

# Benign Myalgic Encephalomyelitis

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IN RECENT YEARS outbreaks of the same pattern of neurological illness with myalgia, pareses and sensory phenomena, and quite distinct from poliomyelitis, have been reported in various parts of the world. The *Lancet* (1956) has suggested the term "benign myalgic encephalomyelitis" as a clinically descriptive name for this syndrome which has as yet no known cause or established pathology but which nevertheless appears to be a nosological entity.

## General Clinical Description

There is an early preparetic stage with mild sore throat, headache, backache, lassitude (marked), drowsiness, giddiness, nuchal pain and low pyrexia. A few days later this is followed by the development in varying combinations of myalgia, neck stiffness, diffuse paresis of a limb or limbs, paraesthesiae, areas of altered cutaneous sensation, impairment of joint and vibration sense, diplopia and other cranial nerve disturbances, and difficulties of micturition. The neurological signs tend to fluctuate and paresis may move from limb to limb or relapse. Twitching and 'jumping' of muscles may disturb the patient, fasciculation may be seen and frank spasm can occur. In affected limbs the muscle tone may be normal, flaccid or hypertonic; the tendon reflexes are preserved and often normal but may be diminished (especially at first) or increased (especially later). The plantar responses have been variously reported as flexor, unobtainable, equivocal or extensor; voluntary movement is often curiously jerky and preceded by a characteristic delay in response to volition; cutaneous sensory changes do not generally correspond to peripheral nerve or spinal segmental areas; there is no proportionate wasting.

The cerebrospinal fluid is usually normal. The blood picture is little affected, but there may be a tendency to neutropenia and occasional atypical lymphocytes are to be seen. Numerous attempts to find a causal agent, especially viral,

have failed. The electromyogram on attempted voluntary movement commonly shows a myopathic pattern and also grouping of motor unit potentials; nerve conduction is normal. Apart from the electromyogram and the qualified hint given by the normal cerebrospinal fluid, diagnosis is purely clinical. Although the paresis even if extensive tends to show marked improvement in a matter of weeks (in general much faster than in extensive poliomyelitis) a proportion of cases relapse or have persisting, usually minor, disability often associated with emotional lability and fatigue. Full details of the clinical picture are to be found in the account given by Dimsdale (1957).

Whilst it may be difficult to be sure that a sporadic example of the condition is not in fact mild poliomyelitis, many are typical of the syndrome. Such was a case, summarized below, which was admitted to hospital during a high local incidence of poliomyelitis in 1957 (Galpine, 1957).

This patient, a woman of 29, had a history of backache, general aches and extreme lassitude for 4 days followed by aching and a feeling of instability in her legs, worse after exertion. The family doctor found a temperature of 99° F. and marked general weakness in her left leg with normal reflexes. In this leg there was diffuse tenderness in the muscles and a tendency to spasm of the hamstrings. The patient also complained of 'flickering' sensations in the limb. The left heel could just be lifted clear of the bed but only after a pause for effort and concentration. Blunting of sensation to touch and pain and to vibration and position was found in the left ankle and foot. There was slight weakness in the left upper arm on testing against resistance, with aching on exertion. The cerebrospinal fluid was normal. The patient improved steadily and when discharged home after 2½ weeks in hospital, sensation appeared normal and she was ambulant although dragging her left leg if she hurried.

### Reports of Outbreaks

In some outbreaks the condition has been found in the general population but the most striking of these have occurred in nurses' homes. As long ago as the summer of 1934 an epidemic diagnosed as poliomyelitis affected 198 of the personnel of the Los Angeles General Hospital (Public Health Bulletin No. 240, 1934) with paresis in 80 per cent, rarely with proportionate wasting. Sensory disturbances were often more pronounced than motor and there was minimal pyrexia and a usually normal cerebrospinal fluid. Contact with patients was blamed for the outbreak. Though hysteria was admitted to be an important factor in some cases, it appeared extremely unlikely that many of the cases were purely hysterical.

The first of the more recent reports describes an outbreak of a disease simulating poliomyelitis in Iceland in the late fall of 1948 (Sigurdsson and others, 1950). It was mainly in Akureyri on the northern coast where it affected 465 persons (6.7 per cent). Paresis was observed in 28 per cent, and in some cases was severe and hardly distinguishable from true poliomyelitis. The cumulative clinical picture, however, differed from poliomyelitis as did the epidemiology with the unusually high morbidity rate, numerous multiple family cases, maximum incidence at age 15-19, female preponderance and lack of any mortality. Personal contact appeared responsible for transmission and the incubation period seemed to be from 5 to 10 days. Of 5 cerebrospinal fluids examined in Akureyri, 2 showed pleocytosis and the others gave normal findings. Attempts to isolate or to find serological evidence of a virus were negative.

A hysterical factor was allowed for in some cases, but this was not considered to cause gross distortion or confusion of the general clinical picture. Complaints of nervous instability and irritability were frequent. Seven to 10 months later, 28 per cent still showed some paresis and 21 per cent sensory disturbance. Sigurdsson and Gudmundsson (1956) following up 39 of the Akureyri cases 6 years later found that 18 still had objective neurological signs and a high number muscle tenderness and subjective complaints. It was surmised that either a poliomyelitis virus of unusual pathogenic properties and of low virulence or an unknown neurotropic virus had been responsible.

