SEVERE TETANOID SPASM LIMITED TO THE WOUNDED LIMB.

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The record here presented relates to one of a group of cases in which, following upon the receipt of a dirty and sloughing wound, there occurs a condition of continuous and long-continued spasm, associated with violent and tetanus-like contractions of extreme severity, but localized absolutely in distribution to the muscles of the part affected by the wound.

The patient, Pte. H., received on December 2, 1915, a shell wound in the right buttock. He was removed to a field ambulance where he received a dose of antitetanic serum, and thence to a clearing station, being detained in the latter until December 8 on account of abdominal pains and vomiting, suggesting intra-abdominal complications. On the latter date he was removed to the Base, and arrived at No. 11 General Hospital, Boulogne, in the early hours of December 9, 1915.

On arrival the patient was pale and collapsed, and obviously much exhausted by his journey; the temperature was 100.8°F., and the pulse 126, of low tension and somewhat thready. He complained of pain in the pelvis, and was much troubled with vomiting of a greenish bile-stained material. The abdomen was a little distended, but the walls of the abdomen were neither rigid nor tender; whilst from the suprapubic region a slight oedema and discoloration extended into either iliac fossa, suggestive of an extraperitoneal haematoma.

On the right buttock, about 1½ inch below the crest, and the same distance external to the sacro-sciatic notch, was a round wound, about ½ inch in diameter, leading downwards and inwards, with sloughing edges, and exuding the thin, brownish, almost faecal-smelling discharge that one associates with anaerobic wound infections. There was no true pus, and no marked oedema of the buttock. No sign of gas formation could be detected.

A skiagram showed a foreign body, in the form of a smallish piece of shell, lying in the pelvis in the mid-line, and situated at a depth of nine centimetres from the surface of the skin over the sacrum.
From December 8 to 15 the patient gradually recovered from his vomiting and collapse; vomiting ceased on the 11th, and patient took plenty of nourishment from then onwards, but was a good deal troubled by pelvic pains. The wound was frequently irrigated with eusol, but remained somewhat malodorous, although there seemed to be no spread of the infection. The oedema and discoloration of the abdominal wall subsided; the temperature rarely rose above 100°F., whilst the pulse, varying between 108 and 80, became much improved in tone.

On December 15, however, thirteen days after the wound had been received, a new element appeared in the form of an occasional twitching of the muscles of the right thigh, both at the hip and at the knee, not at first very violent nor very frequent, but distinctly painful, especially in the upper and anterior parts of the thigh. During that night the condition became aggravated, restlessness increased and the movements became exaggerated and more painful, causing the patient to cry out.

On the morning of December 16 the whole of the right thigh was being thrown into a condition of spasm, during which the limb remained rigid, whilst violent contractions of the flexors of the hip-joint and the extensors of the knee took place. This general rigidity of the muscles of the thigh soon became continuous, so that the limb assumed a permanent condition of extension at both hip and knee, whilst at very frequent intervals a series of violent and exceedingly painful efforts at hip flexion would ensue, causing the patient to cry out in agony. A general impression given was rather that of a series of repeated and violent struggles around the hip-joint on the part of the flexors, battling against an overwhelming and progressively increasing force, attempting to impose upon the limb a state of continuous and laborious hyper-extension. The hamstrings and the quadriceps extensor were the muscles most involved in this condition of continuous spasm, though the sartorius, gracilis, adductors and glutei were all affected to a lesser extent, and enjoyed no interspasmal relaxation; in fact, this state of ceaseless muscular rigidity was actually to continue in the thigh for some weeks, and to terminate only in a very gradual relaxation. These flexors spasms were very frequent, and handling the part did not appear very greatly to aggravate the already grave condition.

The muscles of the leg and foot were not affected, nor were any other parts of the body involved in the spasms; there were never any signs of trismus nor of dysphagia.
In view of the probably tetanic nature of the convulsions, the patient was anesthetized and antitetanic serum to the extent of three thousand units was administered intrathecally by lumbar puncture. The spasms, however, returned with equal intensity as soon as the effects of the anesthetic had worn off, and morphia was then given in $\frac{1}{4}$-grain doses up to one grain.

