Spontaneous motor rhythms of the back and legs in a patient with a complete spinal cord transection

by

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Abstract

Background: Spontaneous activity originating from the spinal cord has been sporadically reported in humans.

Objectives: Investigation of such rhythmic activity of the trunk and legs in a 49 years old male patient who had a complete severance of the spinal cord at the fifth Thoracic vertebra.

Methods: A multi-channel electromyographic (EMG) study was performed together with kinematics measurements obtained from an Optotrak system.

Results: Episodes of rhythmic trunk and lower limb movements started 6-7 years after the spinal lesion, occurred at 2-3 month intervals and continued uninterruptedly for 2-3 days despite continuous delivery of baclofen through an implanted pump.

Several muscles discharged more or less synchronously on both sides but others clearly alternated as, for instance, between hip flexors and knee or ankle extensors. Sensory stimuli (hip position or skin pinch) altered significantly the base line rhythm of about 1 Hz. The patient had both hips injected with corticosteroids and was free of these episodic rhythmic crises for more than 6 months.

Conclusion: It is believed that the rhythmic activity observed in our patient is related to the activation of a spinal pattern generator akin to what has been described in most animal species after complete spinal lesions.
Introduction

Several studies have clearly demonstrated that, after a complete section at mid or low thoracic levels in animals, the spinal cord can generate autonomous and functional locomotor activity of the hindlimbs (1-3). This activity is generally attributed to a spinal Central Pattern Generator (CPG). Over the years, there have also been sporadic reports on rhythmic activity originating from the spinal cord in Man after spinal injury (4-9). Since humans with SCI also show some capacity to regain locomotion after partial SCI (10;11;11-18) and that even complete SCI patients show some rhythmic activity on a treadmill with Body Weight Support (19), it is important to investigate the intrinsic capacity of the spinal cord to generate some organized rhythmic activity.

Here we report on a patient with a complete spinal cord section at T5 and classified as A on the ASIA scale who, several years after his initial lesion, developed, every 2-3 months, continuous rhythmic activity of the lower trunk and legs that would last 2-3 days uninterruptedly, i.e. even during sleep.

Material and Methods

Case History

A 49 year old gentleman sustained a T5-T6 fracture-dislocation spinal injury in a car accident (see Figure 1A) and was classified as ASIA A. Three years later, he was implanted with an intrathecal baclofen pump to diminish his spasticity for a few yeas but developed bouts of continuous rhythmic contractions of the trunk and lower limbs, lasting for 2-3 days often accompanied by severe autonomic dysreflexia. He had an ischiectomy (see Figure 1B) and an ischial flap on the Left side because of pressure sores.
Recording and analyses

1-Electromyography (EMG)

Several muscles (lumbar paraspinal, thoracic paraspinals, above and below the level of the lesion, quadriceps, gastrocnemius and tibialis anterior muscles on both sides) were recorded above and below the spinal lesion with intramuscular electrodes.

Then, the patient was assessed three times with surface EMG electrodes. Two of them were during distinct episodes of spontaneous rhythmic activity (12 months apart). The electrodes were positioned on the erector spinae above (about the level of the spine of the shoulder blade) and below the lesion level, on the right or left side. Abdominal muscles above and below the lesion were also recorded. On both legs, the Gluteus, the Rectus Femoris, the Vastus lateralis, the medial hamstring, the Anterior Tibial and Gastrocnemius muscles were recorded. The EMG were recorded using two NORAXON Telemyo 900 portable integrated EMG multi-channel transmitters (Noraxon, Scottsdale, AZ) and were amplified with an overall gain of 2000 and band-pass filtered (10–500 Hz). The EMG signals were digitized at a sampling frequency of 1200 Hz and stored on a computer using a custom-made Labview program (National Instruments, Austin, TX). The EMG files were converted for analysis with a custom made software (20).

2-Kinematic recordings

Using two synchronised Optotrak motion analysis camera units (model 3020; NDI Technology Inc., Waterloo, Ont., Canada), three dimensional coordinates of three markers placed on each side of the patient (femoral condyle, great trochanter and lateral acromion) were used to record oscillatory movements of the trunk, at a sampling frequency of 60 Hz. Conventional videos were also taken throughout the session for
monitoring purpose. Figure 2A illustrates the position of the optical markers and the EMG electrodes. A typical Optotrak recording of an episode of rhythmic activity is shown in Figure 2B.

3-MRI and X-Rays

Two Magnetic resonance image (MRI) were performed and one is illustrated in Fig. 1A. Based on other work (8) showing a frequent association of myoclonus with hip pathologies, a bilateral hip X-Ray was taken to evaluate the state of the coxo-femoral joints (Fig.1B).

Results

Several questioning sessions did not reveal any obvious trigger for these crises. We could not find any correlation between the periodicity of the baclofen pump filling and crises.

Movement and Electromyographic recordings

Intramuscular recordings of paraspinal muscles above and below the lesion showed that the rhythmic activity occurred only below the level of the lesion.

