RECURRENT POLYNEURITIS IN PREGNANCY AND THE PUERPERIUM AFFECTING THREE MEMBERS OF A FAMILY*

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RECURRENT polyneuritis is not a common condition if one excepts cases due to known toxins such as alcohol and lead.

According to Wilfred Harris,¹ Grocco in 1885 was the first to describe recurrent attacks of polyneuritis in the absence of obvious cause. Eichhorst² and also Mary Sherwood³ described two cases from Eichhorst's clinic, each in men; in one both legs were affected, while in the other only the right arm suffered. Targowla's⁴ case was that of a single woman who had three attacks of paralysis at the ages of 19, 27, and 39 years; although in many ways similar to those described here (Case III), the attacks were unrelated to pregnancy and the paralysis was more extensive, involving both lower and upper limbs and various cranial nerves. Sörgo⁵ recounted the case of a man in whom recurrent polyneuritis was associated with attacks of abdominal pain; and in Thomas'⁶ case the subject was a man who suffered also from chronic indigestion. Rabinowitz⁷ recorded the occurrence of multiple neuritis in a girl 12 years of age, who had had a similar attack at the age of nine. Holmes⁸ mentions three cases observed by himself in one of which the attacks occurred in three successive springs, and cites the seasonal incidence in one of Thomas' cases, where the attacks recurred each June for five years. Bingel⁹ recounted the case of a woman of 30, in whom two relapses of polyneuritis occurred, accompanied in each instance by a recurrence of alopecia. Death occurred from respiratory paralysis. Hoestermann¹⁰ described three cases of recurrent polyneuritis, all in adult males, in one of whom thickening of the nerve trunks was noted. Natrass¹¹ recorded the occurrence of three attacks of multiple neuritis in a boy in whom various peripheral nerves were greatly hypertrophied. Harris and Newcomb¹² described relapsing interstitial hypertrophic polyneuritis in a man of 52, who died in the sixth attack.

The occurrence of polyneuritis in several members of a family is rare. The progressive hypertrophic interstitial polyneuritis of Dejerine and Sottas¹³ was familial in four pairs out of 17 cases reviewed by de Bruyn and Stern.¹⁴ Barker and Estes¹⁵ have described a family hæmatoporphyrinuria sometimes associated with polyneuritis. The two sisters described by Ross

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and Bury¹⁶ appear to be the only recorded instance of *recurrent* polyneuritis affecting more than one member of a family.

Polyneuritis associated with pregnancy or the puerperium has been described, apart from cases due to sepsis or local injury. Farani¹⁷ described a case of polyneuritis beginning in the eighth month of pregnancy, and stated that interruption of pregnancy was sometimes indicated in such circumstances. Albeck¹⁸ recorded four examples in 1926, and later¹⁹ described the symptoms of polyneuritis gravidarum on the basis of nine observed cases. Severe vomiting, a tendency to delirium, marked psychoses, and scanty diuresis appeared to indicate a toxic origin for the polyneuritis. Similar cases have been described by Mills,²⁰ Ledoux,²¹ Bierring,²² and Bode.²³ In the example recorded by Brugi,²⁴ on the other hand, there were apparently no signs of severe intoxication. In the case described by Ormerod²⁵ polyneuritis affected chiefly the flexors of the hips on four occasions, always at the fifth month of pregnancy. Pákozdy²⁶ recorded the gradual painless onset of paralysis of the fingers and elbows and later of the legs, in a woman five months pregnant. Recovery followed the interruption of pregnancy, but three post-partum relapses occurred, the first three years later, and the others at intervals of 18 months.

According to Harris,¹ puerperal polyneuritis is usually not a painful form, numbness and paræsthesiæ predominating, with weakness and loss of knee-jerks. The same author²⁷ describes an example of this condition commencing 10 weeks after birth, and in the absence of sepsis. Paralysis affected first the legs, with some sensory loss, then the extensors of the fingers, and later the face and diaphragm, with complete recovery at the end of four months. Batten²⁸ observed a case in which polyneuritis affecting the hands and the legs with loss of deep reflexes recurred after two confinements. I have been unable to find in the literature any record of polyneuritis, recurrent or otherwise, associated with pregnancy, which has affected more than one member of a family.

The condition described below is of particular interest, in that attacks of polyneuritis of irregular distribution have affected a mother and two daughters at different times over a period of 20 years, and have shown a marked tendency to appear or recur during pregnancy and parturition.

