Is a multidimensional robotic rehabilitation approach feasible in Guillain-Barrè syndrome? Report from a clinical case

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Abstract

We describe the case of a 54-year-old patient, who was admitted to our Rehabilitation Unit with diagnosis of Guillain–Barré Syndrome (GBS). Neurophysiological investigation revealed an axonal polyneuropathy, with impairment of the motor component and preserved sensory function. Despite rapid diagnosis, therapeutic treatment and customized progressive rehabilitation program, nine months after the onset of the disease he presented severe motor sequelae and functional impairment. Patient's hospitalization lasted about two months, while he performed a conventional rehabilitation training. Besides, he underwent a 4-week comprehensive rehabilitation treatment, including both conventional and robotic multidimensional trainings, for 5 1-h sessions per week. Despite the residual global impairment, this treatment, specifically tailored on patient's skills and progress, promoted improvements in functional abilities such as motricity, trunk control, and activities of daily living. Therefore, this case report evidenced the feasibility and efficacy of a multidimensional robotic therapeutic approach along with conventional treatment in the post-acute phase of GBS.

Key Words: Guillain-Barrè syndrome, robotic therapy, comprehensive treatment, disability.
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Guillain–Barré Syndrome (GBS) is an autoimmune inflammatory polyneuropathy characterized by an acute onset, rapid progression of ascending symmetric motor and sensory deficits with frequently associated autonomic dysfunction, Cranial Nerve (CN) involvement and respiratory failure.[1,2] It represents the most frequent cause of acute and sub-acute flaccid paralysis, with a reported annual incidence of 0.5–2 cases per 100,000 people.[3] The treatment of GBS requires a multidisciplinary approach consisting of general medical care and immunological treatment. Indeed, all patients with GBS need meticulous monitoring of respiratory, cardiac and hemodynamic function, and the early start of rehabilitation treatment and psychosocial support is quite recommended.[4]
As regards the therapeutic approach, Plasma Exchange (PE) and Intravenous Immunoglobulin (IVIg) have been used for several years for the treatment of GBS. Anyway, extensive research to elucidate the pathophysiology of GBS has led to the investigation of new potential therapeutic targets and the development of new drugs directed towards them.[5,6] Besides, past research supports an early and intensive rehabilitation approach for patients with GBS to promote functional recovery.[4] About 40% of all cases require inpatient rehabilitation as most patients are very disabled and will require ventilator support during the acute stage of the disease. Intensive exercise, in the inpatient setting, focused on strengthening, endurance training, Activities of Daily Living (ADL) and, when possible, gait training, have proven effective to improve functional independence. Nowadays, less information is available on the impact of rehabilitation in the chronic phases of the disease; however, there is some evidence promoting continued high-intensity and progressive strength, endurance, and functional skill training.[7,8]
Rehabilitation comprehends an interdisciplinary team approach, encouraging active patient and family education and participation, using a goal-oriented functional approach to maximize functions, activities and participation. In this panorama, rapid technological developments that occurred over these years have allowed to optimize rehabilitation intervention. Computer- and robot-assisted devices for upper extremity rehabilitation and virtual reality therapy have

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proven to be an important component for optimizing rehabilitation.[9] The focus of this case report is to describe the feasibility and the effects of Conventional Physiotherapy (CT) intervention, associated with multidimensional Robotic Training (RT) for a patient with a diagnosis of GBS in a post-acute phase of disease.

Case Report

We describe the case of a 54-year-old patient, who presented to the Emergency Department with sudden onset of ascending paralysis involving both upper limbs in April 2022. He had had a preceding onset of mild-flu like syndrome associated with gastrointestinal disturbances. He was diagnosed with GBS after performing a lumbar puncture for the cerebrospinal fluid analysis, and treated with plasma exchange and intravenous immunoglobulin in the Neurologic Unit, performing a total of four cycles of intravenous immunoglobulin along with steroid bolus. Then, he was referred to an intensive-care unit for the severe deterioration of his neurological and respiratory condition.

