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CASE REPORT

Effects of dance-based movement therapy on balance, gait, and psychological functions in severe cerebellar ataxia: A case study

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ABSTRACT

Purpose: Individuals in the later stages of cerebellar ataxia usually experience serious balance and immobility problems. Currently, there is a lack of adequate rehabilitative programs for individuals with severe cerebellar ataxia that can help improve ataxia-related motor impairment. The purpose of the present study was to explore the potential physiotherapeutic benefits of partnered dance on balance, motor functions, and psychological well-being in an individual demonstrating severe cerebellar ataxia symptoms. **Methods**: The individual was a 39-year-old male diagnosed with cerebellar atrophy. He had the disease for more than 15 years prior to the study. The individual attended 24 intervention sessions over an 8-week period of dance-based movement training that aimed to improve his balance and postural stability by facilitating the perception and control of static and dynamic balance movements and body alignment. **Results**: The individual demonstrated improvements in independent standing balance, gait characteristics, and functional mobility. In addition, improvements in self-reported depression and quality of life scores were observed after completion of the intervention. **Conclusion**: Although interpreting the findings of this study is limited to a single participant, partnered dance could be a suitable alternative physiotherapeutic intervention method for people with severely impaired mobility due to cerebellar dysfunction.

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Introduction

Cerebellar ataxia is a neurological symptom associated with damage to the cerebellum, and/or its afferent and efferent connections. It is caused by either focal cerebellar lesions (e.g. stroke and benign tumor) or degenerative diseases. Individuals with cerebellar ataxia typically experience the deterioration of motor control affecting body balance and limb coordination. In the case of degenerative cerebellar ataxia, these motor symptoms worsen over time because of the progressive nature of the disease. Individuals in the later stages of the disease are usually confined to a wheelchair or bed, a result of their immobility (Marsden and Harris, 2011; Morton et al, 2010). This motor impairment frequently affects the independence of patients in terms of their daily activities and their quality of life (QOL) (Schmitz-Hübsch et al, 2010).

Currently, there are no medical treatments available to cure or modify the disease. Nevertheless, recent studies on the development of effective therapeutic interventions have indicated that physiotherapeutic treatments can have beneficial effects on the recovery of motor capacity in individuals with cerebellar ataxia (Burciu et al, 2013; Ilg et al, 2014). For example, rehabilitation studies targeting ataxic impairments of balance and gait have suggested that staged exercise-mediated trainings with various balance and coordinative movement tasks are effective at reducing cerebellar ataxia-related motor dysfunction (Balliett, Harbst, Kim, and Stewart, 1987; Ilg et al, 2009). However, it remains to be seen whether the application of physiotherapeutic interventions is helpful for individuals with severe cerebellar ataxia for whom free standing and walking are restricted (i.e. non-ambulatory patients).

Few studies have investigated rehabilitative programs for advanced-stage individuals with cerebellar ataxia. Locomotor training on a treadmill equipped with a body-weight support system has been used to improve functional independence in walking for non-ambulatory individuals with cerebellar ataxia (Cernak, Stevens, Price, and Shumway-Cook, 2008). This training equipment allows repetitive stepping movements with reduced postural control demands, but this training method needs to be utilized with caution because the use of a harness for body-weight suspension and support may hinder the improvement of balance component of locomotion (Dobkin and Duncan, 2012). Thus it may not be effective

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for people with balance-related impairments such as individuals with cerebellar ataxia. Synofzik et al. (2013) found that the use of interactive virtual video game technology could be effective in facilitating posture, gait, and balance control in sitting and standing in advanced degenerative ataxia. This training strategy helps patients to perform whole-body coordination and balance exercises with greater enjoyment affording home practice, which may offer the benefits of increased levels of motivation and adherence to the training. Nevertheless, attention must be paid to the use of compensatory strategies (e.g. reduced degrees of freedom or range of motion of the joints) that may be adopted by the performer during the accomplishment of a task. When maladaptive control strategies are developed, these compensatory techniques can interfere with the recovery of movement patterns and may limit the potential of patients. Thus, appropriate instructions and guidance need to be provided under the supervision of physiotherapists during video game-based intervention (Synofzik and Ilg, 2014).