Figure 2 B shows the trunk movements (mean frequency of 0.9 Hz) in the sagittal plane with the patient in a resting position for a session performed during a crisis. The maximal excursion, although variable, was under 3\(^{\circ}\).

Figure 3 shows a robust and sustained rhythmic activity in all muscles. However, the rhythm periodically slows down for a few cycles (see 2 cycles in the middle of the traces) and starts again at the same frequency. The overall average period of 36 cycles
(including the 19 cycles shown here) was 1082 ms (0.92 Hz) ±S.D. 284 ms (coefficient of variation of 26%).

Besides the obvious largely synchronous discharge in most muscles, a close examination of Figure 3 also shows that the coupling between the bursts of discharges of various muscles could vary with time. For instance, at the beginning of the record, both Tibialis Anterior muscles are out of phase. However, they gradually become synchronous for most of that recording. At the beginning of the sequence, the Left ankle extensor Gastrocnemius is out of phase with the ankle flexor Tibialis Anterior but then both become synchronous. However, the most clear cut alternation was between the Left Rectus Femoris (hip flexor) and Vastus Lateralis, a knee extensor. This antagonist discharge is sustained throughout the episode (Fig. 4, A and B). In ankle muscles, averages show intermediate bumps suggesting some instability of the coupling with alternate or in phase discharges (Fig. 4, C and D).

**Modulation of the rhythm by sensory stimulation**

Our exploration of the sensory modulation of these rhythms was limited for various reasons. Figure 5 shows the effect of extending the hip joints by lowering the backrest of his stretcher. This extension (shown in Red) resulted in a more vigorous discharge of muscles (Erector spinae and Lateral Gastrocnemius) and, in this case, a lowering of the rhythm from 0.8 Hz to 0.4 Hz.

Figure 6 illustrates one example of a strong skin stimulus applied tonically to the lower back (pinch of skin fold between the thumb and index). This could abolish almost entirely the ongoing rhythm which resumed afterwards often with a rebound, which is
quite clear in both the EMG records and the Optotrak records. This manoeuvre could be repeated several times.

**Further evolution of the patient**

In 2007, an X-Ray examination showed that the patient had a moderate coxo-femoral arthritis (see Figure 1B) which could have acted as a trigger for these episodes without the patient being conscious of this because of his complete spinal lesion (8). Bilateral infiltrations of the hip joint with cortisone were performed. After the first injections, he was free of spasticity crisis for 6 months. The infiltrations were repeated one more time but less successfully.

The patient had an aorto-femoral bypass on the left side and had an amputation of the 2\textsuperscript{nd} toe on the left. These ischemic problems may also have acted as a trigger source for these rhythmic bouts.

**Discussion**

This report complements in many ways the interesting observations made by Calancie (7;8) on complete or incomplete patients presenting occasionally rhythmic movements of the lower limbs. This patient is of supplementary interest firstly because he was classified as an ASIA A and did show some alternate activity between antagonist muscles. Secondly, contrary to other previous observations (8), our patient showed a continuous rhythmic pattern lasting for 2-3 days even when sitting in his wheelchair. Thirdly, this patient had clear signs of autonomic dysreflexia that accompanied these bouts of spinal rhythmic activity, as often seen in patients with lesions above T6 (21).

Remarkable similarities exist between the present observations and a long history of observations animal experiments. Of note is the ability of spinal animals to generate
autonomous spinal locomotor rhythms after complete spinalization and the marked influence of peripheral afferents originating from cutaneous receptors or from proprioceptors, namely from the hip joints, on the rhythmic activity. The reader is referred to various reviews (22;23) for more details. For instance, in acute spinal cats (24), the position of the hips was shown to be of paramount importance since flexion of the hips could abolish the fictive locomotor pattern whereas extension would increase it. Furthermore, as indicated before (8), hip pathologies were often associated with these myoclonus. Our patient had a previous ischiectomy and had signs of coxo-femoral arthritis. The effect of a first bilateral infiltration of the hips also attests the potential importance of signals from the hip.

The bouts of rhythmic activity below the spinal lesion would be classified as spinal myoclonus (6). One problem in the definition of myoclonus concerns its frequency. It varies from single isolated events every 10-20 seconds (periodic) to rhythmic discharges in several muscles.

One misconception is that spinal animals generate a locomotor rhythm continuously for instance after pharmacological stimulation with noradrenergic agonists (25;26). Such is not the case and if the cat is not placed on a moving treadmill, it would be impossible to see he remarkable ability of the hindlimbs to walk. Similarly, our patient sitting in wheelchair or lying down on a stretcher could not be expected to sustain a well-organized bilateral activity of the lower limbs. Whether this rhythmic myoclonus is the same as the CPG in the animal kingdom merits attention. This might determine whether active low level locomotor training of patients with SCI should be vigorously promoted.
given the importance of maintaining the optimal functionality of the spinal cord after SCI in the recovery of locomotor function.