Following stabilization of clinical conditions and after achieving complete weaning from ventilatory support, the patient was transferred to the Rehabilitation ward of a Rehabilitation Respiratory Center in September 2022. Then, he started an intensive rehabilitation program. The patient received a comprehensive rehabilitation treatment, including respiratory and motor rehabilitation. The intensive rehabilitation treatment was continued in our Rehabilitation ward, where he entered in January 2023. At entrance, the upper and lower limbs strength was evaluated by means of manual muscle testing using the Medical Research Council scale (MRC):[10] he was scored 2 at shoulder abduction and wrist flexors, and 1 at elbow extension, hip and knee extension; he was scored 0 at ankle dorsiflexion, bilaterally. Furthermore, the patient presented some stiffness and a degree of contraction in the distal upper and lower limbs: the metacarpo-phalangeal, proximal and distal inter-phalangeal joints and ankle joints revealed the most reduced joint of movement, bilaterally. Evaluation of respiratory gas exchanges revealed a good balance, with no need for oxygen supply. Neurophysiological investigation revealed an axonal polyneuropathy with impairment of motor component and preserved sensory function.

Measures

The patient's functional status and disability were assessed by means of different clinical scales before (T0) and after (T1) one-month comprehensive training period: both the MRC, assessed at T0, and the Motricity Index[11] were used to evaluate segmental force at upper limbs, in particular shoulder flexors-extensors, elbow flexors-extensors and wrist flexors force,[12] while the Trunk Control test[13] evaluated the trunk control. Moreover, disability in basic activities of daily living was assessed by the Barthel Index.

Training protocol

The patient's hospitalization lasted about two months, while he performed a conventional rehabilitation training. Besides, the patient received a comprehensive rehabilitation treatment, including both conventional and robotic multidimensional programmes. The comprehensive rehabilitation accounted for one-month treatment, five sessions per week; each robotic session lasting about an hour. Conventional training lasted about an hour per day, with a progressive increase in duration based on patient's fatigability and achievements. It was mainly focused on daily standardized passive or active-assisted motion sessions of the upper and lower limbs; breathing exercises for respiratory muscles were focused on chest expansion, controlled breathing and diaphragmatic re-education.[12] According to patients' functional improvements, the programme included coordination exercises for trunk control. A gradual mobilization programme was performed to attain independent bed mobility, safe transfer skills and improved sitting balance.

The multidimensional robotic treatment focused on both upper limbs and trunk, and was performed using three different robotic devices focusing on proximal and distal upper limbs and trunk control movement. Each robotic session lasted about one hour and the time spent on each robot was progressively increased along with patients' improvements, with a goal-time of nearly 15-20 minutes on each device. Breaks between each robotic session lasted about 5-10 minutes and were used to recover and discuss progressions and limitations. The choice of motor tasks and progression was specifically tailored to the patient's competencies and needs, under the supervision of a physiotherapist. Specifically, for complexity progression the patient was encouraged to increase his level of functioning among sessions, according to his motor disabilities.