PERSONAL CASES

Case I.—M. W., age 60 years, was healthy until the age of 26 years, when she suffered for about two weeks from 'rheumatic' pains in the left forearm, without loss of power. She married at the age of 22 and had five pregnancies. The first child was N. M., described below (Case III); the second, a female child, died from unknown cause at the age of one year; the third was T. C. (Case II); the fourth is a healthy man, age 25. The fifth pregnancy, 21 years ago, came to full term and a difficult labour terminated with the birth of twins, one living five days and the other five weeks. For three months before the twins were born she suffered very severe pains in the left arm. A few days after parturition she noticed loss of power in this arm, with a complete wrist-drop.

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The wrist remained useless for two or three years, and is still weak. There have never been any paræsthesiæ.

Examination on April 4, 1930, showed a partial wrist-drop on the left side, with weakness and slight wasting of the extensors of the wrist and fingers (fig. 1). The left supinator jerk was absent, but there was no sensory loss. No other abnormal signs were found on examination of the nervous system or elsewhere. Skiagrams of the neck showed no evidence of cervical rib.

Case II.—T. C., a married woman, age 30 years, was first affected in 1924, when, at the

FIG. 1.—The patient M. W. (Case I) illustrating her inability to dorsiflex the left wrist completely (April, 1930).

age of 20, she complained of pain in the left shoulder, which lasted for a few months, travelled down the arm as far as the elbow, and then went away, leaving no weakness of the limb. Up to this time she had been in good health, except for severe headaches, worse in the morning on waking, and accompanied by dizziness and nausea, which began at the age of 17. She remained well until November, 1926, three months before the birth of her first child, when the pain returned to the left shoulder. The pain began gradually, and worked down to the fingers; it remained as a constant sharp pain, day and night, until the birth of the child. Weakness of the left arm developed gradually during this time. In February, 1927, two nights after parturition, which was normal, she was seized with severe pain in the right arm, and suddenly lost the use of

the limb. Thereafter the pains continued with diminished intensity, and the power of the arms improved slightly.

When admitted to the Royal Victoria Infirmary on July 1, 1927, there was wasting of both deltoid muscles, the thenar eminences (abductor pollicis brevis) and of the first interosseous space on each side. On the left side there were, in addition, weakness and wasting of the trapezius, supra- and infraspinati, the biceps and the triceps muscles. On faradic stimulation only the right deltoideus and left infraspinatus

FIG. 2.—The patient T. C. (Case II), showing the area of anæsthesia, enclosed in dotted lines. The right hand has been supinated to show the flattening of the thenar eminence. There is no wrist-drop (April, 1930).

failed to respond. The hand-grip was weak on both sides. The supinator and biceps jerks were absent, but the triceps jerks were active. Sensibility to intermediate degrees of temperature, and to pain, touch and pin-prick was lost over an area along the medial side of both forearms, corresponding to the distribution of the first thoracic nerve. Similar anæsthesia, in this case accompanied by a feeling of numbness, was present over a triangular area on the dorsum of the hands, bounded in front by the roots of the index and middle fingers. Examination of the rest of the nervous system showed no abnormality. No cervical ribs were found on X-ray examination. She was a thin, somewhat sallow woman, but examination of the heart, lungs, abdomen, etc., was negative, and the hæmatological findings were within normal limits. No free

hydrochloric acid was found in the resting juice, but 0.033 per cent. was present after histamine stimulation.

She was discharged from the ward on August 6, 1927, but continued to attend for massage and ultra-violet light treatment for another ten months, during which time there was a gradual increase of power in the limbs.

The patient returned for reexamination in April, 1930. She had remained well for the past two years, except for the area of numbness and hypoesthesia on the dorsum of the hands (fig. 2). Elsewhere sensation was normal. No muscular wasting remained except for a slight degree in the left supraspinatus and in both thenar eminences, and the power of all muscles was good. The supinator, biceps and triceps jerks were not obtained.

No more was seen of her until August 6, 1932. In June she had had acute appendicitis, for which appendectomy was performed. The morning headaches, which had occurred almost daily since the birth of her child five years ago, had become worse during the last two years. At times she experienced a little difficulty in raising the arms above the shoulders, and some aching in the deltoid regions. During very cold weather the hands sometimes went 'dead,' the fingers becoming flexed, so that she was unable to straighten them. The area of numbness on the dorsum of the hands remained, and was accompanied by anesthesia to light touch, and by hypoesthesia to pin-prick and to moderate degrees of heat and cold. The degree of hypoesthesia was less on the left than on the right side. The area of anesthesia to light touch on both hands was greater than at the previous examination, and overlapped the bases of all four fingers. Apart from slight flattening of the right deltoideus, and of both thenar eminences, there was no muscular wasting, and all movements were well performed. Faradic response was good in all muscles.

