Developing a late rehabilitation programme for severe brain damage: A single-subject case study

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Abstract

Objectives: To determine whether an intensive, long-term programme of diverse physiotherapeutic interventions at a late stage after severe brain damage would lead to improvements of mobility and function.

Participant: The patient is a young woman with encephalitis lethargica followed by an anoxic episode, diagnosed as minimally aware.

Interventions: Treatment modalities were wide-ranging, including electrical stimulation (sinusoidal intermittent current interferential) and passive and active exercise using an Oswestry frame, tilt table, motorised bicycle and treadmill with assistance. Treatment was initiated nearly one year after the initial illness, reaching a level of about 25 hours/week after several years.

Results: Initial ES treatment of the throat muscles to promote swallowing allowed removal of the percutaneous endoscopic gastronomy (PEG). ES of the quadriceps and other muscles led to effective bridging and assisted standing, making care considerably easier. After 4 years of treatment, the patient is starting to chew and can stand for long periods with assistance only for balance. There has been a modest improvement in consciousness level assessed with the JFK Coma Recovery Scale.

Conclusions: The patient's mobility and function were improved, and the burden on carers was reduced. Acquisition of weightbearing ability may reduce the risk of complications such as osteoporosis, urinary tract infections and pressure sores. Intensive physiotherapy for severely brain-damaged patients might be economically justified by reduced lifetime care and treatment costs.

Key words: rehabilitation; encepalitis lethargica; brain injury; minimally aware

Introduction

In Britain, rehabilitation following brain injury has been dominated by the Bobath concept of neurodevelopmental treatment since the 1970's [1, 2]. However, some recent reports indicate that an eclectic approach may be more effective [3,4,5], although it remains important to clarify whether all or only some of the component treatments improve outcomes [6].

It is generally accepted that by two years after neurological damage, further recovery is minimal, and rehabilitation programmes after this period have been considered to be of limited value. However, this idea has been challenged [7,8]. Here we present a case study of a long-term programme of intensive, multi-modal physiotherapy in a patient with severe brain damage.

Case presentation

The 17-year-old patient, whom we will refer to as Camilla, initially presented in February 1999 with general febrile illness with rapid onset of neuropsychiatric symptoms and seizures. She was diagnosed with encephalitis lethargica [9], a rare disease that may have a post-infective autoimmune etiology [10]. She was heavily sedated, intubated and admitted to intensive care, where tracheostomy was performed to facilitate ventilation. A percutaneous endoscopic gastronomy (PEG) was fitted soon thereafter. After about 7 weeks, she suffered a cardiac arrest and MRI revealed severe hypoxic brain damage. She was transferred to a neurological unit and six months later was discharged home at her parents' request. No further active rehabilitation

was recommended. All nutrition was via the PEG and all medication was stopped except fluoxetine. She was not catheterised, but remained unresponsive and doubly incontinent, and showed mild flexion contractures and increased tone in all four limbs. She was diagnosed as minimally aware with no discernible hearing or vision. Subsequent reinvestigations revealed adequate hearing and possibly minimal vision. Table 1 at the end of the paper summarizes the timeline of symptoms, treatments and outcomes.

Ethical considerations

Instigated by her family, an eclectic dynamic patient-priority-driven rehabilitation programme was started at home. The positive initial response suggested that mobility and function could be improved. Long-term intensive physiotherapy, even at a late stage, can be beneficial in brain-damaged patients with severe disability [7, 11], and a quasi-experimental study was proposed. As Camilla was unable to give consent, the criteria for treatment methodologies to be included were firstly, that they should do no harm, and secondly, that they might provide benefit. Camilla's parents gave informed consent for the study, and were aware that they could withdraw consent at any time without affecting the availability of any treatment that they wished to be continued.

Treatment programme

The main treatment modalities were exercise (passive and volitional), electrical stimulation (ES), acupuncture and weight-supported treadmill training. Besides the physical benefits, it was hoped that sensory input to the brain from

the treatments might also overcome ‘learned non-use’ arising from non-reinforcement after injury [12].

Leyendecker [13] reported that a combination of electrical stimulation and traditional Bobath-based physiotherapeutic treatment was more successful than Bobath-based physiotherapy alone. Other studies [14-16] have confirmed a role for ES in rehabilitation of brain-injured patients and in reduction of spasticity [17]. We employed a sinusoidal intermittent current (IF) for muscle stimulation as our clinical experience indicates it can be safely applied for long periods. The carrier frequency was set at 4000 Hz, which caused no apparent skin irritation, but resulted in good muscle contraction.

