SUMMARY

Sixteen patients were given prophylactic A.C.T.H. after they had sustained head injuries. Each had shown radiological evidence of a fractured petrous bone and had had bleeding from the ipsilateral ear.

The expected incidence in an untreated group of the same size would have been six patients showing delayed facial weakness.

Only two patients developed delayed facial weakness. In both this occurred on the second day. In one it was mild and lasted only 10 hours; in the other it was complete and denervation occurred despite treatment.

It seems that A.C.T.H. offers some protection against this complication of head injury.

We are grateful to Dr. T. D. A. Hockaday and Mr. George Higgins for their expert help and advice.

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# *Medical Memoranda*

## Sarcoid Myopathy

*Brit. med. J.*, 1967, **4**, 465–466

Only 12 cases (Hinterbuchner and Hinterbuchner, 1964) have been found in the literature where sarcoidosis presented as a pure myopathy without evidence of other systemic involvement. I present another case which has responded dramatically to corticosteroid therapy.

### CASE HISTORY

A 55-year-old Englishwoman presented in March 1966 with a three-week history of calf stiffness alleviated by exercise. She had no other symptoms, was on no drugs, and had no relevant family or social history.

In April she was admitted to hospital, and examination showed severe weakness in her legs, more so on the left, with slight thigh wasting. Weakness was also present in the elbows and wrists, but without wasting. The muscles were non-tender and felt normal. Her gait was stiff and wide-based, and she tired easily. No other significant abnormality was found and no definitive diagnosis made. Physiotherapy produced some improvement, and she was discharged after one month. By August wasting and weakness had developed in all proximal limb muscles. Walking was difficult without support, resulting in several falls.

She was readmitted to hospital in January 1967 with severe weakness in her hands, hardly able to walk even with support, and unable to sit up unaided or do her hair. She had an occasional dry cough but no other complaints. Examination showed obvious muscle wasting of all the limbs, whose tone was slightly reduced. The muscles felt rubbery but were not tender, fasciculating, or showing involuntary movements. Movement was full, but with severe generalized weakness, including the sternomastoids and face. The other systems showed no abnormality.

*Investigations.*—A skeletogram showed mild osteoporosis; W.B.C. 3,900–7,000/cu. mm. (monocytes 8.5–12.5%); serum proteins 6.5–7.2 mg./100 ml. (albumin 3.0–4.4 mg., globulin 2.8–3.5 mg.); normal electrophoresis; latex fixation test weakly positive (Putkonen *et al.*, 1965); urine creatine 460 mg./24 hours (normal 0–50 mg.). Muscle biopsy showed no necrosis or acid-fast bacilli but the characteristics of sarcoidosis—a granulomatous process with small foci of epithelioid cells surrounded by small mononucleated cells and some multinucleated giant cells.

The following investigations showed no abnormality. Blood: haemoglobin, platelets, P.C.V., M.C.H.C., film, L.E. cells, E.S.R., urea, electrolytes, cholesterol, alkaline phosphatase, calcium, phosphorus, liver function, antistreptolysin titre, aldolase, lactic acid dehydrogenase, creatine, creatine phosphokinase, creatinine, lead, culture. Urine: routine analysis, bile derivatives, porphobilinogen, uroporphyrins, Bence Jones protein, calcium, creatinine, renal steroid leucocyte excretion, intravenous pyelogram. Throat swab, culture for tubercle, 1:10 tuberculin test, Wassermann reaction, V.D.R.L., cerebrospinal fluid, electrocardiogram, Kveim test, radioactive iodine uptake.

Treatment was begun in February with prednisone, 15 mg. daily, reducing to 10 mg. daily in June, the only available therapy of value (Crompton and MacDermot, 1961; Silverstein *et al.*, 1965). After three weeks she could sit up unaided, and by April felt much improved and could walk 30 yards (27 metres) with support. Power had improved considerably in her legs and was normal in her arms apart from slight weakness in her right fingers. In June she was walking up to 100 yards (90 metres) with slight support.

Though remissions occur in the sarcoid process, I thought that, since the condition had progressed inexorably for 15 months, corticosteroid therapy had caused rapid improvement.

### COMMENT

The skin lesions of sarcoidosis were first described by Boeck (1899), and literature has amassed covering involvement of nearly all the tissues in the process (Mayock *et al.*, 1963). The diagnosis of sarcoidosis should always be confirmed by histology (Mayock *et al.*, 1963), usually accompanied by an intermittent monocytosis, eosinophilia, leucopenia, raised serum calcium and globulin, and occasionally a raised alkaline phosphatase. A positive Kveim reaction occurs in 60% of patients, with false-negative results in long-standing or inactive disease (Siltzbach, 1964) and during corticosteroid therapy.

The outcome of sarcoidosis is usually spontaneous remission, but irreversible lung or myocardial fibrosis may cause death. Treatment with A.C.T.H. or corticosteroids may produce remission (Silverstein *et al.*, 1965), but 60% of patients do not respond (Crompton and MacDermot, 1961).

Since Sundelin's (1925) original report several cases of muscular involvement in generalized sarcoidosis have been described, taking several forms—asymptomatic microscopic granulomata, palpable nodules, or myopathic. A purely myopathic presentation is very rare, only 12 cases have been found in the literature (McConkey, 1958; Hinterbuchner and Hinterbuchner, 1964). Muscle biopsy shows characteristic sarcoid histology—interstitial infiltration with giant cells, epithelioid cells, and lymphocytes, with tissue replacement by a pallisade-like and reticular collagen fibre network, leading to fibrosis. Occasionally widespread muscle fibre degeneration

occurs with floccular necrosis and macrophagia or epithelioid tubercles and intramysial multinucleated giant cells (Coërs, 1967).

