NEURALGIC AMYOTROPHY IN REITER'S DISEASE*

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Neuralgic amyotrophy is a form of localized neuropathy affecting one or more nerves and leading to wasting and weakness of the muscles innervated by those nerves. During the second world war involvement of the muscles of the shoulder girdle was reported in young men by Spillane (1943) and Parsonage and Turner (1948) and the condition was sometimes called "shoulder-girdle neuritis". It usually occurred among patients who were in hospital because of an acute infection or for an operation.

The condition presents with pain, followed by muscle wasting and weakness. The commonest muscles to be involved are the spinati, the serratus anterior, the deltoid, and the trapezius. Sensory loss over the distribution of the axillary nerve is said to be common when the deltoid is involved. The condition may be unilateral or bilateral. Recovery is usually slow and in severe cases the atrophy of the muscles may be permanent. The cause is not known.

Involvement of the nervous system in cases of Reiter's disease is a well recognized but apparently rare complication. Several observers have reported individual cases but the true incidence of this type of complication is unknown. In Reiter's disease associated with bacillary dysentery, Wilke (1943) described cases of peripheral neuropathy and of so-called "shoulder-girdle neuritis". Optic neuritis was found in two patients by Zewi (1947), and Lindsay-Rea (1947) observed retrobulbar neuritis in a patient who had suffered recurrent attacks of uveitis. Graby, Hosotte, and Armand (1949) reported polyneuritis with foot drop in a patient with amoebic dysentery and Lafon, Pages, Roux, Temple, and Minvielle (1955) described a patient who developed a transient hemiplegia with full recovery of function.

Neurological complications in Reiter's disease associated with genital infection have been reported even less frequently. Csonka (1958) reported details of a patient who developed various cranial nerve palsies and signs of involvement of the pyramidal tracts, and Oates and Hancock (1959) described three patients, one with so-called "shoulder-girdle neuritis", one with lumbar radiculitis, and one with meningo-encephalitis.

This paper describes a patient who developed neuralgic amyotrophy during the course of severe, prolonged, relapsing Reiter's disease associated with genital infection.

Case History

The patient was a 20-year-old English soldier with no history of previous serious illness. In December, 1961, following an exposure to infection, he developed a urethral discharge, pain and stiffness of the right shoulder, aching in the lower part of the back, and pain in both feet on walking. A few days later his eyes became red and sore and he noticed a rash on the glans penis and prepuce. He was admitted to Tidworth Military Hospital and later transferred to Millbank. A diagnosis of Reiter's disease was made and he was treated with bed rest, sulphanamides, analgesics, and physiotherapy. During his 7½ months in hospital he developed pain and swelling of both ankles and of the left knee. Shortly after leaving hospital he was discharged from the Army but still had pain in several joints and walked with a limp.

In August, 1962, he was referred to the Department of Venereology at the General Infirmary at Leeds. He complained of an intermittent urethral discharge, a rash on the penis, soreness and redness of both eyes, pain and stiffness of the right shoulder, the neck, the lower part of the spine, and of both feet and ankles. There had been further recent exposures to infection.

On examination he was a thin, anxious man with a limp due to pain in both ankles on walking. His temperature was 100°F. (37·8°C.). There was marked bilateral conjunctivitis but no evidence of uveitis. The right shoulder was tender to palpation and its movements were considerably restricted because of pain. The movements of his neck were also restricted by pain. There was tenderness over the right sacro-iliac joint, and flexion and extension of the lumbar spines was limited by discomfort. The metatarsal-phalangeal joints of both feet were red, hot, and very tender to palpation. Movement of the mid-tarsal joints of both feet produced pain and there was

* Paper read at MSSVD meeting in Dublin, May 29-30, 1964.
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bilateral swelling of the sub-tendo Achillis bursa. There was a purulent urethral discharge and marked balanitis cincta with erosion of the inner surface of the prepuce. The rest of the physical examination was normal and the blood-pressure was 130/80 mm. Hg.

Gonococci were demonstrated in a Gram-stained smear of the urethral discharge and were also grown on culture. The urine was hazy due to pus in both glasses of the two-glass test but tests for albumin and sugar were negative. The erythrocyte sedimentation rate (ESR) was 30 mm. in one hour (Westergren), the haemoglobin 84 per cent., the white blood cell count normal, the Rose-Waaler sheep cell agglutination test negative, the serum uric acid 3·3 mg. per cent., and the blood Wassermann and Price's precipitation reaction both negative. Radiographs of the chest, feet, ankles, shoulders, spine, and sacro-iliac joints were normal.

