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REPETITIVE SENSORIMOTOR TRAINING FOR ARM AND HAND IN A PATIENT WITH LOCKED-IN SYNDROME

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ABSTRACT. The locked-in syndrome is characterized by quadriplegia, preserved consciousness and inability to respond to the outside world. In recent years, the repetitive execution of identical movements has been demonstrated to be crucial for the recovery of arm and hand function in stroke patients. The present study aimed at investigating the efficiency of repetitive training in a patient suffering from locked-in syndrome due to an occlusion of the basilar artery. Seven months after the brainstem lesion and after a 15-week period of standard inpatient therapy, the repetitive training was applied to the (most affected) right upper extremity in addition to usual therapy. After 42 weeks of the repetitive training for the right arm, it was applied to the left arm. The ranges of active motion as well as functional motor capacity and muscle tone were regularly assessed. During those phases when the repetitive sensorimotor training was applied to the right or left arm, the ranges of active motion, muscle strength and functional motor capacity of the trained arm increased significantly accompanied by a continuous normalization of muscle tone in the flexor muscle groups. Since the prominent functional improvements of the right and left arms were observed during those phases when the repetitive training was applied, these effects were likely to be due to the training rather than to the standard rehabilitation program or extraneous influences. The repetitive sensorimotor training, therefore, appears to be appropriate to improve motor function of the arm and hand and to accelerate the time course of recovery even in patients with almost complete central paralysis of both arms.

Key words: locked-in syndrome; repetitive sensorimotor training; rehabilitation; physiotherapy; arm and hand function.

INTRODUCTION

In 1966, Plum & Posner (20) introduced the term "locked-in syndrome" to characterize patients with preserved full consciousness, but without the ability to respond to the outside world. Lesions in the ventral portion of the pons destroy corticospinal and corticobulbar fibres as well as motor nuclei. Structures within the dorsal part of the pons controlling consciousness and transmitting sensory information to the cortex usually remain intact. Cranial nerves I–IV and midbrain structures above the pons are also spared.

Three varieties of the locked-in syndrome have been distinguished by Bauer et al. (2). The classical locked-in syndrome comprises complete quadriplegia, lower cranial nerve paralysis with more or less intact vertical eye and upper eyelid movements. Patients with incomplete locked-in syndrome have residual voluntary contractions, particularly in distal parts of the arms and legs, but these are weak and, in most cases, functionally useless. Patients who are totally immobile including eye and eyelid movements belong to the complete category (2). The most common aetiology of the locked-in syndrome is basilar artery occlusion, although the same clinical picture may result from a pontine haemorrhage or tumour, encephalitis or traumatic brainstem injury.

The mortality rate within the first year is approximately 60% (19). Death occurs most frequently within the first 4 months (19) and is most often attributed to pulmonary complications or an extension of the lesion. For those patients who survive, chances for a significant recovery are considered to be poor (16). Nevertheless, McCusker et al. (17) report that recovery may continue for years. Prognosis appears to be more favourable in patients with non-vascular rather than vascular aetiology.

In recent years, a specific repetitive sensorimotor training (RST) has been developed to improve the outcome of motor rehabilitation of the centrally paretic arm and hand in stroke patients (6, 8, 9). In daily training sessions, patients are asked to perform voluntary isometric and isotonic contractions repetitively. Isometric contractions are to be executed as rapidly and as strongly as possible; isotonic contractions must cover the entire range of motion that is actively available for the patient at a respective joint. In cases lacking active movement, the intended movements are repetitively carried out by an occupational therapist or physiotherapist. The patients' movement performances are closely observed and arm muscle tone is frequently assessed in order to detect the occurrence of undesired associated movements and an increase in muscle tone. For both assessments, the training is interrupted for a short period and continued after a sufficient reduction of muscle tone.