I am grateful to Dr. T. Grahame-Wilson for allowing me to study this case, and to Professor W. Blackwood, Dr. G. M. Churcher, Dr. H. Greenbergh, and Miss J. K. Bowman for their help and advice.

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# Transfusion Malaria in a Man with Christmas Disease

*Brit. med. J.*, 1967, 4, 466

Malaria as a complication of blood transfusion has been reported in the United Kingdom only four times (Thomas *et al.*, 1936 ; Nabarro and Edward, 1939 ; Rogers, 1947 ; Grant *et al.*, 1960), though it is a relatively common occurrence in countries where malaria has until recently been endemic. The rarity of this complication in England can make diagnosis difficult, as is illustrated in the following case.

## CASE REPORT

An Englishman aged 33 who had never travelled abroad had been admitted to hospital many times since childhood for haemarthroses and haematuria, secondary to Factor IX deficiency. He was first admitted to the Churchill Hospital on 14 April 1966 for the treatment of a haematoma in the arm caused by an intramuscular injection.

Examination showed a pale, anxious man with a large tense haematoma of the deltoid muscle. His haemoglobin was only 47% (6.9 g./100 ml.), though he had been given 3 pints (1.7 litres) of blood during the previous week. Investigation revealed chronic renal disease with raised blood urea and proteinuria ; an intravenous pyelogram later showed small contracted kidneys. Coagulation studies showed Factor IX deficiency.

The deltoid haematoma was treated during the next two weeks by the infusion of three doses of Factor IX concentrate (equivalent to 1.5–3 litres of fresh plasma), three doses of plasma (dose volume 800 ml.), and packed cells from 4 pints (2.3 litres) of blood. The latter was given on 14 and 15 April. Three more doses of plasma were given later for the treatment of haemarthroses.

He was discharged home on 4 June, only to be readmitted on 17 June with a haemarthrosis of the knee of five days' duration. This was treated by immobilization without intravenous therapy.

On 29 June he developed an unexplained irregular fever and at the same time became hypotensive and developed pancytopenia and splenomegaly.

*Investigations from 30 June to 5 August.*—The following tests gave normal results: blood, urine, stool, and throat-swab cultures ; chest *x*-ray examination ; salmonella and brucella agglutinations ; L.E. cell preparations ; one search for malaria parasites ; a sternal marrow puncture ; and cultures of marrow and urine for tuberculosis. During July his anaemia was aggravated by recurrent haematemeses associated with rigors. Thrombocytopenia probably contributed to the gastrointestinal bleeding (platelets 45,000–120,000 cu. mm.). He was transfused with fresh packed cells from 10 pints (5.7 litres) of blood during this time. Meanwhile he developed boils on his buttocks and his blood urea rose from 100 to 186 mg./100 ml. After four weeks of irregular fever the temperature chart began to show a regular periodicity with two clear days between each attack. The character of the fever became strongly suggestive of quartan malaria, and on 5 August parasites of *Plasmodium malariae* were found in the peripheral blood. A review of blood films showed that parasites first appeared in small numbers in a blood film on 5 July, while six previous films between 3 April and 1 June failed to show parasites.

Chloroquine 500 mg. followed by 250 mg. daily for three days and primaquine 15 mg. base daily for 14 days were given. During the writing of this paper I have been informed that malaria transmitted by blood transfusion does not give rise to exoerythrocytic forms of the parasite. Treatment with primaquine was therefore unnecessary (P. G. Shute, 1967).

Blood counts returned to normal, the boils healed, and the blood urea fell to its former level. During the next three weeks five doses of plasma (dose volume 800 ml.) were administered for haemarthroses, which developed during mobilization, and the patient was discharged home on 10 September.

## COMMENT

Investigation of the blood donors in this case revealed one Englishman who had suffered malaria in 1946 while serving with the Army in the Far East. He had been in good health since that time. During prolonged searches of his blood films one young malaria parasite was found. This donor had given 34 pints (19.3 litres) of blood in the past and an attempt was made to discover whether previous recipients of his blood had contracted malaria. A temperature chart suggestive of quartan malaria was found in the records of one recipient 39 days after transfusion. This could not be confirmed on the only available blood film.

In a series of 36 cases of transfusion malaria studied by Lepeš (1965) in Yugoslavia the incubation period varied between 5 and 70 days, the infected blood having been stored 1 to 10 days. The incubation period in the present case was 74 days, the blood having been stored 10 days.

Transfusion malaria should thus be remembered when a patient develops a pyrexia of unknown origin as late as 10 weeks after receiving blood, for in spite of the precautions taken by the National Blood Transfusion Service the occasional donor may be a chronic malaria carrier.

I would like to thank Professor R. G. Macfarlane for encouraging me to publish this case ; Professor P. B. Beeson for helping to make the diagnosis ; Dr. Jean Grant, who investigated the blood donors ; Dr. F. G. Bolton, Mr. P. G. Shute, and Dr. F. E. Lipscomb, who spent much time studying blood films from the patient and donors ; and Dr. W. d'A. Maycock for his helpful advice.

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