EMG, and muscle biopsy support of the diagnosis. At the time of his first presentation his quadriceps biopsy was histologically normal, when his gastrocnemius was very inflamed. Despite a continuous marked elevation in CK levels, the patient has remained well, fully active, and with no evidence of generalised myopathy. Immunofluorescence testing revealed C₃ and IgM within blood vessels, supporting the evidence of a vasculitic element in the pathogenesis of polymyositis. We have chosen not to treat an elderly man with steroids since he has no disability, despite the fluctuating CK levels.

In the past, there has been confusion between localised nodular myositis, chronic focal polymyositis, and focal myositis. The first two are unusual manifestations of polymyositis, whereas focal myositis is a benign inflammatory pseudotumour. In chronic focal polymyositis, highly selective muscle wasting and weakness remains confined to individual groups of muscles. This has been described for the forearm flexors, brachio-radialis and quadriceps,⁶ and, more recently, the bulbar muscles.⁷ Focal myositis⁸ presents as a lymphocytic infiltration, scattered muscle fibre necrosis, and interstitial fibrosis. There is never sign of diffuse muscle involvement or systemic disease, and both blood levels of CK and ESR remain normal. The aetiology is unknown.

Whether the condition of localised nodular myositis is a distinct entity is not clear. In the literature, previously reported

cases have been a mode of presentation of polymyositis. It is still possible that a generalised myopathy may develop in this man. It is remarkable that it has not happened in the 3 years that he has been under review.

Localised nodular myositis must be remembered in the differential diagnosis of the focal tender muscle mass, which includes myositis ossificans, proliferative myositis, pseudosarcomatous fasciitis, granulomatosis and pyomyositis.

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Rhabdomyolysis associated with simultaneous Epstein-Barr virus infection and isolation of echovirus 6 from muscle: a dual infection

Sir: Acute rhabdomyolysis has been associated with a number of viral infections,¹⁻¹⁰ which has been confirmed by a significant rise in serum antibodies or by

isolation of virus from throatwashing, CSF, urine or stool. Previous studies reveal that only influenza A and B, adenovirus type 2 and Coxsackie-virus type A9 have been isolated from muscle tissue.² We report a case of rhabdomyolysis associated with a dual viral infection. Epstein-Barr virus (EBV) was identified serologically and echovirus isolated from muscle.

A 17 year old boy was suffering from a flu-like disease with headache, fever (40°C), and progressive weakness of legs and arms. When after 3 days he started voiding dark coloured urine and when amoxicillin medication failed to lower his body temperature, he was admitted to hospital. His previous medical history was non-contributory. Previously he had been a healthy young man attending secondary school without exercise-related muscle complaints and his family history gave no evidence of muscle disorders.

On admission the rectal temperature was 39°C and the patient appeared mildly dehydrated. Blood pressure was 120/60 mm Hg and pulse rate 92 beats per minute. Some small axillary and inguinal lymph nodes were palpated. Liver and spleen were not enlarged. Thigh muscles were tender to palpation and weak against resistance. The patient was unable to stand alone without support. The result of the remainder of the physical examination was within normal limits. On admission a ESR of 3 mm/h was noted. WBC was 9.2/mm³ with 17% segmented forms, 14% bands, 61% lymphocytes, and 8% monocytes. Serum enzymes were strongly elevated: CK 54 050 U/l (normal 15-91 U/l), ASAT 864 U/l (normal 7-25 U/l), ALAT 198 U/l (normal 5-25 U/l) and LDH 5100 U/l (normal 155-275 U/l). Gamma-GT was normal. Urine analysis showed proteinuria (2+) and many pigmented casts were detected. Immunoassay for myoglobin revealed a very high level in serum of 11 100 ng/ml (normal 0-85 ng/ml) and presence in urine of 824 500 ng/ml. AST and ANF were normal and circulating immune complexes absent. The qualitative mononucleosis slide agglutination test was positive. By indirect immunofluorescence IgM and IgG antibodies to EBV were detected at titres of 1:512 and 1:2048 respectively. Complement fixation antibody titres to adenovirus, influenza virus, mycoplasma pneumoniae, parainfluenza virus, measles and herpes simplex virus were negative.