RST improves functional motor capacity of the centrally paretic upper extremity and accelerates the time course of recovery significantly. Similar positive results are observed when EMG-initiated electrical muscle stimulation is repetitively applied to functionally important muscle groups of the upper extremity (8, 15). The common feature of both training procedures is the repetitive voluntary activation of functionally important muscle groups of the arm and hand. The proprioceptive and cutaneous impulses generated repetitively and time-locked to the unfolding voluntary movement are supposed to form the basis of motor learning (10) as well as motor recovery in stroke patients irrespective of which methodology is employed. Both approaches are appropriate to strengthen sensorimotor coupling within motor regions of the brain involved in the movement.

The aim of the present study was to investigate whether the RST originally designed for hemiparetic stroke patients is of value for motor function in a quadriplegic patient suffering from locked-in syndrome. This study is part of a series of investigations concerning the functional value of repeated motor practice in patients with central motor disorders.

PATIENT AND METHODS

The 45-year-old right-handed patient reported on in the present paper was admitted to hospital because he developed rapidly progressive quadriplegia due to an ischaemic lesion of the ventral pons (and a small infarction of the left thalamus) caused by persistent occlusion of the basilar artery, as demonstrated angiographically. His cardiovascular and pulmonary situation stabilized after a few weeks so that rehabilitation could be started in the acute hospital. It consisted of regular occupational and physiotherapy, including mobilization. After 7 months he was transferred as an inpatient to our rehabilitation hospital. On examination, he was anarthric and quadriplegic, except for some small and weak voluntary hand, finger and elbow extensions on the left side (about 10% of the range of motion). On command, he was able to produce slight contractions of the extensor muscle groups of the right forearm, but without any visible motion. Sensation was normal. Reflexes were hyperactive and he had bilateral Babinski signs. He could communicate by vertical eye movements (upward gaze for "yes") and by eye blinking (eyelid closure for "no"). According to his clinical picture, the patient was classified as suffering from incomplete locked-in syndrome (2). He took no neuroactive drugs during any phase of the study.

Rehabilitation program

The rehabilitation program started immediately after the patient's admission to our rehabilitation department and was applied during all phases of the study in addition to RST. The rehabilitation strategy followed a coordinated team approach. Nurses were responsible for monitoring pulmonary function, mouth care, tracheal suctioning, proper head and body positioning, observing susceptible skin regions, and so on. Within two weeks, the patient learned to use a computer-assisted communication system that could be controlled by eyelid movements. Standard physical exercise therapy was applied daily, including passive movements of the arms and legs (in part using a motor-driven cycling device), bringing the patient into an upright position by means of a tilt table and combating spasticity by slow sustained muscle stretching. The patient received daily conventional occupational and physiotherapy according to the Bobath concept (3, 4) (individual therapy for at least 2 hours a day) supplemented by speech therapy and psychological assistance. Physiotherapy particularly aimed at facilitating trunk, leg and arm movements, whereas occupational therapy was concerned with arm and hand activation, predominantly on the left side, which presented some residual motor function (see above).

Repetitive sensorimotor training (RST)

During the first 15 weeks after admission to our rehabilitation hospital, the patient received the conventional occupational and physiotherapy described above. During this time and the following phases of the study, motor function of the upper extremities was assessed every 3 weeks. Starting at the beginning of the 16th week, RST was practised 5 days per week in addition to the standard rehabilitation program described above. To demonstrate the influence of the training in comparison to conventional occupational and physiotherapy and in order not to overstrain the patient, RST was applied only to his right arm, whereas the conventional rehabilitation program focused on arms, legs and trunk. After the patient had been informed in detail about its aim and design, he agreed to participate in the study, which had been approved by the ethical board of the Free University of Berlin. In particular, the patient was made aware that he could withdraw his consent at any time, and that RST would neither modify nor diminish the standard rehabilitation program he received daily in addition to the repetitive training.