When determining the effective treatment program for a specific form of impairments in movement-related functions in advanced cerebellar ataxia, a suitable training program may be dance or dance-based movement therapy. In essence, dance-based movement involves balance and coordinative functions associated with the step-by-step control of goal-directed movements (Earhart, 2009). Since balance and posture impairment is typically exacerbated in severe cerebellar ataxia, a form of partnered dance can provide physical support and guidance, enabling the patient to function more easily and challenge their limits of stability during training (Hackney and Earhart, 2010). When partnered with a trainer or therapist, individual supervision and direction regarding the desired stepping kinematics and posture can be provided to the patient. In addition, the trainer can adjust the amount of hand-held support so that the dependency-producing effects of the partner as a supportive aid can be reduced when appropriate. Moreover, the incorporation of music into a dance program and access to social interaction (e.g. partnership and peer support) may increase enjoyment of and engagement in the exercise, which not only improves physical condition and mobility, but may also enhance the emotional state and QOL of the patient (Hackney and Earhart, 2009).

Although dance-based movement as a rehabilitation approach appears to offer suitable training components to overcome limitations in activities arising from cerebellar dysfunction, no study has yet assessed its potential therapeutic benefits for individuals with cerebellar ataxia, particularly those who suffer severe balance and mobility problems. The aim of the present study is to explore whether dance-based movement training improves ataxia-related motor symptoms and the QOL of an individual with advanced cerebellar ataxia.

Case description

Participant background and examination

The individual was a 39-year-old male (height: 175 cm, weight: 73 kg) living with his father and sister. He was diagnosed with *spinocerebellar ataxia type 2 (SCA 2) as confirmed by his neurologist using clinical examinations, MRI records, and genetic testing at the age of 28. Ataxic symptoms began when he was in his early 20s, which worsened over time. Following the progressive declines in gait and stance functions as well as impaired limb coordination, he became wheelchair-bound at the age of 36. At the time of this case he demonstrated severe balance problems and was not able to stand independently, requiring the assistance of two people to walk. He used grab bars installed on a wall to ambulate within the home. He previously received no rehabilitative treatment.

The individual underwent the Mini Mental State Examination to evaluate his cognitive status and scored normally (≥25). The severity of his cerebellar symptoms was evaluated using the International Cooperative Ataxia Rating Scale (ICARS) (Trouillas et al, 1997) and the Scale for the assessment and rating of ataxia (SARA) (Schmitz-Hübsch et al, 2006). He had coordination difficulty in finger chase, nose-finger touch, fast alternating hand movement, and heel-shin slide tests, along with mild dysarthria and writing problems. He exhibited no extracerebellar signs (e.g. weakness or tremors) and had no other clinically significant orthopedic, visual, or psychiatric dysfunction. Daily activities included stretching exercises, doing sit-ups, playing computer games, and helping the light household tasks while sitting or lying on the floor. He was on no medications at the time of participation in the study. Informed consent was obtained from the individual in accordance with the declaration of Helsinki.

Design and outcome measures

The basic design of this research was a single-subject case study, type A-B-A, of an individual with cerebellar ataxia. Phase A was a pre-test in which outcome measures were conducted a week prior to the initiation of the intervention. Phase B represented a post-test performed after the end of the training program. The second Phase A was a follow-up test assessed 4 weeks after the completion of the training. Each assessment included clinical outcome measures with additional kinematic evaluation of balance and gait performance taken every 2 weeks during the intervention period.

Gait performance data were collected from an OPTITRAK 3D optoelectronic camera system in the laboratory (NaturalPoint Inc., Oregon, USA) with a sampling rate of 120 Hz. The individual was asked to walk along a 6-m long pathway at his preferred speed with manual assistance from the trainers. The following outcome variables for balance and gait performance were collected: step width, step length, relative double-support time, and balance time. Step width was calculated as the horizontal distance between the toe marker of one foot to the toe marker of the opposite foot, step length was defined as the sagittal distance from one heel strike to the next heel strike, and relative double-support time was determined as the time spent with both feet in contact with the ground expressed as a percentage of the total stride time. Balance time was measured as the time spent in maintaining balance independently in the standing position with the eyes open. The criteria for minimal detectable change (MDC) for gait parameters have not been established in individuals with cerebellar ataxia.

