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Case Report

Effects of Intensive Coordination Training While Walking In Parallel Bars with Visual Feedback in a Case of Spinocerebellar Ataxia Type I: A Case Report

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ABSTRACT

Background: This study attempts to explore the effect of 3 months long intensive bi-manual coordination training, through visual and auditory bio-feedback, along with concurrent gait training on parallel bars, on the symptoms of ataxia in a 18 year old case of Spinocerebellar ataxia Type-I. The patient was 18 year old female, with complaint of difficulty in walking in a straight line and reaching targets with her hands.
Method: Coordination training in the form of task oriented bimanual reaching activities for training of the upper extremities; along with gait training on parallel bars in front of a mirror was implemented thrice a week, for 3 months. The patient was asked to perform the same activities at home, each day for 1 hour during these 3 months. Primary outcome was assessed by symptom evaluation using "International Cooperative for Ataxia Rating Scale". Secondary outcome was assessment of mobility, derived from "Performance Oriented Mobility Assessment". Both outcomes measures showed significant improvement by the end of 3 months intervention.
Conclusion: It may therefore be concluded that "intensive coordination training for upper extremity reaching activities along with balance and gait training on parallel bars supplemented with mirror and verbal feedback" improved symptoms associated with spinocerebellar ataxia.
Key words: Spinocerebellar Ataxia, International Cooperative of Ataxia Rating Scale, Performance Oriented Mobility Assessment, Motor Control Theory, Biomechanical Ankle Platform System board.

INTRODUCTION

Spino-Cerebellar Ataxia (SCA) Type I is one of the subtypes of Autosomal Dominant Cerebellar Ataxias, which is a neurodegenerative condition characterized by degeneration of cerebellum, affecting its afferent and efferent connections. [1]

Though the prevalence of Spino-Cerebellar ataxia is about three cases per 100,000, Saleem et al (2000) concluded that among Indian population, SCA Type 1 is prevalent in less than 5%. [2]

Koeppen et al (2005) stated that the ataxia cause lesions in the brain invariably affect the "cerebellar module" that is defined as a reciprocal circuitry between the cerebellar cortex, the dentate neuclei and the inferior olivary nuclei. [3] Sasaki et al (1996) found a pan-cerebellar syndrome; with ataxia of gait, stance, and limbs, dysarthria and Oculomotor abnormalities like gaze evoked nystagmus, saccadic smooth pursuit, impaired visual suppression of the vestibulo-ocular reflex, and decreased optokinetic nystagmus. Pyramidal signs like spasticity, hyperreflexia, and extensor plantar responses are common. Amyotrophic changes and sensory loss may also occur. [4]

Recent years has seen a growing part of evidence from the field of neurophysiology that has advanced the understanding of function and dysfunction of the cerebellum in motor control and motor learning. [5-7] Building on this work, theories have developed about possible responses of the cerebellum at a neural level (neuroplasticity) to respond to rehabilitation interventions. [8-12]

In most cases conducted by several authors, interventions included proprioceptive neuromuscular facilitation, Frenkel's exercises, dynamic postural stability training during activity, muscle strengthening and flexibility exercises, balance reeducation and gait training. [13-15]

Jeka (1997) reviewed a series of studies on postural control, using light touch feedback through contact of fingertips or a walking aid as a means of support. Clinical observation suggests that some individuals with ataxia may find light touch more useful as a feedback strategy rather than a conventional walking aid. [16]

The effect of weighting the axial skeleton was studied by Folz and Sinali (1995) who reported subjective improvements in posture, gait and feelings of steadiness. Conversely Clopton et al (2003) reported gait characteristics changed unpredictably with axial loading, worsening more often than improving. [17,18]

The current case report evaluated the efficacy of gait and balance training on ataxic symptoms using mobility aids such as parallel bars with continuous visual and auditory feedback and thus aimed to add value to the present resources for rehabilitation of such patients.

CASE HISTORY

The patient was an 18 year old female diagnosed with genetically induced spinocerebellar ataxia type 1, age of onset being 15 years. Her primary complaint was difficulty in walking in a straight line and reaching targets with both upper and lower extremities. Patient came into the clinic walking independently with an unsteady gait pattern, suggestive of classical 'Ataxic Gait', characterized by Patients standing with feet well apart and frightened to stand due to fear of fall. Patients tend to reel to the side of unilateral lesion, or from side to side if central or bilateral (even if supported). Walking along a line of the floor demonstrates minor degrees of gait unsteadiness. [6,7]

Clinical history: Onset of symptoms was insidious; started initially with unsteadiness of hands while reaching for objects, hoarseness of voice and difficulty in writing. All symptoms worsened gradually over time.

Family history: Reportedly other family members of the patient had similar symptoms, her father being affected in fifth decade and her elder brother in second decade of life. She was diagnosed to have Spino-cerebellar ataxia Type1on the basis of genetic testing and MRI.

Physical examination:

Examination of eye and speech: Oculomotor nerve testing revealed impaired dilatation of pupillary reaction to light and saccadic eye movements. Moderate dysarthria was present with mild delay in swallowing, without pharyngeal phase dysphagia.