In this case, we proposed a widely used commercial array of devices furnished by Tyromotion GmbH (Graz, Austria). The array included a robotic device for treatment of the hand, AMADEO, an electromechanical device for the bilateral treatment of the upper limbs, DIEGO, and a sensor-based system, PABLO for the treatment of the trunk and the upper limb (Figure 1). At the beginning, the hand robot (AMADEO) helped to train various aspects of the grasping movement and to work on targeted finger flexion-extension training. Active participation of the patient's affected hand was promoted by biofeedback and assist-as-needed interactions implemented in a highly motivating therapeutic gaming environment. The entire hand and individual finger exercises were controlled through isometric force or range of motion and were used to train different functional activities. With the progressive improvement of motor recruitment at the level of the shoulder girdle, DIEGO and PABLO devices were included in the training protocol. An electromechanical device, such as the DIEGO, helped to train functional reaching movements as a whole focusing on lifting, reaching, and transferring components of the movement, or targeting a specific joint movement. This system owned an arm weight compensation system, enabling unilateral or bilateral assistance as needed that adapts to the actual position of the upper limb in 3-dimensional space. The distal wrist and elbow assistance increased proximal trunk and shoulder stability and helped to decrease compensatory movements. The third device, PABLO, enabled the therapist to extend and focus the therapy on specific joint districts of the upper limb and trunk. In particular, with the DIEGO, active-assisted and active exercises were performed for the proximal muscular districts of the upper limbs in three-dimensional space with large numbers of repetitions in a motivating therapeutic gaming environment, while with PABLO, exercises were included using two additional tools, namely the multiboard and the multiball, to improve upper limb dexterity and trunk control.[9]

Results

Results are related to a 4-week comprehensive rehabilitation treatment, including both conventional and robotic multidimensional trainings. In fact, nine months after the onset of the disease, he was hospitalized for a two-months period, while performing a conventional rehabilitation training. Additionally, he underwent a 4-week comprehensive rehabilitation treatment, including both conventional and robotic multidimensional training. Results from functional scales revealed an improvement of patient's motor abilities (Figure 2) from T0 to T1. With respect to the Motricity Index, we found a relative recovery for upper extremities strength on both right (T0: 9/100 vs T1: 39/100) and left side (T0: 9/100 vs T1: 28/100), Figure 2a. Similar results were provided by the MRC scale, showing an increase in strength at the shoulder (2/5 at T0 vs 3/5 at T1), elbow (0/5 at T0 vs 2/5 at T1) and wrist flexors (0/5 at T0 vs 2/5) (Figure 2b). Moreover, we showed a partial increase in trunk control especially as regards the sitting balance as showed by Trunk Control Test (T0: 12/100 vs T1: 25/100), Figure 2c. Indeed, the patient showed improvement in the ability to maintain a seated position. Finally, overall functional abilities were improved after rehabilitation training, as evidenced by Barthel Index score (T0: 10/100 vs T1: 35/100), Figure 2d. In this regard, the improvements involved feeding capacity, bowel and bladder dysfunction and postural transfers ability.

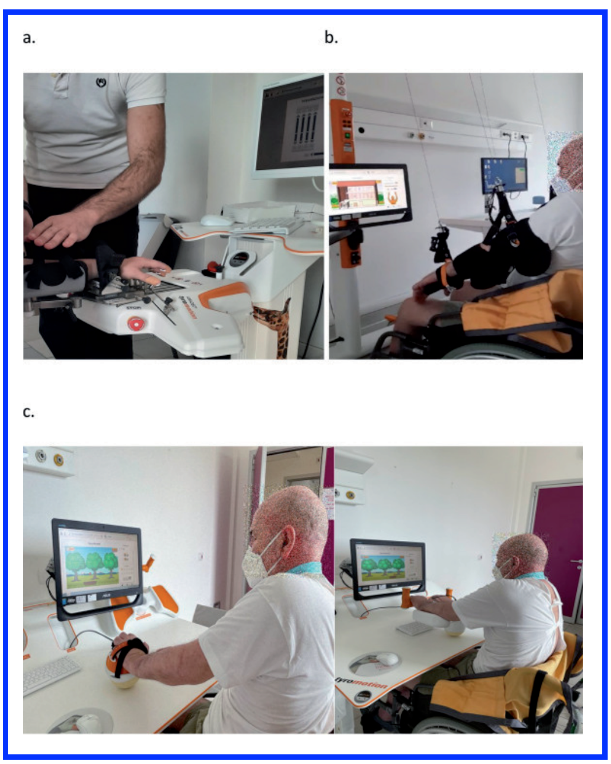

Figure 1. *Multidimensional robotic treatment. The array includes two robotic devices, AMADEO and DIEGO, and a sensor-based system, PABLO (Fondazione Carlo Gnocchi): a) a robotic device for the treatment of the hand (AMADEO, Tyromotion), b) a robotic arm-shoulder therapy system (DIEGO, Tyromotion); c) a sensor-based therapy system for the entire upper limb and truk (PABLO, Tyromoyion).*