The individual then underwent subjective measures of mobility and psychological function. Since there are no assessment tools specifically designed to measure balance confidence in cerebellar ataxia, we chose some standardized clinical outcome measures frequently used to document self-perception of balance capacity. We characterized balance confidence using the Berg Balance Scale (BBS) and the Fall Efficacy Scale (FES). The BBS evaluates the balance performance in 14 functional tasks that are common in daily activity (Berg, Wood-Dauphinee, and Willizma, 1995). Although this scale has not been validated for use with the cerebellar ataxia population, it has been shown to be a good indicator of balance confidence level in other neurological papulations (Newstead, Hinman, and Tomberlin, 2005; Whitney, Wrisley, and Furman, 2003). The maximum score is 56, with higher scores indicating greater balance. The MDC for the BBS was not established in cerebellar ataxia, but reported to be 2.5 points in chronic stroke (Liston and Brouwer, 1996). The FES measures perceived fear of falling while performing 10 activities of daily living with a total possible score of 100 (perfect balance confidence) (Tinetti, Richman, and Powell, 1990). This scale was chosen for its ability to measure perceived body stability as fear of falling is common in individuals with cerebellar ataxia (van de Warrenburg et al, 2005). The MDC for the FES has not been established in the literature. The Barthel Index is a measure of the functional independence of a participant from nursing care in basic daily activities, testing 10 items. The highest attainable score is 100, indicating total self-care functional ability (Shah, Vanclay, and Cooper, 1989). The BI was devised for use in rehabilitation for individuals with stroke and other neurological deficits (Hobart et al, 2001; Wade and Collin, 1988). The MDC for the Barthel Index (BI) was not established in cerebellar ataxia, but reported to be 4.02 points in chronic stroke (Hsieh et al, 2007). The individual also completed the Beck Depression Index (BDI), a 21-item self-rating questionnaire, to evaluate the symptoms of clinical depression, with lower scores indicating less severe depression (Beck et al, 1961). The BDI is one of the most widely used tests for assessing the severity of depression. Although this measure has not been validated in individuals with cerebellar ataxia, the BDI was included in outcome variables because severe illness is often accompanied by depression (Miller et al, 2010). The MDC for the BDI has not been established in the literature. Perceived QOL was measured using the short version of the health-related QOL, a 26-item survey that assesses physical health, mental health, social relationships, and environment (WHOQOL Group, 1998). This measure is one of the few validated clinical measures for individuals with cerebellar ataxia (Schmitz-Hübsch et al, 2010). Higher scores on the QOL reflect a better QOL. The MDC for the QOL has not been reported in the literature.

Intervention

The dance protocol intervention included the basic steps and simplest movements of the tango, modified to accommodate cerebellar ataxia. The main aim of the treatment was to improve balance and postural stability by facilitating the perception and control of static and dynamic balance movement and body alignment. The dance program was led by a certified dance instructor in a progressive manner with the help of one or two trained assistants. The instructor had extensive knowledge and experience in mobility problems associated with cerebellar ataxia. The instructor served as a dance partner, providing physical support and close supervision during training. Initially the individual practiced static balance while standing upright with partner's hand-held support. The individual was asked to stand upright with his body weight evenly distributed on both feet and to hold a stable stance. He was then instructed to slowly shift his body weight laterally onto one leg while maintaining an upright trunk and limb alignment. Once proper weight bearing and balance on one leg had been accomplished, this was repeated for the other leg. After this weight transfer task side by side, the individual practiced a body weight shift in the mid-stance position of the gait (i.e. one leg forward and the other leg behind). The instructor monitored and directed the individual's attention in controlling the location of his body center and his posture during the static balance tasks.