It was decided that ES would be used initially to re-educate the pharyngeal muscles, as effective swallowing would facilitate removal of the PEG. Swallowing is a complex sequence of events involving obligate muscles that work in a sequence of excitatory and inhibitory events, but once the pharyngeal phase of swallowing is initiated, it becomes an irreversible motor event. Therefore, we focused on the mylohyoid, digastric and internal pterygoid muscles. To achieve accurate targeting with minimum discomfort to the patient, we used small adhesive electrodes to deliver ES at a frequency of 0-10 Hz and an intensity of 15-20 mA, which produced visible muscle contraction. Thirty-minute sessions were administered daily. As swallowing of saliva improved, feeding of liquids and soft foods was commenced, and this proved successful enough to allow removal of the PEG (see Table 1).

Bridging was also an early focus, as a precursor to standing. ES was administered at 0-10 Hz to the gluteal muscles through 6 cm x 7.5 cm rubber electrodes placed at either end of the muscles. Initially, an intensity of 100 mA

produced no apparent contraction, indicating marked muscle weakness, and emphasis was placed on prone lying and stretching of the slightly shortened hip flexor muscles. The intensity required to produce contraction was gradually reduced to 30-60 mA. Supported standing using a tilt table and active bridging with assistance were encouraged, and the caregivers were taught the appropriate techniques for daily treatments. Camilla gradually developed the ability to bridge well enough to make toileting and dressing considerably easier.

Treatment was continued with concomitant ES of the quadriceps and gluteals, together with a tilt table, by means of a cross-over technique with 2 machines, employing two 6 cm x 7.5 cm electrodes on the gluteals and two on the opposite quadriceps muscle, initially at 130-120 mA. A vector surge frequency of 0-10 Hz appeared to decrease the flexion reflex that was preventing weight-bearing. ES was also used to work the hamstring muscles under stretch to reduce spasticity. Passive stretching of all limbs was continued and trunk stability was worked on using a physio ball, with active assisted high kneeling exercise to encourage hip stability. Treatment conditions are summarized in Table 2 at the end of the paper.

Camilla became able to stand in an Oswestry frame with the aid of two people. She actively used her quadriceps and hip extensors to push herself upright, but was unable to support her body weight for more than a few seconds. A Biodex unweighing system was introduced to allow standing while supporting only partial body weight, with continued ES to the gluteals and the quadriceps. The intensity of physiotherapy was increased to 25 hours/week, and after several months, Camilla could support her full body weight in a standing position for more than 40 minutes.

Inversion and plantar flexion of the feet were correctable manually, but continuing occasional severe spasms ruled out polypropylene in-shoe orthoses. Increased gentle mobilisation and stretching along with ES to the peroneal muscles were used to prepare Camilla for bilateral below-knee callipers with T-straps. The orthoses improved stability, and Camilla became able to stand with the assistance, mainly for balance, of one person.

Daily work on a Reck Moto-Med motorised bicycle was introduced to mobilise the lower limbs, encourage volitional movement, and increase muscle strength and stamina. The resistance was initially set to zero, and gradually increased. Camilla's strength and stamina increased markedly, with the distance actively cycled reaching 60-80% of the session distance. A treadmill-based locomotor training programme was also introduced. This provided variety in the exercise programme, as Camilla's achievement level was better maintained or increased if treatment methods were varied.

Outcome measures

Several interim measures were used to guide treatment. In the case of ES, the milliamperage needed to obtain a muscle contraction when a muscle was electrically stimulated was used. To follow progress in weightbearing ability and muscle strength, the Biomed unweighing system was initially used, but once Camilla was able to support her full body weight this was no longer appropriate. The Motomed bicycle, which registers volitional activity, was used to measure the distance and duration of active cycling and these were

recorded as a percentage of total distance or time, respectively, in each session. The resistance offered by the machine can be adjusted appropriately.

For overall treatment outcome, we considered that the most suitable measures in this single-patient case study would be objective achievements directly related to the level of care required by the patient. We also employed the JFK Coma Recovery Scale - Revised (CRS-R), which is appropriate for evaluating minimally aware patients [25]. The widely used Glasgow Coma scale is unsuitable for evaluating changes in such patients, and the FIM/FAM scale is also unsuitable to measure small improvements that are nevertheless significant in terms of care. For example, although Camilla became able to chew and swallow, she still required complete assistance with feeding.