The patient was admitted to hospital and treated with fortified procaine penicillin, 800,000 units intramuscularly in a single injection, rest in bed, and calcium aspirin tablets every six hours. The urethral discharge disappeared in 24 hours but recurred again three days later. Smears and cultures showed many pus cells but no gonococci or other organisms were demonstrated. This non-specific urethritis was treated with tetracycline, 250 mg. six-hourly for seven days. The discharge did not respond to the treatment. Two weeks later the right knee, right wrist, and the proximal interphalangeal joints of the right hand became tender and swollen. The pain in the right shoulder increased in severity and movements were very restricted. There was pain in the thoracic area of the spine and pain over the upper right costo-chondral junctions. There was a large effusion in the right knee and tenderness on palpation over the inter-spinous ligaments of T2 to 4 and T11 and 12. Superficial erosions of the mucous membranes of the mouth and tongue were discovered and shortly afterwards purulent and crusted lesions of keratoderma blennorrhagica appeared on the right heel. The ESR was now 110 mm. in one hour. Treatment with bed rest, phenylbutazone (200 mg. three times daily), calcium aspirin (two tablets six-hourly), and gentle active and passive movements of the joints did not produce any improvement. The pain in the right shoulder became increasingly severe and movements of the joint were very limited due to weakness and pain. Three weeks later marked wasting of the muscles of the right shoulder girdle with fasciculation was noticed. The wasting was most marked in the supraspinatus, infraspinatus, deltoid, and trapezius muscles (Fig. 1), but all the muscles of the shoulder girdle were involved as well as those of the upper arm and forearm. Measurements showed ½ in. of wasting of the upper arm and ⅛ in. of wasting of the forearm on the right as compared with the left arm. Sensory testing revealed an area of anaesthesia (Fig. 2) to pin prick and light touch measuring 4 in. by 2 in. on the lateral aspect of the right upper arm in the region of the insertion of the deltoid muscle in the distribution of the axillary nerve (C5 and 6). Early morning smears and cultures showed the non-specific urethritis to be still present. The balanitis became scaly and psoriasiform in type and fresh lesions of keratoderma blennorrhagica appeared on the feet and on the chest.

Electromyographical studies on the wasted muscles showed significant fibrillation activity at rest and a
decreased jerky pattern of motor unit activity on voluntary contraction in the right infraspinatus and deltoid muscles. These changes were considered to be in keeping with a diagnosis of neuralgic amyotrophy. Clinical assessment by a neurologist (Dr Deryck Taverner) confirmed that the physical signs were compatible with neuralgic amyotrophy.

Because of the unsatisfactory response to the usual remedies steroid hormones were administered. Prednisone, 15 mg. daily, produced no improvement and the dose was increased to 30 mg. daily. By this time the patient had lost 3 st. (19.1 kg.) in weight since his illness began and had run an intermittent fever for several months. There was a normocytic, normochromic anaemia with a haemoglobin of 70 per cent. Radiographs showed osteoporosis of the right wrist and right shoulder. Tests of the cerebrospinal fluid were normal.

Immediately following the increase in the dose of prednisone the left knee became swollen and an effusion developed in the joint cavity. The arthritis of the small joints of the hands and the feet worsened, and fresh lesions of keratoderma blennorrhagica appeared on the soles of the feet and on the chest. Gradually, however, during the ensuing weeks the condition of the joints improved and the lesions of keratoderma blennorrhagica decreased in size and eventually disappeared. The pain in the region of the right shoulder slowly improved and the muscles of the right shoulder girdle gradually returned to normal with exercises. The area of cutaneous anaesthesia decreased in size and eventually disappeared. Nine months after starting treatment with prednisone the patient was free from symptoms and there were no abnormal physical signs. The evidence of urethritis had disappeared, but tests on the prostatic secretions still showed pus to be present. The prednisone was slowly withdrawn over a period of several months, and one year after stopping all treatment the patient was quite well, there was no evidence of arthritis, muscle wasting, or cutaneous sensory loss. Tests of the prostatic secretions were now normal. The patient was attending a rehabilitation centre and was able to play football.

Discussion

Neuralgic amyotrophy is frequently precipitated by a febrile illness and appears to be more common in young men than in women. Although the cause is unknown, Hughes (1944) has suggested that a virus may be responsible for some cases. The cause of Reiter's disease is also unknown but there is some evidence that it could be due to a virus. If this were proved to be the case, it is possible that the two conditions in our patient might have been caused by the same virus. An alternative suggestion as to the cause of neuralgic amyotrophy has been put forward by Miller and Stanton (1954), who suggested that the condition might be due to hypersensitivity and considered that there were many similarities with cases which followed serum injections and prophylactic inoculation.

Our patient had severe, chronic, relapsing Reiter's disease with a prolonged episode of fever which did not respond to the usual remedies of tetracycline and phenylbutazone. However, the arthritis, the keratoderma blennorrhagica, the circinate balanitis, the urethritis, and the neurological symptoms and signs all improved and eventually disappeared after the administration of prednisone. There were no residual abnormalities and the patient was healthy when last seen one year after stopping all forms of treatment.

Summary

The history, physical signs, and results of investigations in the case of a patient with chronic, relapsing Reiter's disease, who developed neuralgic amyotrophy, are described.

The condition failed to respond to the usual remedies used in the treatment of Reiter's disease, but eventually disappeared with prednisone.

After studying the literature, we believe that this is only the second recorded case of neuralgic amyotrophy occurring in a patient with Reiter's disease associated with genital infection.

We would like to thank Dr Deryck Taverner for arranging the electromyographical studies and for his interest in this patient.

REFERENCES

Spillane, J. D. (1943). Ibid., 2, 532.

Amyotrophie neuralgique dans la maladie de Reiter

RÉSUMÉ

On présente l’observation d’un malade atteint de maladie de Reiter chronique et récidivante chez qui se développait une amyotrophie neuralgique.

Cette condition ne réagit pas au traitement habituel de la maladie de Reiter mais finit par disparaître grâce à la prednisone.

Après maintes recherches nous pensons que ce cas n’est que le deuxième cas d’amyotrophie neuralgique survenant dans une maladie de Reiter associée à une infection génitale, rapporté dans la littérature médicale.