The rhythmic locomotor-like activity described here should indeed not be too surprising given earlier (and mostly forgotten) description of autonomous movements in patients with complete section of the spinal cord. A few descriptions are worth remembering. The first (9) describes a patient injured by a bullet which left “a complete absence of spinal cord substance at the vertebral levels of D-3, D-4 and D-5”. Kuhn describes: “the act of reclining supine on a flat bed initiates a complex series of muscle movements below the level of cord injury. The lower extremities, observed throughout a period of ten minutes maintained continual motion. The involuntary activity manifested by this man is unlike the stereotyped flexor and extensor activity seen in most other subjects... Attempts to handle the extremities during this period of activity are met by bursts of powerful, alternating flexion and extension, and the limbs become momentarily completely unmanageable”.

Similar findings were reported previously by Lhermitte (4) who describes a 30 year old patient with a complete section of the thoracic cord by a bullet (the autopsy clearly showed that there was no remaining nervous tissue). Although the patient died 5 months later, he showed, starting after 4 months of the injury, some vigorous automatic movements of the lower limbs. In our own translation, Lhermitte writes « the lower limbs are agitated by continuous spontaneous movements that involved the trunk and obliged to strap the lower limbs to the bed”.

It is believed that the rhythmic activity observed in the patient here is related to the activation of central spinal pattern generator. Future recordings in other patients will
help to better document the characteristics of these rhythmic patterns and their modulation by various sensorimotor manipulations and the potential therapeutic advantage to harness them.

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Legends
Figure 1. MRI, Hip X-Rays and Intervals between episodes of rhythmic movements and pump fillings.

A. T2 Image viewed in the Sagittal plane. This demonstrate a complete transection of the spinal cord at the T5-T6 level with a spinal cord atrophy above the lesion (the high thoracic area), and a stable syringomyelic cavity of less than 3 cm length below the lesion.

B. X-Rays of the lower back showing the Baclofen pump on the Right, the site of the left ischiectomt performed, and signs of coxo-femoral arthritis.

C. The intervals (in days) between the rhythmic episodes and pump fillings are displayed over a period of about 2 years. We could not see any correlation between the two series of events.

Figure 2. Movements recorded by the Optotrak and the EMG Systems

A, shows the placement of the Optotrak diodes and EMG electrodes placed on the subject on its left side during a crisis.

B-Graph showing the flexion-extension angles of the trunk during an episode of spontaneous rhythmic activity.

Figure 3. Continuous multi-channel EMG recordings during an episode of rhythmic movements.
Nine muscles with good signals are represented. The stability of the rhythm is noticeable although there are periods where clearly the rhythm slows down to about half the frequency.

**Figure 4. Patterns of synchronization between pairs of antagonist muscles.**

A, B, C, D,....

A, illustrates an average of EMG signals over 32 cycles triggered on the onset of Rectus Femoris. An alternation between Rectus Femoris and Vastus Lateralis on the left side is very robust. The pattern of synchronization of the averaged signals of the most distal muscles Tibialis Anterior and Lateral Gastrocnemius is much less clear with a second bump appearing in between suggesting periods where the muscles are out of phase rather than in phase.

B, shows individual examples of 5 cycles where Rectus Femoris is clearly out of phase with Vastus Lateralis as in A.

C, Tibialis anterior muscles on both sides are most of the time out-of-phase in this short episode.

D, Average of 23 cycles of Left Tibialis and Left Gastrocnemius (used as the trigger for the average) showing a clear out-of-phase pattern.

**Figure 5. Effect of extending the hips.**

By lowering the backrest of the stretcher from 120 to 130 degrees, the rhythm was slowed down from 0.8 Hz to 0.4 Hz and the amplitude of the muscle discharge in the
back muscle was increased. The EMG of the Left Erector Spinae and Left Lateral Gastrocnemius are shown. The averaged rectified EMG in Black is before extension and the Red traces are after the stretch.

**Figure 6. Effects of pinching the skin of the lower back on EMG activity and flexion-extension movements of the trunk.**

A) A manual pinch applied to the lower back (in between the two arrows) could almost stop the back activity completely for several seconds. This activity resumed often with a surge after the release of the pinch.

B) The same manual pinch (start and end indicated by the arrows) modified also the flexion-extension movements of the trunk reducing the frequency at 0.5 Hz.
Cycle duration = 1082 ms, sd=284 ms, coefficient of variation = 26%, n=39
Figure 4

A

cycle duration 1409 ms

Rectus Femoris L

n=32

Vastus Lateralis L

Tibialis Anterior R

Tibialis Anterior L

Lateral Gastrocnemius R

Lateral Gastrocnemius L

Phase of step cycle

B

Rectus Femoris L

Vastus Lateralis L

1 s

C

Tibialis Anterior R

Tibialis Anterior L

1 s

D

Tibialis Anterior L

Lateral Gastrocnemius L

n=23

Phase of step cycle
Figure 5

- 120°
- 130°
- Erector Spinae L
- Lateral Gastrocnemius L
- 0.8 Hz
- 0.4 Hz
- 3 s
Figure 6

A

Erector Spinae L
Tibialis Anterior L
Lateral Gastrocnemius L

B

Sagittal Trunk Angle (°)

Mean frequency after pinch: 0.5 Hz
Max excursion: 2°