On the 9th day after admission a needle biopsy of the quadriceps muscle was carried out. The structure of muscle fascicles was well preserved. Necrosis was absent; some mononuclear cell infiltration could be still

Fig Biopsy from quadriceps muscle showing an unidentified NADH-positive structure with small round clear particles (bar = 50 μ m).

noticed. One basophilic fibre was seen; one fibre showed an unidentified NADH-positive structure filled with small round clear particles (fig). Electronmicroscopically Z-band streaming and some cytoplasmic bodies were seen. No viral inclusions could be detected.

For virus isolation cultures of primary monkey kidney cells, of human diploid fibroblasts and of Hep-2 cells were inoculated with patient's material by standard procedures.¹¹ Inoculated cultures were examined for the development of a cytopathogenic effect for a period of 3 weeks. Isolated viruses were typed with the use of type specific neutralising antisera. The isolate was identified as echovirus 6.

The patient was immediately treated with infusion therapy of 41 NaCl 0.9% per 24 hours and forced diuresis in order to prevent acute renal insufficiency as consequence of myoglobinuria. Urine was kept alkaline by NaHCO_3 1.5% infusion. No signs of renal failure developed. On the 8th day of admission a maculopapulous rash developed, which is very characteristic of a hypersensitive reaction to penicillin in infectious mononucleosis patients. The muscle weakness gradually disappeared within 2 weeks and full recovery took place.

The diagnosis rhabdomyolysis was established at admission, because of tenderness and weakness of both legs, myalgia, dark-brown coloured urine and strongly elevated levels of myoglobin and CK in serum. Adequate therapy¹³ possibly prevented the development of renal insufficiency, a well-known complication of rhabdomyolysis. Infectious mononucleosis was suspected because of fever, lymphadenopathy, atypical lymphocytosis and the characteristic skin rash as a reaction to amoxicillin. An EBV infection was serologically confirmed.

It is not always possible to make a clear distinction between postinfectious ("toxic") rhabdomyolysis and true viral myositis. These diagnostic terms are used synonymously in most case reports about rhabdomyolysis related to viral infection. This probably illustrates the existence of a wide spectrum of muscle pathology extending from a toxic muscle fibre necrosis following febrile disease, to acute inflammatory myositis with rhabdomyolysis due to viral infestation. It would be better, however, to limit the diagnosis viral myositis to those cases in which inflammatory changes in muscle are present and/or virus has been isolated from muscle. Especially in familial cases is a toxic origin most probable.

In the present case it is clear, however, that this previously healthy young man suffered from a true acute viral myositis with mild rhabdomyolysis. Echovirus 6 was isolated from muscle and thought to be the causative agent of rhabdomyolysis. Still the question remains whether any relation exists between echovirus 6 infection and EBV. The contribution of EBV to the pathogenesis of the attack of rhabdomyolysis is unclear. In the literature rhabdomyolysis has been reported to be associated with EBV in four patients in whom the infection was serologically confirmed and rhabdomyolysis was evident from weakness, elevated CK and renal insufficiency.^{7,9,14,15} Only in the patient reported by Schlesinger *et al* was a muscle biopsy carried out, which showed no inflammatory changes.⁷ To our knowledge EBV has never been detected in muscle and any direct causal relationship between mononucleosis and rhabdomyolysis has not yet been documented. In the three patients of Schlesinger *et al*, Kantor *et al*, and Friedman and Libby a postinfectious toxic factor may have played a role in the development of rhabdomyolysis.^{7,9,14} Also in our patient a direct effect of EBV on muscle seems unlikely. One might hypothesise that EBV has altered the patient's immune system and made him more susceptible to the effects of echovirus infection. EBV infection might have made it possible for echovirus 6 to invade directly into muscle tissue and thus to trigger rhabdomyolysis. The same mechanism may have played a part in the dual infection with streptococcus and picornavirus in the case reported by Porter *et al*.³ Also Fukuyama *et al* describe a patient in whom Coxsackie virus infection was determined by serology and picornavirus-like crystals were found in muscle.⁴ These infections might have interacted synergistically.