The following year, 1949, witnessed an outbreak of illness in Adelaide during the Australian winter. Diffuse muscular weakness was common,

but recovery was excellent and there were no deaths (Pellew, 1951). Fever was trivial. A serious epidemic of true poliomyelitis had broken out earlier in that year and it appears that it became mingled with the more benign illness later in the season. In convalescence there were complaints of muscle pain and depression and difficulty in mental application.

In the late summer of the next year, 1950, an epidemic took place in New York State in which cases were mostly labelled as poliomyelitis, abortive poliomyelitis or 'poliomyelitis suspect'. White and Burch (1954) studied 19 cases in detail (15 women and 4 men) and noted a close similarity to the description of the Icelandic outbreak. Lymphadenopathy was noted as in the Royal Free outbreaks mentioned later. Here again was muscle tenderness and aching, paresis unaccompanied by much change in deep reflexes or by wasting, occasional sensory loss and a tendency to mental depression. Two only of 11 cerebrospinal fluids showed a (low) pleocytosis. It was concluded that the condition, though resembling it, was not poliomyelitis.

In England in the late summer of 1952, 14 nurses resident at the Middlesex Hospital were affected by an illness designated as encephalomyelitis, with severe muscular pain, neck stiffness, a diffuse mild involvement of the central nervous system and a normal cerebrospinal fluid (Acheson, 1954). Extensor plantar responses in over half the cases and frequency of brain-stem signs were features of the outbreak. Although Type III poliomyelitis virus was isolated from the stools of 1 out of 4 patients so tested, the evidence was insufficient to confirm an aetiological role for it. The view was expressed either that the characteristics of poliomyelitis are changing or the outbreak was due to an unidentified neurotropic agent or agents epidemiologically linked with, but less virulent than, poliomyelitis.

In Coventry a year later in the late summer and autumn of 1953, 13 out of 49 staff employed on wards to which cases of poliomyelitis were being admitted, became ill with a condition at first thought to be poliomyelitis (Macrae and Galpine, 1954). In 12 of the patients limb paresis was found, but scrutiny of some cases and of the outbreak as a whole showed divergencies from poliomyelitis of the kind already described. Electromyograms obtained in 3 patients showed a discrete pattern of motor potentials with some large polyphasic on attempted voluntary movement (Shulman,

1953). Virus investigations undertaken primarily to exclude or confirm poliomyelitis were negative. In that season in Coventry increasing numbers of 'poliomyelitis' patients, a high proportion female, were admitted suffering from mild paralysis with early recovery and having a normal cerebrospinal fluid. It was considered that the staff illness resulted from personal contact with the patients and to a lesser extent among themselves.

In South Africa, early in 1955 and towards the end of a poliomyelitis epidemic in the general community, an outbreak of over 90 cases of parietic illness affected the nursing staff of the Addington Hospital, Durban (Hill, 1955). Affected muscles were described as extremely painful, tender and of a rubbery consistency. Paresis, initially flaccid, tended to progress to hypertonicity and early depression of reflexes to exaggeration. When tested against resistance the muscles contracted in a 'curious interrupted clonic-like fashion'. Areas of hyperaesthesia not conforming to any anatomical pattern were found. A number of similar cases were admitted to hospital from the Durban area. Investigations which were continuing at the time of the report referred to here were all negative for antibody, viral and toxicological tests.

The most dramatic outbreak in this country was that in the latter half of 1955, especially July, August and September in the Royal Free Hospital Teaching Group. Out of a total population of about 3,500 (staff, patients and medical students) some 300 experienced an illness described by Crowley and others (1957) as a lympho-reticular encephalomyelopathy. Membership of the nursing staff and residence in the hospital were special, though far from exclusive, qualifications for attack. Instances of presumable case to case infection were recorded from the outset with an incubation, in the main, of 5 or 6 days; in some cases the existence of symptomless carriers was presumed.

The diversity of irritative or depressant disturbances in the central nervous system gave rise to differing views on pathology, but opinion in the hospital was united in postulating an infective origin in spite of the failure to find an infectious agent. More than half the cases showed lymphoreticular disturbance with tenderness and occasional enlargement of lymph glands, liver and spleen; morphological abnormalities also occurred in the circulating leucocytes. These were consistent with those described in infective and also allergic diseases.

Several cases with positive Paul-Bunnell reactions were not, on that account, excluded and reference was made to the view that infectious mononucleosis may essentially be a form of hypersensitivity excited by reinfection.