The principle of RST is that it aims at facilitating selective movements at shoulder, elbow, wrist and finger joints. Movements instead of isometric contractions or postural activity are predominant and primary. If the intended movement cannot be executed actively, the motion is carried out by the physiotherapist. Thereafter, parallel to the recovery of voluntary muscle control, selective movements must be performed more and more against gravitational pull, covering the entire range of motion. In a further step, the patient is asked to perform the movements as fast as possible, but to avoid an increase in muscle tone or the occurrence of undesired associated reactions (18). Finally, in order to enhance muscle strength, movements are performed at various resistance levels exerted by weights (100 g to 3 kg) attached to the upper arm or forearm and to the hand dorsum, depending on the target movement. After the patient has achieved voluntary motor capacity at a single joint, movements are trained that involve several (proximal and distal) segments of the arm, including postural and dynamic activities. In our patient, RST lasted between 60 and 70 minutes for each arm. It was applied by two advanced physiotherapists who were regularly supervised by other members of the research group in order to ensure that the RST was administered in an appropriate manner.

Assessments

In order to evaluate the patient's motor capacity, the active range of motion (divided into five categories at 0, 25, 50, 75 and 100% of the total range of motion, both for active movements without gravitational influence and for active movements against gravitational pull) was assessed every three weeks for those movements listed in Table I. Furthermore, motor capacity of the upper extremity was regularly quantified by means of the Fugl-Meyer test (7). Muscle tone was assessed by means of the modified Ashworth scale (5). Ratings were performed by two experienced members of our research group who had years of experience in administering the Fugl-Meyer test and the modified Ashworth scale. Interrater reliability was regularly checked and amounted to 90%.

The Fugl-Meyer test (7) was originally designed to evaluate motor function, balance, some sensation qualities and joint function in stroke patients. In our patient, the development of motor capacity of the upper extremity was regularly assessed by means of items A–C in the arm section of this test. This section comprises reflex activity of the biceps, triceps and finger flexors (max. score 4), voluntary movements completely within flexor and/or extensor synergies (max. score 18), voluntary movements mixing flexor and extensor synergies (max. score 6), voluntary movements with little or no synergy dependence (max. score 6), voluntary movements of the wrist (max. score 10) and hand (max. score 14) and, in later stages, the normalized activity of tendon reflexes (max. score 2). The Ashworth scale (5) was used to assess muscle tone of various parts of a limb. It ranges from 0 (flaccid tone) to 5 (rigid fixation). Conventional testing of muscle strength, for instance by means of the MRC scale, appears to be a somewhat inadequate assessment instrument, as motor activity in patients with brain lesions is often dependent on synergies.

Course of the study

The rehabilitation program started, as mentioned above, about 7 months after the acute stroke. The active ranges of motion (Table I) as well as muscle tone and functional motor capacity of the arm were assessed every 3 weeks. Fifteen weeks after admission to our rehabilitation department, RST started for the right arm in addition to the usual inpatient rehabilitation program. To demonstrate the influence of RST, it was applied—during the first 42 weeks—only to the right arm, which had shown no voluntary motor activity at the beginning of the study. If the ranges of voluntary movement and/or the functional motor capacity increase at the moment when RST is introduced, it is likely due to the training rather than to the standard rehabilitation procedures or extraneous events. From the beginning of the 43rd week, RST was also applied to the left arm, the residual motor capacity of which had been slightly superior to that of the right arm at the time of admission to the rehabilitation department. After a total of 78 weeks of inpatient rehabilitation, RST had to be interrupted due to the patient's dismissal from hospital. The time course of the training can be inferred from Fig. 1.

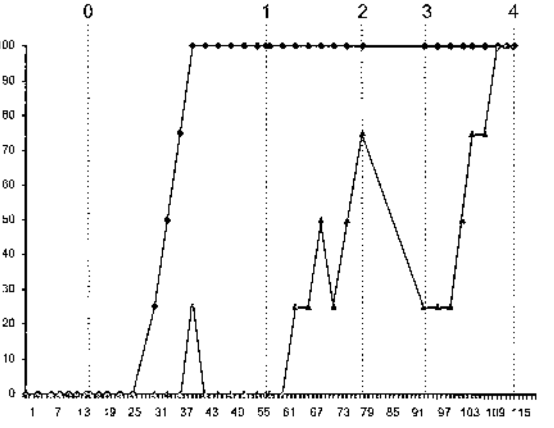

Fig. 1. Time course (x-axis in weeks) of the range of motion (y-axis in % of the maximum range of motion) for active anteversion of the upper arm (without gravitational pull). 0: Beginning of repetitive sensorimotor training (RST) for the right arm; 1: Beginning of RST for the left arm; 2: Dismissal from the rehabilitation hospital; 3: Resumption of RST; 4: End of the study. Right arm: ◆; left arm: ▲.