Discussion

We described the case of a middle-aged man who was diagnosed with axonal polyneuropathy with prevalent impairment of motor component and preserved sensory function. We proposed a comprehensive rehabilitation treatment, including both conventional and robotic multidimensional training. This case report evidenced the feasibility and efficacy of such an approach in the post-acute phase of GBS disease. Despite the residual global impairment, the comprehensive treatment, specifically tailored to the patient's competencies and progress, was feasible, and promoted improvements in functional abilities.

The main findings of our study are related to the improvements evidenced in clinical and functional scales. Our motor function measure assessed using the MI was able to track improvements in upper limb abilities after training intervention. Currently, it is known that this test gives an overall indication of patient's limb impairment. Of note, literature evidences a strong correlation between motor function assessed by MI and activity function measured by BI in post-stroke hemiplegic patients. Indeed, it has been shown that the less severe the mobility impairment the greater the patient's Activities of Daily Living (ADLs) function.[15] According to current literature, this result is quite important, considering that not all GBS patients recover completely within one year. Indeed, for GBS survivors, the time-frame of maximum recovery has been observed within the first year after the disease,[2] with the best recovery achievements occurring during the first six months. In this case, after about nine months from the onset of the disease, we have shown an increase in MI values by twice the minimum detectable change after training, thus suggesting that the trajectory of recovery has indeed been positively influenced by our comprehensive rehabilitation training.[16] Besides improvements in upper limb abilities, our patient experienced the effects of treatment also on trunk control, as shown by changes in the TCT test scores. The TCT is widely used in the clinical setting to evaluate a patient's ability to turn to either side of the bed and get up, and it is an important predictive feature of functional recovery in stroke patients.[14] Moreover, this test is a reliable and early predictor of ADLs, suggesting a positive correlation with future disabilities at hospital discharge.[17] For these reasons,