RESULTS

Intensive physiotherapy of this initially immobile, minimally aware patient led to removal of the PEG, and recovery of swallowing and chewing ability, as described under *Treatment programme*. She became able to bridge effectively, to bear weight, and to stand for long periods with assistance only for balance (Table 1). Assessments using the JFK Coma Recovery Scale indicated a modest improvement in the level of consciousness, although she remained minimally aware (Table 3 at the end of the paper).

Discussion and Conclusion

Recent reports [7, 21, 22], and our results, suggest that intensive rehabilitation can produce improvement even at a late stage after severe brain injury. Since diagnosis of consciousness level is not straightforward, and the late recovery rate of patients initially diagnosed as being in a vegetative state (VS) is as high as 14% (number recovering after 1 year/number still in VS at 1 year) [23, 24], the rehabilitation needs of minimally aware or vegetative patients must be considered in the light of potential consciousness recovery. Giacino [25] noted that inability to predict the specific functional outcome of an individual patient gives rise to ethical and use of resource issues.

Electrical stimulation of muscles formed a large part of the treatment programme used here. There is considerable evidence [13,15,26] that ES is a useful adjunct to functional exercise in an aggressive approach to rehabilitation following brain injury, and our results support this.

Our results also support the idea that long-term physiotherapy for severely brain-injured patients has a number of advantages, and may offer cost savings in lifetime care. Firstly, treatment focusing on weight bearing and strength makes it easier for carers to transfer, toilet and dress the patient with less risk of injury to themselves. Secondly, weightbearing is expected to reduce the risk of osteoporosis and fractures, urinary tract infections and pressure sores. Thirdly, carers can be trained to carry out daily physiotherapy routines; the physiotherapist needs only to re-assess on a regular basis, so the cost need not be high. We think a larger-scale study would be justified to see whether the

lifetime cost of support for severely brain-injured patients would be decreased by a long-term programme of physiotherapy.

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Ethical approval: The patient's parents gave informed consent for this study on a continuing basis. The study was approved by the Ethical Committee of the Royce Clinic.

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Table 1
Timeline of symptoms, interventions and outcomes