Her condition at the present time (April, 1933) is unchanged, so that in the absence of further pregnancies she has remained free from relapse for five years.

Case III.—N. M., a married woman, 38 years of age, had always been healthy except for rheumatic fever at the age of six, and occasional headaches before the menstrual periods.

On October 1, 1929, she complained of pain over the spine of the right scapula, which was severe, constant and worse at night. During the ensuing week the pains travelled to the radial side of the right forearm, and to the dorsum of the wrist and three middle fingers. The fingers began to curl up from weakness of the extensor muscles and the hand-grip weakened. During the second week she developed a complete wrist-drop, with loss of grip and an almost complete inability to move the fingers. The pains then markedly diminished. There was no history to suggest pressure injury, or exposure to cold; she attributed the wrist-drop partly to the fact that at this time she was using the right wrist very much in her work of numbering articles, etc.

During the fourth week (on October 23) she was seen at the out-patient department, where the condition was diagnosed as a musculospiral paralysis due to neuritis or some pressure injury. Massage and movement were commenced, and the wrist supported by a splint.

At the sixth week intermittent pain was felt in the opposite shoulder, again in the region of the spine of the scapula. There were occasional sharp shooting pains in the knees, with some weakness in the legs.

On January 8, 1930, 14 weeks from the onset, pains were still present in the left shoulder and arm. There was no improvement in the power of the right forearm. It was clear that the paralysis was not confined to the distribution of the musculospiral nerve, as she was unable to separate the fingers, or to oppose or adduct the thumb. The supinator jerk was absent on the right side, but the deep reflexes of the upper and lower extremities were otherwise normal. The abdominal reflexes were

brisk on the right side ; on the left side the reflex was diminished in the upper quadrant and absent in the lower. The plantar reflex was flexor on the right side, and doubtful on the left. There were no other positive findings in the examination of the limbs, and the optic fundi and cranial nerves were normal.

Reaction of degeneration was present in the muscles supplied by the right musculospiral nerve, except in the triceps and supinator longus. A good galvanic response was obtained. Other muscles responded normally to faradism. Skiagrams revealed no cervical ribs and no abnormality of the lower cervical or upper dorsal vertebræ.

FIG. 3.—The patient N. M. (Case III) attempting to dorsiflex both wrists. In addition to drop-wrist on the right side, some flattening of the thenar eminence of the left hand may be seen (April, 1930).

By the end of January there was obvious wasting of the right supinator longus. Early in February the pains increased in the left arm and there developed a feeling of 'pins and needles' in the ring and little fingers, and later in the middle finger, the paræsthesiæ being increased if she put the arm behind her.

She was admitted to the Royal Victoria Infirmary on February 21, 1930, by which time there had been slight improvement in the grip of the right hand. The bowels were constipated, and the appetite poor, and she had lost a little weight. Menstruation was heavy and occurred every three weeks. Examination showed a thin intelligent woman, of sallow complexion, with no abnormality of the heart, lungs or abdomen. The teeth were artificial, the tongue slightly furred. The optic fundi and cranial nerves

were normal. There was wasting of the extensor muscles of the right forearm, with wrist-drop (fig. 3). Power in the interossei and adductor pollicis was now normal, and the hand-grip was fair. The left arm showed no loss of power, but some slight wasting of the thenar eminence. The supinator jerk was absent on the right side, but the reflexes of the upper limbs were otherwise normal. There was quantitative diminution of sensitivity to cotton-wool and pin-prick over the ring and little fingers of the left

FIG. 4.—Shows Case III in June, 1932, after the second attack, with wrist-drop on the left side.

hand. On one occasion, when the pain was severe, the patient was unable to feel a pin-prick in this area.

The abdominal reflexes were diminished on the left side. The lower limbs showed normal power, tone, coordination, and no sensory loss. Knee- and ankle-jerks were brisk and equal. A flexor response was easily obtained on stimulation of the sole of the right foot, but the response was less definite and less easily obtained on the left side.

Cerebrospinal fluid obtained at lumbar puncture was normal with regard to its content of protein, chlorides, sugar and cells. A Wassermann test of the blood was negative. No lead could be detected in the urine. The gastric juice contained a plentiful amount of free hydrochloric acid.

She left hospital on March 25, 1930, but continued to attend for massage and electrical treatment. Pains persisted in the left arm, passing down to the ring and little fingers, for which ultra-violet radiation with a mercury lamp was tried.

With continued massage the right wrist improved so that by the end of May she was able to extend it fully; the fingers remained flexed, however. Both supraspinatus muscles showed slight wasting, but reacted normally to faradic current.