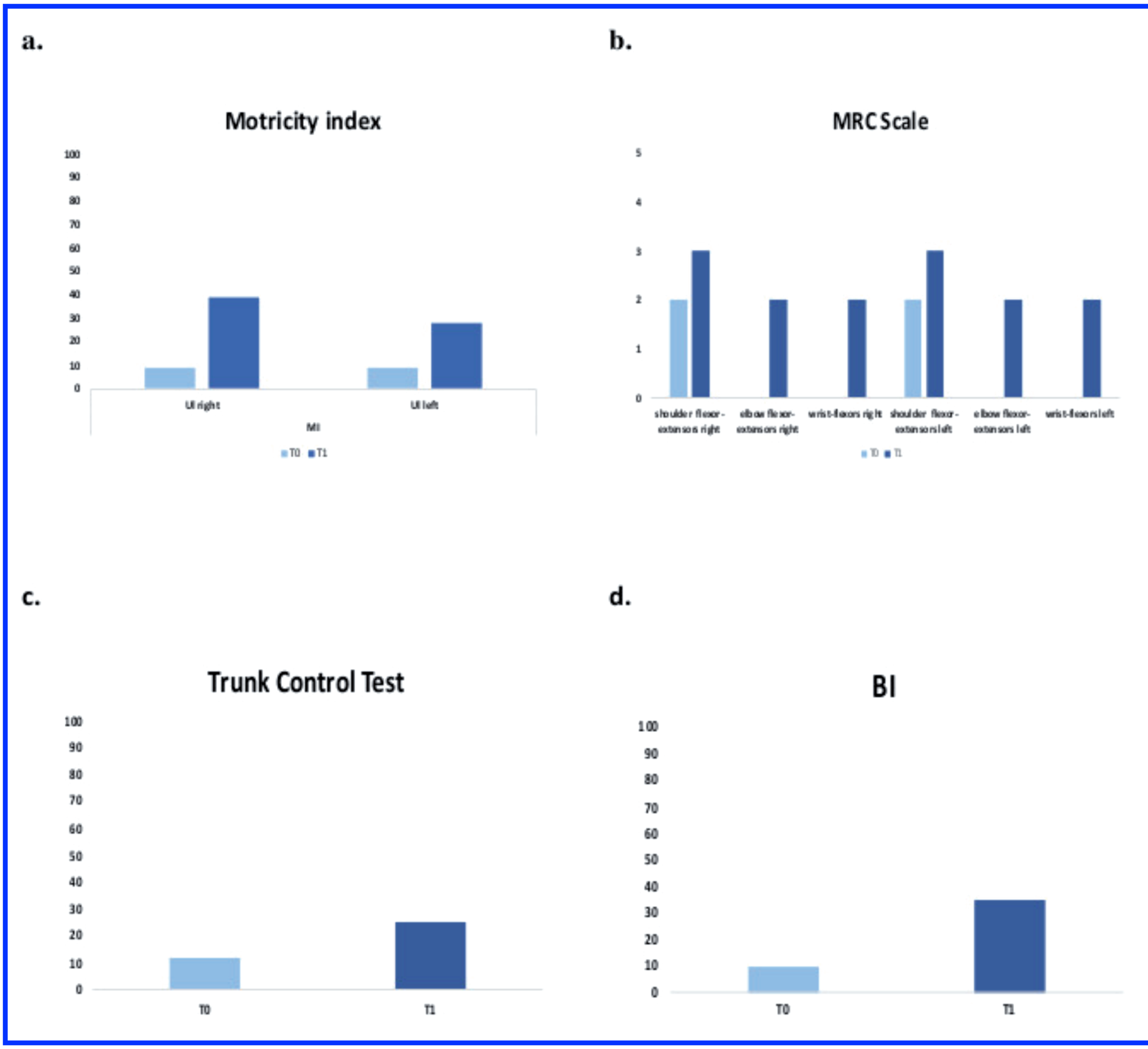

Figure 2. *Functional and disability scales before (T0) and after (TT) training protocol. MI, Motricity Index; MRC scale, Medical Research Council Scale; BI, Barthel Index, Ul, Upper Limb.*

what is remarkable is the fact that after attending our comprehensive rehabilitation protocol, our patient experienced an improvement in upper limb abilities and trunk control, likely impacting his possibility of future disability. Furthermore, another aspect to be noted is a slight increase in upper limb strength after the training period, as evidenced by segmental manual muscle testing (MRC scale). In this regard, recent studies pointed out the prognostic value of assessing skeletal muscle strength in GBS. Indeed, more severe weakness with consequent low MRC score on admission appears a reliable predictor of outcome at 6 months, and probably later.[18] According to this evidence, our patient presented an important motor impairment as a *sequela* of GBS infection disease, and, despite the overall aforementioned improvement, his motor impairment remained of high severity. Another stimulating result is represented by evidence of benefits not only on motor impairments, but also in the limitations observed in ADL. In fact, our patient improved his global ADL performance, as demonstrated by Barthel Index changes form T0 to T1, both statistically and clinically significant.[14] Limitations in the ability to perform ADL increase the susceptibility to care dependency, thus reducing the quality of life of both patients and caregivers. Indeed, the impact of GBS on ADL, social activities and health-related quality of life is extensive, and it can persist even two years after the onset of the disease, despite significant improvements over time.[19] Previous studies have shown a close correlation between MI score, TCT score and Barthel Index score. Then, the relevance of these data lies in the evidence that our comprehensive rehabilitation protocol was