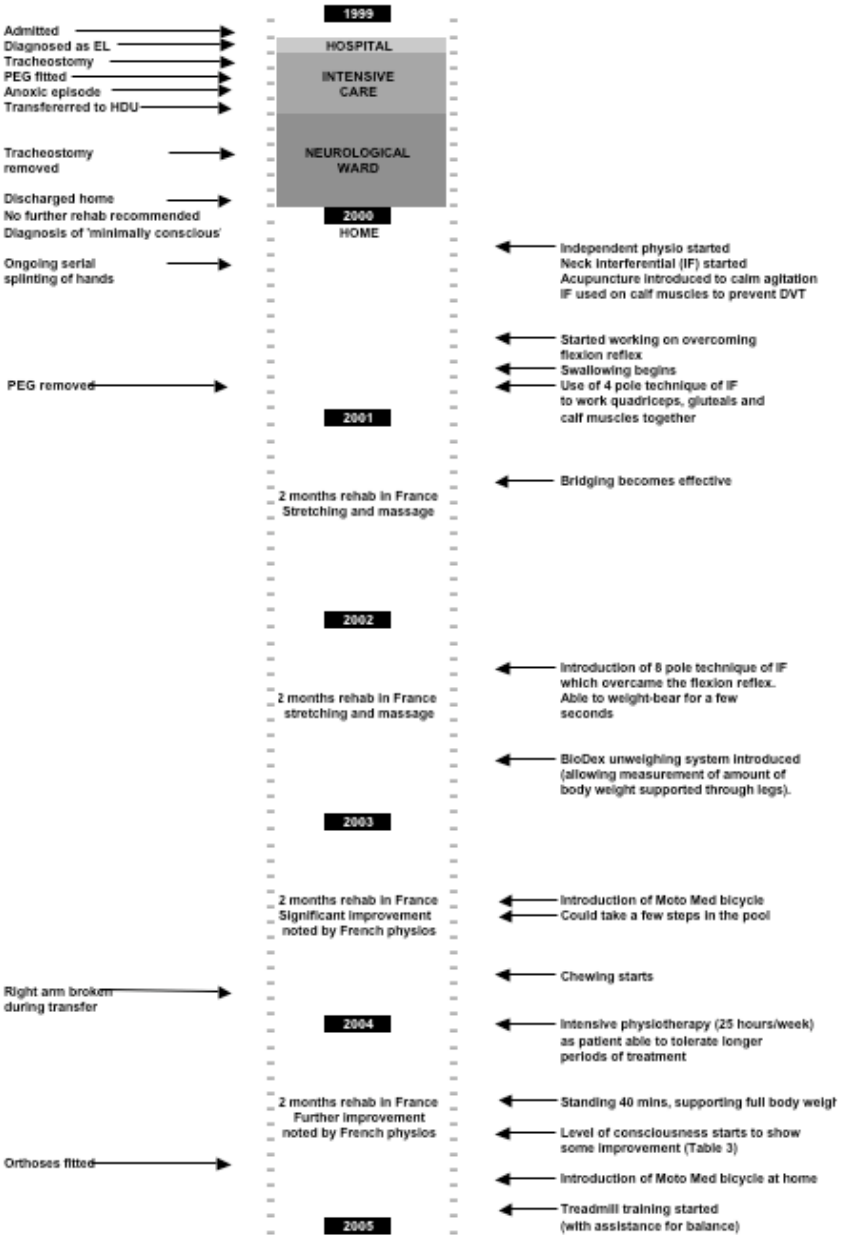

Table 2
Treatment aims, interventions and conditions

Treatment Aims	Interventions	Conditions
Re-educate swallowing	Electrical stimulation*	Sinusoidal intermittant current (IF) Carrier frequ. 400 Hz Freq. 0-10 Hz Intensity ~43 mA
Promote normal muscle tone	Electrical stimulation*	IF Carrier freq. 400 Hz Frequ. 150-250 Hz Intensity - varied with muscle group
	Stretching	Passive - all limbs
	Exercise	Passive and active as able
Strengthen muscles	Active exercise	Motomed bicycle - gear 2 Sit - stand in Biodex unweighing frame
	Electrical stimulation*	Sinusoidal intermittant current (IF) Carrier freq. 400 Hz Freq. 0-10 Hz Intensity - varied with muscle group
Encourage active movement	Active exercise	Motomed bicycle - gear 1 Sit - stand in Biodex frame Locomotor training with treadmill
Improve level of awareness	Active exercise	As above
	Acupuncture	GV20, GB20, B10
Relaxation/stress reduction	Acupuncture	GV20, Ex-HN1, Ex-HN6, GB14, GB20, LI4

* See text for details.

Table 3

Score sheet of changes in JFK Coma Recovery Scale - Revised

Patient: Camilla Date:	**11.07.04**	**10.09.04**	**12.11.04**	**04.03.05**	**16.05.05**
AUDITORY FUNCTION SCALE					
4 - Consistent Movement to Command					
3 - Reproducible Movement to Command					
2 - Localization to Sound			**X**	**X**	**X**
1 - Auditory Startle		**X**			
0 - None	**X**				
VISUAL FUNCTION SCALE					
5 - Object Recognition					
4 - Object Localization: Reaching					
3 - Visual Pursuit					
2 - Fixation					
1 - Visual Startle	**X**	**X**	**X**	**X**	**X**
0 - None					
MOTOR FUNCTION SCALE					
6 - Functional Object Use					
5 - Automatic Motor Response					
4 - Object Manipulation					
3 - Localization to Noxious Stimulation					**X**
2 - Flexion Withdrawal	**X**	**X**	**X**	**X**	
1 - Abnormal Posturing					
0 - None/Flaccid					
OROMOTOR/VERBAL FUNCTION SCALE					
3 - Intelligible Verbalization					
2 - Vocalization/Oral Movement	**X**	**X**	**X**	**X**	**X**
1 - Oral Reflexive Movement					
0 - None					
COMMUNICATION SCALE					
2 - Functional: Accurate					
1 - Non-Functional: Intentional		**X**	**X**	**X**	**X**
0 - None	**X**				
AROUSAL SCALE					
3 - Attention					
2 - Eye Opening w/o Stimulation		**X**	**X**	**X**	**X**
1 - Eye Opening with Stimulation	**X**				
0 - Unarousable					
TOTAL SCORE	**6**	**9**	**10**	**10**	**11**