Following approximately 20 minutes of static balance practice, the remaining 30 minutes of the intervention were dedicated to basic tango step activities. Dance movements included multi-directional steps, turns, and rocking actions, with an emphasis on a full shift of the center of mass and the maintenance of balance and postural alignment over the new base of support, particularly during the single-limb stance position. The partner's support in a closed-practice arm position (i.e. holding elbows while facing each other) was provided to guide the step movements as they were performed in a slow and rhythmic manner so that the individual could develop a conscious awareness of adequate balance and postural control. The speed of movement was gradually increased to synchronize with the tango music played in the background as progressed. The restoration of body stabilization was emphasized over any other activity in the case of trunk sway or the loss of balance during practice. The dance program took place in his living room on a hard-surfaced floor (6 m × 4 m) because of restrictions in his mobility and transportation.

Outcomes

The individual completed all 24 intervention sessions over an 8-week period of training. His clinical measures for ICARS (60 points with a maximum possible score of 100) and SARA (28 points with a maximum possible score f 40) did not change across the three assessment periods (i.e. the pre-, post- and follow-up tests). Thus, his overall ataxia symptoms remained stable over the study period.

The kinematic measurement of balance and gait performance obtained during each of the assessment sessions are presented in Figure 1. The individual improved for nearly all kinematic measures over the course of the intervention and the improvements were highest for the post-intervention evaluation. However, many of the gains were lost by the time the follow-up evaluation conducted 1 month after the intervention session. Prior to training, the individual's independent balance time was 0.56 seconds and he was virtually unable to maintain standing balance. He improved his standing balance over the

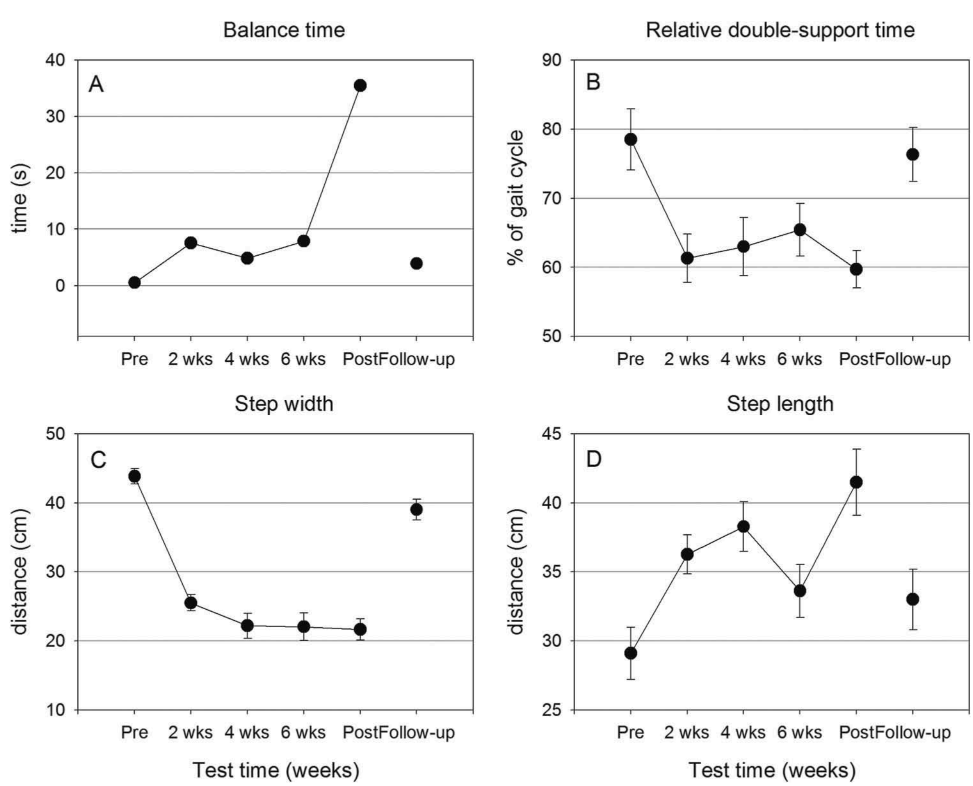

Figure 1. Plots of standing balance time, relative double-support time, step width, and step length across the assessment period.