RESULTS

Figure 1 illustrates the influence of RST on the range of motion of active anteversion of the upper arm. After 15 weeks of repetitive training, active movements covering about 25% of the range of motion were observed. The complete range of active motion was reached after 24 weeks. During the following weeks, the patient experienced further recovery of strength in the involved muscle groups. Similar developments were observed with the other movements that were regularly assessed. The improvement in active range of motion began at the moment when the RST was started. This held true for both right and left arms (Table I).

The improvement in range of active motion at various joints was accompanied by increasing scores on the Fugl-Meyer scale (Fig. 2). At the end of the study, the patient was able to use both arms effectively in several activities of daily living provided that they did not require high degrees of muscular force. Furthermore, RST contributed to the reduction of enhanced muscle tone, as is evident in Fig. 3 for those muscle groups acting on the elbow. Muscle tone began to diminish for the right arm after the 15th week and for the left arm after the 57th week, i.e. the moments at which the repetitive

Table I. *Ranges of active motion (right and left arm) (in % of the total range of motion)*

	0[a] R[b]	0 L	1 R	1 L	2 R	2 L	3 R	3 L	4 R	4 L
Movements without gravitational pull										
Shoulder										
Anteversion of the upper arm	0	0	100	0	100	75	100	25	100	100
Retroversion of the upper arm	0	25	100	100	100	100	100	75	100	100
External rotation	0	50	100	50	100	100	100	50	100	100
Internal rotation	0	25	100	100	100	100	100	75	100	100
Abduction	0	25	50	50	100	75	100	50	100	100
Adduction	0	75	100	50	100	100	100	100	100	100
Elbow										
Extension	0	25	100	50	100	100	100	75	100	100
Flexion	0	25	100	75	100	100	100	75	100	100
Pronation	0	75	100	75	100	100	100	50	100	75
Supination	0	25	100	50	100	100	100	50	100	100
Wrist										
Extension	0	50	100	50	100	75	100	75	100	100
Flexion	25	50	100	75	100	100	100	50	100	100
Ulnar abduction	0	50	100	50	100	100	100	50	100	100
Radial abduction	0	100	100	75	100	100	100	50	100	100
Fingers										
Extension	0	25	100	50	100	75	100	50	100	75
Flexion	25	50	100	75	100	100	100	75	100	100
Abduction of the index finger	0	0	100	50	100	75	100	50	100	100
Adduction of the index finger	0	0	100	75	100	75	100	50	100	100
Thumb										
Opposition	75	75	100	25	100	50	100	25	100	100
Extension	75	0	100	0	100	75	100	50	100	100
Flexion	0	0	100	25	100	75	100	75	100	100
Movements against gravitational pull										
Shoulder										
Anteversion of the upper arm	0	0	75	0	100	0	100	0	100	0
Retroversion of the upper arm	0	0	75	100	100	75	100	0	100	50
External rotation	0	0	100	0	75	25	100	0	100	50
Internal rotation	0	0	100	0	100	50	100	0	100	75
Abduction	0	0	75	0	100	0	100	0	100	0
Adduction	0	0	100	0	100	25	100	0	100	50
Elbow										
Extension	0	0	100	0	100	75	100	0	100	100
Flexion	0	0	100	0	100	100	100	0	100	100
Pronation	0	0	100	0	100	50	100	0	100	0
Supination	0	0	100	0	100	50	100	0	100	100
Wrist										
Extension	0	0	100	0	100	0	100	0	100	75
Flexion	0	0	100	0	100	25	100	0	100	75
Ulnar abduction	0	0	100	0	100	50	100	0	100	25
Radial abduction	0	0	100	0	100	50	100	0	100	75
Fingers										
Extension	0	0	100	0	100	0	100	0	100	0
Flexion	0	0	100	0	100	75	100	0	100	75
Abduction of the index finger	0	0	100	0	100	0	100	0	100	0
Adduction of the index finger	0	0	100	0	100	0	100	0	100	25
Thumb										
Opposition	0	0	100	0	100	0	100	0	100	50
Extension	0	0	100	0	100	0	100	0	100	50
Flexion	0	0	75	0	100	0	100	0	100	0