On the following day (December 17) the spasms were more frequent, more violent, and more painful, the pain being situated mainly in the anterior muscles about the level of Scarpa's triangle. A further complication arose in the onset of a hemorrhage from the wound necessitating operation. Under anesthesia the wound was opened up and bleeding vessels in the glutei found and secured. The great sciatic nerve, being exposed, was found to be markedly purplish and bruised in appearance, but no gross injury could be detected. The bony edges of the sacro-sciatic notch were found to be broken, and loose portions of the spine and adjacent parts of the ischium were removed. The track into the pelvis was followed up, and the piece of shell lying in the hollow of the sacrum was removed from a cavity filled with clot and foul exudation. This cavity was then washed out with eusol and packed with gauze around a rubber catheter.

The spasms returned with consciousness, but patient was able to take plenty of food shortly after operation. Morphia was given in $\frac{1}{4}$-grain doses up to nearly three grains, whilst chloroform was administered during the worst spasms.

On the 18th and 19th the condition showed no improvement, and on the latter day, on account of the agonizing nature of the pain, a full intrathecal dose of stovaine was administered, without any satisfactory result ensuing, the spasms returning as soon as the effects of the chloroform given had worn off; that is to say, within less than one hour after the injection of the stovaine. A mixture of chloral and bromide was given, but the patient vomited after each dose and it was discontinued in favour of morphia, of which nearly three grains were given hypodermically on the 19th, and a full three grains on the 20th, whilst a slight chloroform anesthesia was repeatedly induced both day and night.

At 3 a.m. on the 21st, the patient became suddenly much worse, the temperature rose from normal to 101°F.; the pulse flew up to 144, and the beat became feeble, irregular, and even intermittent. The colour became dusky, vomiting set in, and patient was in a highly critical condition. He rallied later, however, and the pulse improved during the day, though still maintaining its
irregularity. Large doses of morphia were given daily, combined with administration of chloroform, in spite of which the patient obtained very little real rest, sleeping but in snatches insufficient to compensate for the extreme exhaustion produced by the repeated very severe spasms, and aggravated by a troublesome vomiting which prevented him from obtaining sufficient nourishment. The position of the limb had been becoming more and more extended at the hip, until hyper-extension was now maintained, in spite of the violent attempts on the part of the anterior muscles to obtain flexion.

The vomiting began to subside on the 25th, and the patient was enabled to take more nourishment. Constipation was troublesome, partly due, in all probability, to the morphia, the daily dose of which now averaged $2\frac{1}{2}$ grains.

On the 26th, the spasms began to lose some of their former violence and became rather less frequent; pain was still very great in the upper thigh muscles, and two grains of morphia had to be given on that day. However, from this date onwards until January 3 improvement was steady. Vomiting ceased entirely on the 31st, and constipation was yielding to purgatives. The necessary daily morphia diminished in quantity until it reached $\frac{1}{4}$ grain, on January 2 chloroform being entirely discarded. Spasms ceased on January 2, but the limb remained in a condition of semi-rigid hyper-extension at both the hip and the knee. The temperature was constant between 97° F. and 98° F. since the 25th.

On the evening of the 3rd, a smart haemorrhage occurred from the wound; chloroform was given and the wound packed tightly with gauze, and pressure applied with firm pad and bandage. On the morning of the 4th, however, the haemorrhage having recurred, the wound was once more opened up freely; bleeding vessels in the muscles were secured, and the sciatic artery ligated. The intrapelvic well of the wound was washed out thoroughly with eusol, and some particularly foul-smelling pieces of bone were removed from the ischium in the region of the spine. The sciatic nerve, on exposure, was found to be much less inflamed than during the previous operation, and though slightly discoloured, the former deep purple colour had now gone. The wound was packed with gauze around the catheter as before.

There were no evil effects from the haemorrhage and no recrudescence of spasms arising from the manipulation; and the patient's progress was uninterrupted from that time forward. The pain rapidly disappeared, appetite returned, and the general condition became quite satisfactory.
The state of hypertonus in the limb gradually diminished, and on discharge from hospital the right lower limb was held stiff at the knee, but slight degrees of flexion at the hip were readily obtained. The ankle and the joints of the foot were never affected throughout and remained quite mobile. Babinski's sign was present on the affected side, but no ankle-clonus; the knee was held stiff, and so no jerk could be obtained. The hamstrings and quadriceps, though less tense than formerly, were still rigid; there was still slight hyper-extension at the hip and knee when lying at rest, but a gradual relaxation of the muscles of the thigh was becoming manifest.