Abnormal electromyograms were found and described by Richardson (1956) as essentially showing an abnormality in the recruitment of motor units. Instead of contractions beginning with small units in normal numbers, maximum volitional effort showed a reduction of motor unit potentials with initial ones of quite long duration, generally polyphasic and tending to group, producing a tremulous contraction which rapidly fatigued. This resembled the findings in the so-called myelopathic lesion (Bauwens, 1955) with involvement of the motor unit at the level of the cord as in poliomyelitis and syringomyelia — in which diseases prolonged paralysis also produces signs of lower motor neurone degeneration, a finding absent in the Royal Free cases in 27 out of 28 tested. The peculiar grouping of potentials has not been described before and its origin is obscure. In a further paper dealing with this outbreak (medical staff of the Royal Free Hospital, 1957) some advantage is conceded to the term benign myalgic encephalomyelitis but criticism is made not only of the adjective 'benign' as misleading in view of the possible severity and permanency of the neurological manifestations, but also of the failure of the term to indicate lymphoreticular involvement.

In the same summer as the Royal Free Hospital staff outbreak, similar cases were admitted to the Infectious Diseases Department, Royal Free Hospital, 8 of which have been described by Ramsay and O'Sullivan (1956). Electromyograms resembled those obtained in the Royal Free staff outbreak. Five patients showing behaviour disorder were electroencephalographed and all showed abnormalities, though non-specific and possibly constitutional. None of the patients had been in contact as far as was known with any of the affected Royal Free Hospital staff. Five of the 8 had some residual disability when seen 8 or 9 months later. A further small outbreak of 'acute infective encephalomyelitis' occurred in the Preliminary Training School, Royal Free Hospital, in May and June, 1956 (Geffen and Tracy, 1957). The 8 staff affected had periods off duty of from 2 weeks to 5 months.

In Florida in the spring of 1956 an outbreak of encephalomyelitis occurred in Punta Garda.

(Bond, 1956). This followed a similar outbreak in Tallahassee in 1954 variously known as 'Housewives' Polio or 'Tallahassee Strain Polio'. Bond says there was little true paralysis but restriction of movement was often due to pain, sensory disturbances and incoordination. The illness mainly selected adult females in the white population, the ratio being almost 3 females to 1 male and the average age 32. Bond considers these outbreaks sufficiently similar to the English reports to be classed with them under their title of benign myalgic encephalomyelitis.

In the summer of 1956, after an apparent absence locally of 3 years, 7 sporadic cases of benign myalgic encephalomyelitis were admitted to the infectious diseases unit in Coventry (Galpine and Brady, 1957). One patient had had a similar attack in 1953 at the time of the outbreak in the nursing staff. Electromyography gave in all cases an abnormal pattern with grouping of action potentials. Electroencephalography was done in 3 cases and in 1 of them showed minor irregularities similar to those mentioned by Ramsay and O'Sullivan (1956). One case (of at least 2 months' standing and probably in a phase of relapse) had the blood picture and antibody reaction of glandular fever. Again in the summer of 1957, 3 cases, all adult females, were admitted in 2 instances as ? poliomyelitis and in 1 as ? meningitis.

### Conclusion

The broadly similar reports from widely separate parts of the world, summarized above, provide a considerable volume of clinical and epidemiological evidence in favour of the existence of a separate infectious entity to which the clinically descriptive term 'benign myalgic encephalomyelitis' can be applied. It is variably characterized by minor symptoms of encephalitis, lesions of cranial nerves, myalgia, paresis and cramp, and emotional disturbances; though the outcome is relatively benign, some cases have a prolonged course with relapses and persisting, usually minor, disability.

It is natural that some outbreaks of benign myalgic encephalomyelitis should have been mistaken for poliomyelitis. The seasonal incidence of the two diseases is similar, outbreaks of both have occasionally arisen in the general population in the same season and in both, paralysis is often a presenting feature. Moreover, it is difficult when faced with a new thing to get away from accepted disease concepts

even if the latter do not seem wholly appropriate. The usually normal cerebrospinal fluid in benign myalgic encephalomyelitis is a clue which will remain hidden if lumbar puncture is omitted whenever acute paralytic disease prompts the assumption of poliomyelitis.

So far as experience in Coventry is concerned, it was the bizarre outbreak of myalgic encephalomyelitis in the nursing staff that focused attention on its existence in the area. After this it was easy to see that similar cases had been admitted from the general population in the same season. Other conditions that may need differentiation are 'poliomyelitis with functional overlay', hysteria, pyrexia of unknown origin, meningitis, rheumatism and polyneuritis. The suspicion of hysteria, especially in sporadic cases of benign myalgic encephalomyelitis, may be aroused by finding, for example, monoplegia without spasticity along with active tendon reflexes and no wasting, possibly associated with cutaneous sensory loss unrelated to segmental or peripheral nerve areas as well as emotional instability and fatigability—a situation neatly epitomized in the phrase "organically determined hysteria" (Ramsay, 1956). The peculiar nature of the motor weakness has received attention in the account given by the medical staff of the Royal Free Hospital (1957) who point out that the occurrence of hemiplegia without spasticity due to cortical or subcortical lesions has long been recognized. In difficult cases the electromyogram may give valuable evidence by confirming organic disturbance.

In conclusion, although the nature of the lesions is hypothetical and the cause unknown, benign myalgic encephalomyelitis remains a clinical reality to be reckoned with, especially in its favourite guise of '? poliomyelitis'.

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