[a] 0: Beginning of repetitive sensorimotor training (RST) for the right arm; 1: Beginning of RST for the left arm (42 weeks after the beginning of the training for the right arm); 2: Interruption of the training for 4 months; 3: Resumption of the training; 4: End of the study.
[b] R: right; L: left.

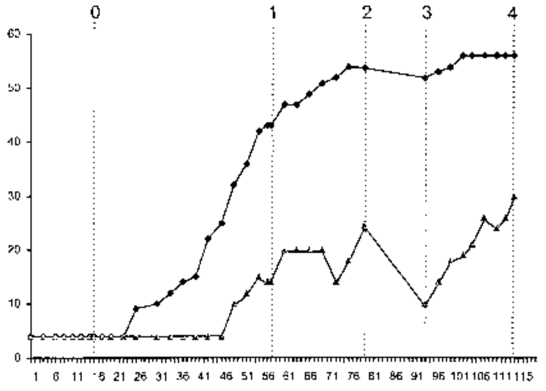

Fig. 2. Time course (x-axis in weeks) of the scores (y-axis) of the Fugl-Meyer Scale (upper extremity section). 0: Beginning of repetitive sensorimotor training (RST) for the right arm; 1: Beginning of RST for the left arm; 2: Dismissal from the rehabilitation hospital; 3: Resumption of RST; 4: End of the study. Right arm: ◆; left arm: ▲.

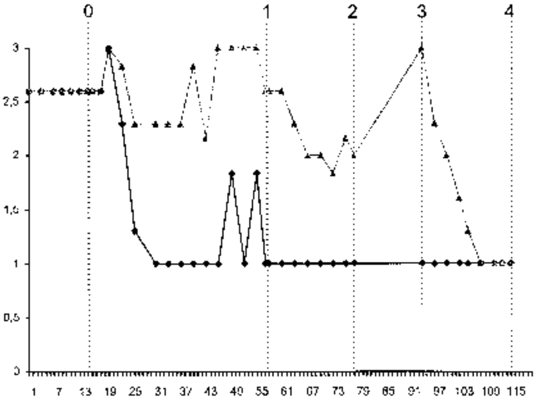

Fig. 3. Muscle tone (assessed by means of the modified Ashworth scale) for the flexor muscles acting on the elbow and on the hand (x-axis: time course in weeks, y-axis: average of the individual Ashworth scores). 0: Beginning of repetitive sensorimotor training (RST) for the right arm; 1: Beginning of RST for the left arm; 2: Dismissal from the rehabilitation hospital; 3: Resumption of RST; 4: End of the study. Right arm: ◆; left arm: ▲.

training was introduced for the right and left arms, respectively (Fig. 3).

DISCUSSION

According to Patterson & Grabois (19), spontaneous recovery in patients suffering from locked-in syndrome is expected to be most prominent during the first 4–6 months. Katz et al. (13) emphasize that lack of recovery within the first 6 months predicts persistent locked-in syndrome. Nevertheless, according to McCusker et al. (17), recovery may continue for years, particularly in patients with non-vascular rather than vascular aetiology. Since our patient was admitted to the rehabilitation department about 7 months after the acute brain lesion, and because RST started 15 weeks later, more than 10 months had passed when RST commenced. Therefore, spontaneous recovery likely only played a minor role in functional restitution. Furthermore, it had been conclusively demonstrated in previous studies that the effect of repetitive training is indeed specific and not simply due to enhanced treatment time (6, 9).