REMARKS.

Here, then, was a case in which, upon the thirteenth day after an injury, there commenced a series of twitches and painful spasms such as not infrequently herald the onset of tetanus. Stiffness of the muscles in the neighbourhood of the wound soon followed, accompanied by spasmodic contractions of certain groups in excess of the others, notably in the flexors of the hip-joint and, at first, in the extensors of the knee, though the latter soon assumed a condition of permanent rigidity. The most marked feature was an appearance of complete and continuous stiffness of the limb as a whole, a condition which, actually, was limited to the muscles of the thigh and spread neither below the knee nor to the abdominal wall, whilst the remainder of the body was entirely unaffected. A definite posture was gradually assumed in which the affected limb showed hyper-extension at both hip and knee, lying stiff as a poker, only moving as the spasms occurred, and then to but a slight extent, since the constant extensor influence was sufficient to overcome to a great extent the spasmodic efforts of the flexors. The disease ran an acute course over a period of twelve days, after which a gradual improvement set in: the last spasms occurred on the eighteenth day, leaving a residual rigidity, which was slowly relaxing when the patient left for England a fortnight later.

DIAGNOSIS.

The main question in the diagnosis rests as to whether or no the condition is one of true tetanus localized to one limb. The outstanding features of the case are, firstly, the complete localization of the trouble to the muscles in the immediate neighbourhood of the wound; and, secondly, the extreme severity of the condition.
A number of cases of tetanus have occurred during this War in which the condition has started in an injured limb, has remained localized in that limb for a varying period of time, and has later involved other parts of the body, usually going on to produce trismus, as in a case reported by Captain T. R. Mout [1]. In that instance the onset of the disease was manifested by twitchings in the region of the wound for some twenty-five days, followed by involvement of the muscles of the shoulder girdle on the same side. Four days later the muscles of the opposite shoulder were affected, the head became retracted, and trismus appeared. An amelioration, and later a complete cessation, of the symptoms followed administration of antitetanic serum. Such may be truly described as a case of delayed tetanus, localized, for a time, in one limb.

A second and very similar case occurred in No. 11 General Hospital, under the care of Captain J. Campbell, by whose courtesy I am allowed to publish the following details: The patient was wounded on November 15, 1915, and was admitted to No. 11 General Hospital on November 16, 1915, with multiple wounds of the right thigh and leg, and of the left ankle. On November 25, 1915, he complained of marked pains over the sole of the left foot, and a few twitches were noticed in the muscles of the calf. On the 27th definite spasms ensued, very painful, tonic in character, with almost complete interspasmal relaxation, and involving the muscles of the left leg and also, to a lesser degree, of the left thigh, but accompanied by neither trismus nor dysphagia. Ten thousand units of antitoxin were given intravenously, which afforded slight relief for twenty-four hours. On the 29th, 5,000 units were given subcutaneously, whilst the wound was explored. On the 30th spasms were as bad as ever, and the condition of the ankle necessitated amputation below the knee, 10,000 units again being administered intravenously. On December 1, the spasms were definitely less frequent and had not spread to other parts; but on December 2 the abdominal muscles of the left side had become involved and 3,000 units were given intrathecally. No spasms then occurred for three days, but on December 5 there was a recurrence of severe twitching in the stump, and the patient complained of some discomfort about the jaw, though its movements were quite free. Three thousand units were given once more intrathecally; slight twitches of the stump were noticed on the 6th, but after that day there was no recurrence of the spasms at all.

In both these cases there occurred a condition of spasmodic contractions, starting in one limb, at the site of a wound, and slowly
travelling in a central direction. In the first instance, after three or four weeks of local twitching, the condition slowly spread to the shoulder girdles and thence to the neck and to the jaw, thus disclosing a condition of true tetanus. In the second instance a very similar state of affairs was present—twitching and pain in the foot and calf, spreading to the thigh and finally to the abdomen; administration of antitoxin followed by alleviation, then a few days later a recurrence, a further course of antitoxin, and finally a cessation of the spasms. Here apparently the condition was cut short before it had been able to reach the masseters by repeated intrathecal injections of antitoxin.