training period from 4.87 seconds at 2 weeks up to 7.89 seconds at 6 weeks. In the post-intervention assessment, his balance time improved to 35.47 seconds, but then was markedly decreased to 3.90 seconds in the follow-up assessment 1 month after cessation of the intervention (Figure 1A). The relative value of double-support time was 78.58% prior to training. His relative double-support time was decreased and maintained over the 8-week training period (61.31–65.42%). After completing the intervention, his relative double-support time was 59.78%, but then was increased to 76.37% in the follow-up testing (Figure 1B). A gradual decrease in step width was observed during the training period. His step width was 43.82 cm in the first assessment, and was decreased up to 22.05 cm at 6 weeks. In the post-intervention evaluation, step width was 21.66 cm. However, gains in step width were not maintained and increased to 39.01 cm in the follow-up (Figure 1C). The individual demonstrated continued improvement in step length with some fluctuations from the baseline to the post-intervention period. His step length was 29.10 cm in the initial assessment, was increased to 33.61 cm at 6 weeks. In the post-intervention measurement, step length was further increased to 41.48 cm. However, most of the gains in step length were lost and decreased to 33.20 cm by the time of the follow-up testing (Figure 1D).

The scores for the self-reported functional measurements are shown in Table 1. In addition to the improvements in kinematic performance, the individual improved in nearly all functional measures over the 8-week training period, with some of these gains retained at the time of the 1-month follow-up. His BBS score improved by 2 points from pre- to post-training. He scored 5/56 prior to training, and 7/56 after the intervention on the BBS. However, this gain was not maintained and he scored 5/56 in the follow-up testing. His FES score improved by 8 points from pre- to post-training. His pre-test score was 22/100 and post-test score was 30/100 on the FES. The FES score was further increased to 31/100 over the 1-month follow-up period. There was an improvement in the Barthel Index of 7 points from pre- to post-training. He scored 27/100 and 34/100 on the BI in the first and post-intervention assessments, respectively. The post-test score was maintained during the 1-month follow-up period. The individual demonstrated a marked reduction in his BDI score of 29 points from pre- to post-training. He scored 37/63 prior to training, and 8/63 after the training on the BDI. Some of the improvement was lost and he scored 11/63 on the BDI over the 1-month follow-up period. He also demonstrated an improvement in the QOL score of 24 points from pre- to post-training. His QOL scores increased from 63/130 prior to training to 87/130 after the training. The QOL score was further increased to 99/130 over the 1-month follow-up period.

Discussion

The present study examined the effects of partnered dance intervention on balance and mobility functions in an individual suffering from severe cerebellar ataxia. After 8 weeks of 1-hour partnered tango dance therapy three times per week, the individual demonstrated improvements in independent standing balance, gait characteristics, and functional mobility. In addition, improvements in self-reported depression and QOL scores were observed after the completion of the intervention, indicating increased psychological well-being. While most of the gains in balance and gait capacities were not retained in the 1-month follow-up evaluation, some of the gains in self-reported functional mobility and psychological benefits persisted after the cessation of the program.

The beneficial effects of the tango dance on balance and gait in the individual with severe degenerative cerebellar ataxia may be due to the training components adopted in the current intervention program. One of main symptoms of cerebellar ataxia is a difficulty in maintenance of body balance while standing. Such postural instability may necessitate safety strategies in gait such as wide-base walking with reduced step length (Hudson and Krebs, 2000). Tango involves weight shifting movements that emphasize maintaining an upright posture and balance with performing coordinated stepping movements (Earhart, 2009). Participants must focus on the continuous control of balance and step-by-step adjustment of steps while dancing. These aspects of the tango require a more functional and complex task performance than

Table 1. Intervention outcomes for balance confidence, activity of daily living, depression, and quality of life.

Outcome measures	Pre-training	Post-training	Follow-up	Change (%) pre–post-training
Berg Balance Scale	5/56	7/56	5/56	40
Fall Efficacy	22/100	30/100	31/100	36.3
Barthel Index	27/100	34/100	34/100	25.9
Beck Depression	37/63	8/63	11/63	−78.3
Quality of Life	63/130	87/130	99/130	38.1

traditional therapeutic exercise programs, and may help to improve balance and gait characteristics of the individual. The functional features of movement patterns in the tango more likely target the goals of physiotherapies for rehabilitation in cerebellar ataxia. There is strong evidence that task-specific training can have more beneficial effects on functional recovery than nonspecific rehabilitation training in clinical practice (Bayona, Bitensky, Salter, and Teasell, 2005).