Our knowledge about this type of muscle disorder could be augmented by carrying out

more frequently histological studies together with isolation of virus in case of postinfectious rhabdomyolysis. A needle biopsy, as in our case, suffices.

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Rheumatoid nodule formation within the lumbar extradural space

Sir: The presence of rheumatoid nodules is the major extra-articular feature of rheumatoid disease, occurring typically within the subcutaneous tissues in relation to the extensor surfaces of the wrist and elbow. Rheumatoid nodule formation within the central nervous system is rare.¹ We report a case of a lumbar extradural rheumatoid nodule giving rise to spinal nerve root compression.

A 58 year old female with a 3 year history of seropositive but quiescent rheumatoid arthritis affecting multiple joints was referred to the Neurosurgical Unit with a 3 month history of left sided sciatica and parasthesiae. Examination revealed typical rheumatoid features in both hands but no subcutaneous nodules were seen. There was flattening of the lumbar lordosis and marked restriction of right lateral flexion and extension. Although the left lower limb tendon reflexes were diminished no focal neurological deficit was noted. A lumbar myelogram and CT of the spine revealed a marked stenosis at the L3/4 disc level with a filling defect most prominent in the left postero-lateral position (fig). A mid-lumbar exploration was performed. Exposure of the dura at this level lead to the extrusion of a small amount of pultaceous material. Within the extradural space there was an encapsulated nodule 2 cm long, lying postero-laterally, and loosely adherent to the dura. The precise origin of the lesion was obscured by fibrous tissue. The mass was removed piecemeal. Histological examination of the lesion showed extensive areas of fibrinoid necrosis, surrounded, in parts, by poorly formed palisades of histiocytes and chronic inflammatory cells. The appearances were those of a rheumatoid nodule. The patient's sciatica resolved after the operation and she made an uneventful recovery. Her rheumatoid arthritis remained quiescent on penicillamine therapy.

Direct involvement of the brain, spinal cord and meninges by rheumatoid nodule formation is very uncommon. Eleven previous cases of rheumatoid nodule formation within the brain and cerebral meninges have been described,¹ but only three previous cases of involvement of the spinal cord and spinal meninges.²⁻⁴ Less than 55% of the cases of rheumatoid nodule formation in the brain and cerebral meninges had clinical signs or symptoms (and of these a proportion could be attributed to other coexistent CNS pathology.¹ By comparison all cases with spinal cord involvement (including our case) had neurological symptoms and signs of spinal nerve root compression. This reflects the anatomical site of the lesion rather than any special feature of spinal nodules. Unlike its cerebral counterpart, the diagnosis of spinal rheumatoid nodule does appear to have some therapeutic significance for in two of the three previous cases (as well as in ours) the lesion was readily amenable to surgery. Excision produced resolution of the patients' symptomatology.

Our case is unusual in that it occurred in a patient with inactive disease of only 3 years duration and apart from the extradural

nodule there were no other extra-articular features. All previous examples of CNS involvement by rheumatoid nodules have occurred in patients with marked synovitis and/or joint deformities. Extra-articular features were also frequent with three patients having ocular involvement, five patients with pulmonary disease and seven with subcutaneous nodules.¹ Unlike our case, previous examples of CNS rheumatoid nodule formation appear to be a feature of chronic disease. The duration of disease varied from 6 to 30 years (although one case with nodules of the falx cerebri and choroid plexus had only a 4 month history of disease).¹

Our case, and the previously reported cases, illustrate the widespread nature of rheumatoid disease, but in this case CNS involvement has occurred in the absence of active disease or other extra-articular manifestations.

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Notice

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Fig Lumbar myelogram showing a postero-lateral filling defect and marked stenosis at the level of the L3/4 disc space.