The design of the present case study is appropriate to document the impact of the repetitive training precisely (14), since improvements in the range of active motion and in motor function were observed exclusively for the right arm after training for that arm had started. The same holds true for the left arm, whereas only minor changes in motor parameters occurred during the first 57 weeks when conventional occupational and physiotherapy were applied, and during the 15 weeks when the training was interrupted. During the period when RST was interrupted, the patient experienced a decline in motor function of the left arm. Unlike the right arm, the function of the left arm was not yet sufficient to allow him to perform everyday life activities. This fact could have contributed to a reduction in voluntary activation of this arm, and to a subsequent decrease in the active range of motion.

During all phases of the study, the patient received conventional physiotherapy according to the Bobath concept (3, 4), supplemented by occupational therapy. Both therapeutic approaches aim at reducing spasticity, whereas RST seeks to activate centrally paretic muscle groups directly. As was pointed out convincingly by Bütefisch et al. (6) for the centrally paretic hand, therapeutic measures focusing on muscle tone reduction contribute little to functional motor improvement. On the other hand, when RST was applied for the right arm its motor function improved, whereas no relevant changes were observed for the left arm. At the moment when the repetitive training started for the left arm, both active range of motion and functional motor capacity increased.

RST improved motor function significantly. The patient was able to perform useful activities such as shaving and brushing his teeth without external aid. Nevertheless, he had to be supported in motor activities requiring higher levels of muscular force. These facts justify the application of RST. Furthermore, the additional costs appear to be relatively low. The physiotherapist, who spends between 120 and 140 minutes per day on the training, i.e. around 25% of her or his daily working time, contributes to this added expense. The additional costs due to RST, therefore, run up to $1000 per month.

In contrast to the repetitive training as it was described previously (6, 8, 9) for the affected hand of stroke patients, the training described in the present paper was applied to all functionally relevant muscle groups of the centrally paretic arm. Furthermore, the patients included in the studies by Bütefisch et al. (6) and Hummelsheim et al. (8, 9) had some residual motor function of the hand, whereas the patient described in the present study was completely paralysed, except for some slight contractions of the extensor muscles of the left arm and forearm. The repetitive training, therefore, started with motions carried out by the physiotherapist. Nevertheless, the repetitive motor action was appropriate to initialize and maintain functional recovery.

An important feature of RST is the fact that it places high value on facilitating selective movements and emphasizes the avoidance of gross synergistic patterns. In fact, the degree of spasticity decreased in parallel with the growing voluntary motor capacity, as was observed in previous studies with stroke patients (6, 8).

Both the modified RST described here and the repetitive training described in previous studies (6, 8, 9) consist of stereotyped repetitive motor activities. According to Asanuma & Keller (1), the proprioceptive and cutaneous impulses generated repetitively and time-locked to the movement are thought to form the basis of motor learning as well as of motor recovery in patients with lesions in the central nervous system. Since our patient presented normal sensory perception, the repetitive afferent stimulation together with movement-related activity in motor centres of the brain could have induced unmasking phenomena within pre-existing neuronal circuits (11) and probably enhanced cortical representational zones (12). Furthermore, long-term potentiation phenomena brought about by repeatedly arising afferent stimuli might have facilitated the excitation of respective subsets of motor cortical neurones activating particular muscles or muscle groups (10).

The striking but long-term recovery induced by RST is probably supported by the known high degree of redundancy within the motor system formed by the parallel and independent organization of inputs and outputs for different representations of muscle groups. This has been illustrated in detail by Strick (21) for the hand representation in motor, premotor and supplementary motor areas of the cortex. These areas differ in their access to brainstem motor centres as well as to spinal motoneurones and interneurones. RST could have activated alternative representational zones in secondary motor areas or in the brainstem, which possess relatively independent descending pathways that bypass the lesioned brainstem regions. Future prospective studies should investigate the influence of repeated motor practice in patients suffering from less severe brainstem or incomplete spinal cord lesions.

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