Both these cases, however, differ in a marked degree from the case under the writer’s care. There the spasms and rigidity remained entirely localized in the muscles first involved, and were in no way alleviated by intrathecal administration of antitoxin; nor, when the latter was discontinued, did the spasms spread, either upwards to the abdominal walls or downwards to the muscles of the calf or foot. The severity of the condition was unmistakable: very frequent spasms on top of the continuous state of rigidity in the opposing muscles was such as is only seen in the most acute cases of generalized tetanus. There was none of the delay between the onset of twitching and the development of acute spasms, such as was noted in the other two cases.

On the other hand, there have been three cases recorded by Lieut.-Col. Rudolf [2] in which the course of the disease and the definite limitations of the parts affected are almost identical with those of the case here recorded. In each case a wound of the thigh or buttock was followed by a condition of constant rigidity, localized in the muscles of the limb affected, during which violent contractions would occur. In two of the three cases the spasms commenced on the eleventh day after the infliction of the wound, whilst in the third they actually started as soon as the man received his injury. In all three of these cases there occurred the same rapid onset of acute symptoms, the same absolute localization of the spasms, even in the most acute stages, and the same persistent rigidity of the limb over a long period, gradually passing off during convalescence—just as occurred in the case under the writer’s care—and together presenting a clinical picture quite different to that which was seen in the two cases of delayed tetanus in which the symptoms were, for a time, localized in the wounded limb.

On comparing these two groups of cases one is driven to the conclusion that they must represent two quite separate clinical and
pathological conditions. The one group would appear to illustrate a condition of true tetanus, probably modified by a previous prophylactic injection of antitoxin, of gradual onset and slowly advancing, but none the less quite capable of extension as far as the masseters, and culminating, unless freely treated with antitetanic serum, in a typical condition of lockjaw. In the second group, however, the condition never advances beyond the immediate neighbourhood of the wound, and, in its own closely confined area, runs an exceedingly acute course quite unaffected by antitetanic serum; it would be comparable rather to a most violent attack of tetanus absolutely cut off from the rest of the body, unlimited in severity but limited most markedly in distribution. Can this condition be considered as due to tetanus at all when compared with the first group of cases, which do undoubtedly belong to that category? Or might the cause not be found in some other organism, associated probably with anaerobic wound infection and capable of producing a toxin similar to that of true tetanus, but differing in its distribution and methods of spread in the body? It would be of great interest to try and produce such toxin by growing in vitro the various anaerobes found in sloughing wounds, and then, by animal inoculation, to attempt to reproduce the clinical conditions described above.

The question of the spread of the toxin requires elucidation; the possibility of the lymphatic distribution to the muscles affected, and a course then along the specific nerve trunks to definite cornual cells, without power of further intracornual migration, might be considered, in view of the entire freedom of the muscles of the foot and leg in the case where the whole thigh was involved in agonizing spasms.

In three out of the four cases of this type there was apparently a definite incubation period of from ten to twelve days, which closely resembles that most commonly occurring in cases of true tetanus. In the fourth case, however—the first in Colonel Rudolf's series—the spasms started immediately the wound was inflicted, a condition which would coincide neither with tetanus nor with any other wound infection, but which would suggest a direct introduction of the toxin into the wound, with a purely local action, rather than any poisoning of the cornual cells.

As far as treatment goes, in view of the failure of antitetanic serum, one was forced to fall back upon purely symptomatic remedies. Morphia was given in great quantities—twenty-five grains in ten days, and as much as four grains in twenty-five
hours; a toleration for the drug was apparently developed, as no sign of pin-point pupil was ever seen, and no depression of the respiratory centres. Chloroform, as in generalized tetanus, was invaluable, and was administered just as in labour, very little at a time being sufficient to allow of a brief rest, and to give the morphia a chance to produce its effects.

The exhaustion produced by the spasms was so great that the question of myosection of the affected muscles became worthy of serious consideration. Possibly in the future a new antitoxin will be available which will affect this condition of localized spasm, just as the antitetanic serum is able to attack the poison of true tetanus, whether localized in its action to one extremity or generalized over the muscles of the whole body.

Such, then, is a brief account of a condition which might almost be termed "para-tetanus." Rare in the extreme, even by contrast with true tetanus, it presents certain features worthy of description, and appears to merit classification as a definite clinical entity for which, up to the present, no adequate pathological explanation has been forthcoming.

REFERENCES.