associated to relevant benefits on functional capacities and activities of daily living, indicating a virtuous circle between physical function and disability status.[8]
Our data are in line with recent literature describing the positive influence of rehabilitation treatments on functional measures and health-related QoL scores in GBS survivors.[4,8,20] Recent reviews underline the importance of approaching a rehabilitation intervention as a Multidisciplinary (MD) rehabilitation care, seen as a coordinated interdisciplinary team approach, under medical supervision, which aims to limit patient symptoms, enhance functional independence and maximize participation, as defined by the International Classification of Functioning, Disability and Health.[8] MD rehabilitation is designed to be patient-centred, time-based, and functionally-oriented and aims to maximize activity and social integration using a biopsychosocial model. Existing clinical guidelines for GBS recommend comprehensive, flexible coordinated MD care, usually tailored to suit an individual's needs, and appropriate follow-up, education and support for patients (and caregivers).[8] In this regard, recent reviews support the importance of improved disability in the short term (less than 6 months) with high-intensity inpatient multidisciplinary rehabilitation. Overall, various types of exercise programmes improve physical outcomes such as functional mobility, cardio-pulmonary function, muscle strength, and work rate and reduce fatigue in GBS, although the low quality of evidence of most studies decreases their external validity.[8,20] Cycling training seems the most warranted type of program, although strengthening exercises and endurance training, including gait training, can also target physical outcomes. Anyway, for optimal recovery, a two-phase rehabilitation process should take place: the first in the early stages of recovery to reduce the disability burden and the second in the later stages of the disease to support reconditioning.[20]
As regards the latter aspect, less information is available on the impact of rehabilitation in the chronic phases of the disease. However, evidence promotes continued high-intensity and progressive strength, endurance, and functional skill training.[7,8,20] Given these premises, we suggest that a comprehensive rehabilitation intervention, including both conventional and robotic multidimensional training, may be feasible and effective in the post-acute phase of GBS disease. Despite the residual global impairment, the comprehensive treatment, specifically tailored on the patient's competencies and progress, promoted improvements in functional abilities and disability status. As regards this point, we have to acknowledge that the functional gains were influenced by the combination of both training approaches. Therefore, we may not disregard the effects of the constant conventional training on overall improvements. Anyway, an important novelty of our study is represented by the use of robotic treatment, tailored to the patient's competencies and functions under the supervision of a physiotherapist. In particular, the different robotic devices involved proximal and distal upper limbs reinforcement and trunk control movement: the task-oriented repetitions in a motivating therapeutic gaming environment may have increased the gains in the ability to maintain a seated position and in abilities involving upper limb function and trunk control. Therefore, in addition to documenting the feasibility of our training program on outpatient services, the present study showed that such an approach might be a valuable treatment option in the post-acute phase of GBS disease. This training protocol should be tested out in future large sample randomized controlled trials compared to usual care, and including a follow-up assessment, to confirm the potential utility of the presented approach.

Conclusions

This study evidenced the feasibility of a multidimensional robotic therapeutic approach associated with conventional treatment in the post-acute phase of Guillain-Barrè Syndrome. Before and after asesessments revealed clinically significant improvements associated to this approach on motor recovery and global disability status.

Authors' contributions

CT performed functional evaluations, contributed to data collection, analysis and interpreted the results. BG contributed to data collection, analysis and interpreted the results. PG and SG contributed to data collection. MG, FC, IA and PB contributed to manuscript supervision. All authors read and approved the final version of the manuscript.

Patient consent for publication

The patient gave their written consent to use their personal data for the publication of this case report and any accompanying images.

Availability of data and materials

All data underlying the findings are fully available.

Conflict of interest

The authors declare to have no conflict of interest, since they did not receive any funding for the present research.

Ethics approval and consent to participate

No ethical committee approval was required for this case report by the Department, because of the use of routinary functional tests and scales, which are commonly performed in actual clinical practice. Informed consent was obtained from the patient included in this study.

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