She married in June, 1930. From July to October her chief complaint was of a tightness in the epigastrium, having no relation to food, and associated with slight

FIG. 5.—Illustrates the winging of the right scapula, the drooping of the shoulder, and the wasting of the trapezius, supra- and infraspinatus muscles (Case III, N. M., in June, 1932).

tenderness and rigidity of the right upper rectus. The bowels remained very constipated.

The upper limbs continued to improve, and massage was discontinued in October, 1930. By February, 1931, she felt absolutely well, and remained so until two weeks after the birth of her first child on February 29, 1932.

Because of the paralysis which her sister had suffered following child-birth, the patient decided to go for her confinement to the Princess Mary Maternity Hospital, where she stayed for 11 days. Four days after returning home pains began in the right shoulder blade, passing down the right arm, as in her former attack. Two days

later the left shoulder and arm were similarly affected. Subsequently the left leg felt numb and 'dead,' so that she experienced difficulty in walking. Weakness developed in both arms and progressed so that she could not lift the baby, nor raise her arms above her head. The right arm became numb if the weight of the child was allowed to rest upon it.

On April 4, 1932, she still complained of pains in both arms. Examination showed some weakness in the deltoid muscles, with difficulty in raising the arms laterally above the head, and a compensatory rotation of the scapula. All other movements of the upper limbs were normal and there was no wrist-drop. No wasting was present in the muscles of the shoulder girdle, but the thenar eminences showed slight flattening.

FIG. 6.—Shows 'main-en-griffe' on the left side. There is wasting of both deltoids, of the clavicular fibres of the pectoralis major muscles, of the right biceps, and of both thenar eminences (Case III, N. M., in June, 1932).

The remainder of the neurological examination was negative, and no abnormality in the reflexes was found.

She was given a further course of ultra-violet radiation to the shoulders and arms, and told to rest the upper limbs as far as possible until the pains ceased.

A week later she had developed wrist-drop on the left side, and was having severe pains in the left shoulder. The arm reflexes were active and equal. There was diminished sensitivity to touch and pin-prick in the left thumb. Faradic response was obtainable in the paralyzed muscles of the left forearm, but a very strong current was required. The wrist was splinted in a dorsiflexed position.

By May, 1932, the severe pain in the shoulders had disappeared but there was aching in both arms from shoulder to elbow, sometimes passing to the volar surface of the right wrist. She was still unable to dorsiflex the left wrist. Slight wasting of the thenar eminences was present. Massage was recommenced.

On June 20, 1932, examination showed that although the pains had ceased, the

wasting and weakness of the arms were increasing. Appetite was good, she was not losing weight, and her baby was doing very well. She had difficulty in raising the arms above her head, and wasting was present in the following muscles (figs. 4, 5 and 6): both deltoids, more on the right; the clavicular fibres of the pectoralis major on each side; the right trapezius, supra- and infrapinati, the right scapula being much lower than the left, and the upper angle inclined inwards; the right serratus anterior, with gross winging of the scapula, when the arms were held forward, and to a less extent a similar condition on the left side; the right latissimus dorsi; the right biceps; the left triceps, with inability to extend the elbow completely; the left supinator longus, together with all the muscles on the extensor surface of the left forearm, associated with complete wrist-drop and inability to extend the fingers; both thenar eminences (abductor pollicis brevis), and the left hypothenar eminence. Adduction of the thumb was defective in the left hand. Opposition of fingers and thumb, and movements carried out by the interossei, were not affected. The flexors of the wrists and fingers showed a fair amount of power, but the grip of the left hand was poor. The reaction to faradic current was absent in the following muscles on the left side: serratus magnus, supinator longus, extensor carpi radialis longus and brevis, extensor communis digitorum, extensor pollicis longus and brevis, extensor carpi ulnaris, and abductor pollicis brevis; there was only a very faint faradic response in the extensor indicis. The reaction to galvanic current was delayed, especially in the deltoideus, and was completely absent in the abductor pollicis brevis. On the right side only the serratus magnus failed to respond to faradic current.

Numbness was present over both thenar eminences, but no loss of sensation to light touch or pin-prick could be detected. The skin of the left hand was shiny and atrophic. The deep reflexes were altered as follows: biceps jerk absent on the right side, supinator jerk diminished on the right side and absent on the left. Hoffman's sign and the paradoxical flexor-extensor reflex were negative; and although Wartenberg's sign was a partial positive on the left side the affection of the small muscles of the hand makes it of doubtful value, in this case, as a sign of pyramidal tract involvement. The reflexes of the trunk and lower limbs were normal, and there were no other positive findings in the neurological examination. The movements of the diaphragm, as observed on radiographic examination, were normal.