Second, feedback and physical guidance were given to the individual in an explicit way in order to increase conscious attention to movement quality and practice performance. Since implicit learning and automaticity in motor skills were known to be disturbed in individuals with cerebellar dysfunction (Lang and Bastian, 2002), we assumed that the explicit strategy might help patients to be aware of their actual state of movement and allow them to modify their actions when necessary. Consistent with this notion, Taylor, Klemfuss, and Ivry (2010) reported that using such treatment strategy could be beneficial for sensorimotor learning in individuals with cerebellar diseases.

Third, the role of the partner may also be of great importance in the rehabilitation treatment of individuals with severe ataxia. As the individual was unable to maintain standing balance, the physical support provided by the partner might help the individual to more dynamically control their body balance and learn dance movements more quickly than without one (Hackney and Earhart, 2010). In addition, the partner monitored and coached proper trunk alignment and weight shifting with limb kinematics during the intervention. This direct training and supervision by the partner could prevent the development of compensatory or incorrect movement patterns. The partner also adjusted the amount of manual assistance as the training progressed to encourage independent balance and mobility functions while reducing dependency-producing effects of the partner.

We postulated that these training components and strategies incorporated in the dance program might yield specific effects on improvements in balance and gait functions in the individual with severe ataxia. However, these motor gains were not maintained after the cessation of the intervention program and declined nearly to the baseline level. The reason for this attenuation of functional status is not fully clear, but we believe it may be attributed largely to the natural disease progression and decreased physical activity at home after the intervention. Studies regarding natural course of degenerative cerebellar diseases indicated that ataxic symptoms were progressively worsening over time and the mean annual increase of the SARA points was 1.61 in individuals with cerebellar ataxia type 3 without any training (Jacobi et al, 2011). Miyai et al. (2012) noted that functional improvements achieved by training were less sustained in individuals with advanced cerebellar neurodegeneration. In addition, the individual reported a substantial reduction in physical activity at home after the intervention period. He reported feeling slightly depressed and less motivated to do physical activity/exercise after removal of the intervention. Therefore, continuous rehabilitative training seems crucial for the retention of the physiotherapeutic benefits in individuals with severe cerebellar ataxia.

The dance intervention also had a positive effect on depression and the QOL of the individual. The beneficial effects of dance programs on psychological well-being were noted in other studies using different neurological patients (e.g. Parkinson's disease and stroke) (Hackney and Earhart, 2009; Hackney, Hall, Echt, and Wolf, 2012). Depression is a common comorbidity in individuals with cerebellar ataxia, and more severe cases of the disease are thought to be associated with a higher risk of depression (Schmitz-Hübsch et al, 2011). It is also thought to have a negative impact on the perception of the patients' functional status and QOL. Previous work showed that dance-based therapy programs were particularly beneficial for psychological well-being (Earhart, 2009) probably due to the inherent social and supportive nature of the dance program, including the perceived benefits of functional mobility and balance. In the present study, the 8-week dance training program had a positive and lasting effect on the individual's QOL, even in the later stages of cerebellar degeneration. In informal exit interviews, the individual mentioned enjoyment and an interest in continuing the program, which may promote adherence to the program over the long term.

Conclusion

The present study demonstrated the beneficial effects of dance-based movement training on the rehabilitation of motoric and psychological function in an individual with severe cerebellar ataxia. Although the findings of this study are limited to a single participant, the tango as a form of partnered dance may be a suitable alternative method of physiotherapeutic intervention for individuals with severely impaired mobility due to cerebellar dysfunction. This is because various components of the tango allow for the activation of postural and neural control mechanisms relevant to the rehabilitation of ataxia-related disabilities. Future studies should attempt to elucidate the pathophysiological mechanisms underlying the beneficial effects of dance-based movement training. There is also a need for studies with a larger sample of patients and longer-term intervention to improve the clinical feasibility of dance training programs for individuals with cerebellar ataxia.

Declaration of interest

The authors declare no conflicts of interest.

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