Her condition at this time bears a striking similarity, in the distribution of muscular wasting, to that found in her sister T. C. in 1927.

From now on there occurred a progressive improvement especially in the power of the right arm. On August 6, 1932, the deltoid muscles showed less wasting, and she was able to raise the arms above the head. Improvement in power and diminution of wasting were noticeable in all the affected muscles, with the exception of the thenar eminences, and of the extensor muscles of the left forearm. There was slight improvement in the ability to extend the fingers, but the wrist-drop remained unchanged. On the right side there was a good response to the faradic current in the serratus magnus, but the rhomboidei reacted poorly, and the biceps gave a negative result with both faradic and galvanic stimulation. On the left side the serratus magnus now reacted neither to faradic nor galvanic current; as on the previous occasion the extensors of the wrist and fingers, with the exception of the abductor pollicis longus, showed no faradic response. Careful testing of the area of numbness on the left thenar eminence showed loss of sensation to light touch (cotton wool), and a diminution to pin-prick and to moderate degrees of heat and cold. A small area of hypoesthesia to light touch was present over the left deltoid muscle; hypoesthesia to pin-prick was present in the central part of this area, but heat and cold were correctly distinguished.

When last examined (April 3, 1933) the wrist-drop had almost recovered, the improvement having occurred chiefly during the past five weeks. There was no longer any sensory loss over the thenar eminences or over the left deltoid muscle.

DISCUSSION

The symptom-complex is that of a recurrent polyneuritis, each attack consisting of three stages :—

1. Severe pains, usually radiating from the spine of the scapula down into one upper limb.
2. The rapid onset of paralysis of lower motor neurone type in groups of muscles of the same limb as the shoulder girdle, sometimes accompanied by sensory loss. The pains remit during this period. Extension of the paralysis, usually accompanied by further pains, may occur during the next few months.
3. Gradual recovery follows during the ensuing 12–18 months, sometimes leaving residual paresis, wasting or sensory loss.

Noteworthy features are the incidence of attacks in three members of a family at long intervals, and the tendency to appear or recur in relation to pregnancy and the puerperium. The paralysis is asymmetrical and frequently widespread, but confined to the upper limbs and shoulder girdles, and not corresponding in its distribution to the areas of supply of peripheral nerves or spinal segments. Sensory loss, when present, is not of glove-and-stocking type, but is confined to small areas of unusual distribution, which may or may not be bilaterally symmetrical, and which may remain after the paralysis has recovered.

Investigations have so far failed to throw any light on the etiology of the condition. The incidence of the disease in three members of a family at different times over a period of 20 years is against the possibility of lead or other extrinsic toxic substance as an etiological factor, nor has there been any evidence of such intoxications. There have been no signs of syphilis or other infection, of pregnancy toxæmia, or of vitamin B deficiency. Examination of the cerebrospinal fluid, gastric contents, blood and urine has revealed no significant abnormality. Hæmatoporphyrinuria has never been a feature of these cases. No cervical ribs and no changes in the vertebral column were visible on radiological examination. There are no nodules to be felt along the peripheral nerves, such as might suggest hypertrophic neuritis.

SUMMARY

A syndrome is described consisting of lower motor neurone paralysis of irregular distribution, preceded and accompanied by sensory disturbances, which affected three members of a family at different times over a period of 20 years and showed a marked tendency to appear or recur after childbirth.

The mother developed unilateral wrist-drop at the age of 39, a few days after the termination of her fifth pregnancy, having suffered from severe pains in the affected arm for three months beforehand. Similar pains had occurred at the age of 26, but without loss of power. Now, 21 years later, there is some residual paralysis of the extensors of the wrist and fingers. The

younger daughter, at the age of 23, suffered from similar pains in one shoulder and arm during the latter months of her first pregnancy, with rapid involvement of the other limb after parturition, and subsequently showed in the upper limbs asymmetrical wasting of peculiar distribution, with localized areas of anæsthesia ; gradual but almost complete recovery followed.

The other daughter, at the age of 29, developed severe pains in the right shoulder and arm, followed soon afterwards by wrist-drop, and paralysis of the small muscles of the hand on the affected side. Pains and localized anæsthesia occurred in the opposite arm. Recovery followed and she married. Two weeks after the birth of her first child pains recommenced in both shoulders with paresis of both deltoid muscles. Wrist-drop again developed, this time on the left side, and was followed by extensive but asymmetrical wasting of both shoulder girdles, and of other muscles of the upper limbs. The subsequent recovery has been almost complete.

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