Motor examination:

Tone and deep tendon reflex: On evaluation of muscle tone, upper and lower extremities had tone of 1 and 0 respectively on Modified Ashworth Scale and bilaterally decreased reflexes in biceps 0/4, triceps 1/4, and brachioradialis 0/4. Lower extremity showed absent deep tendon reflexes and a positive Babinski. No proximal or distal muscle weakness in the extremities was detected by Manual muscle testing.

Range of motion: Goniometric evaluation of upper extremity revealed full joint range of motion. Lower extremity assessment showed mild tightness in the hamstring and Achilles tendons bilaterally.

Sensory examination: On sensory evaluation, superficial sensations were found to be intact with mildly reduced distal perception of deep sensation like vibration in both upper and lower extremities.

Balance and coordination examination: Non-equilibrium coordination tests: There was impaired coordination in both upper extremities, elicited with non-equilibrium testing, such as finger-nose-finger maneuver and finger-pointing test. Movements were slow and unsteady, requiring moderate contact guarding. There was impaired coordination with non-equilibrium tests in lower extremities such as heel-knee-shin maneuver, alternate heel to knee and heel to toe maneuvers, with mild bilateral distal tremor was found. [19,20]

The patient was able to sit and stand without assistance, but with both arms pressed against the trunk as if trying to maintain body's center of gravity within her base of support. The base of support in standing was slightly widened, with normal body sway.

Equilibrium coordination tests revealed moderate impairment during single limb loading. While standing on one leg, the patient tilted the trunk laterally on load bearing side, with moderate unsteadiness requiring moderate contact guarding. [18]

Functional evaluation: On analysis of functional activities, patient was independent in all self care and was able to walk indoors without any assistance, but was unable to walk for longer distances in the community. The patient exhibited mildly impaired coordination in leg movements, resulting in abnormal and variable swing trajectories and foot placement, resulting in increased variability in length and timing of steps, slow walking speed, and an increased base of support during gait.

Mobility: Walking showed slowness and mild to moderate unsteadiness which was more prevalent with narrowed base of support. Although the patient was able to reach targets, the step length and base of support varied widely with every step, with wobbling of trunk. Tandem standing and walking was severely impaired, the patient being able to initiate with difficulty but unable to complete the movement due to fear of fall, needing maximal contact guarding. Ankle and hip strategy of balance was significantly impaired, as the patient showed dominance of stepping strategy even on mild perturbation.

Medications: The patient was on long term medication with Idebenone 45m.g and Amantadine 100mg, taken twice daily, since one year.

OUTCOMES: The primary outcome measure used in this study was International Cooperative for Ataxia Rating Scale (ICARS) which has been approved as a valid measure of the disease severity in spinocerebellar ataxias. The semi quantitative 100-point ICARS consists of 19 items divided into four unequally weighted sub-scores: posture and gait disturbances (7 items; 34 points), limb kinetic functions (7 items; 52 points), speech disorders (2 items; 8 points), and Oculomotor disorders (3 items; 6 points). The scale defines and semi quantitatively scores many of the classic clinical signs of the ataxic syndrome and, therefore, has high face validity for neurologists. On assessing the patient using ICARS, her pre-intervention score (41/100) was suggestive of stage 2 ataxia according to Klockgether and colleagues (Table1). The patients with late disease onset (>14 years of age) were predicted to decline into the disease by~1.8±0.27 points per year for the first 20 years of the disease in a recent study by G. Metz et al in 2013. On evaluating mobility, scores in performance oriented assessment of mobility –I (POMA-I) was mild to moderately impaired (table 2). [21-24]

Table 1. ICARS scores

OUTCOME	PRE-INTERVENTION	POST-INTERVENTION
ICARS:		
Posture and gait disturbance	15/34	11/34
Kinetic functions	22/52	18/52
Speech disorders	2/8	2/8
Oculomotor disorders	2/6	1/6
TOTAL SCORE	41/100	32/100

Table 2. POMA Score

OUTCOME	PRE-INTERVENTION	POST-INTERVENTION
POMA I		
Balance	11/21	14/21
Time required for bending over	3 seconds	3 seconds
Time required to rise from chair three times	17 seconds	15 seconds
Gait	4/9	5/9
Timed walk for 15 foot walkway At normal pace: As fast as feels safe:	28 seconds 27 seconds	28 seconds 26 seconds
TOTAL	15/30	19/30

Another outcome used was Performance oriented mobility assessment (POMA-I) which is a task-oriented test that measures an older adult's gait and balance abilities. The POMA-I and its subscale POMA-B (balance subscale) have adequate reliability and validity for assessing mobility in older adults as stated by Faber et al in 2006. POMA-I is useful for demonstrating intervention effects at the group level. Changes within patients, however, should be at least 5 points before being interpreted as reliable changes. [24]

Intervention:

We used facilitative strategies to minimize functional limitations and restore function.

On the basis of various electrophysiological studies of the normal function and consequences of lesions of the cerebellum by Eccles, 1969 and Fisher et al. 1965, it was suggested that the cerebellum acts as comparator between sensory input and motor output. [25,26]

Moreover, Ito in 1970, proposed that if cerebellum is damaged, movement will then be guided by slow sensory feedback loops through the cerebellum, just as in learning a new skill, and incoordination will result. [27]

In the current study we implemented coordinative training through many repetitions and proper sequencing of tasks to facilitate learning. Moreover, external cues in the form of auditory and visual feedback appear to facilitate movement by utilizing different brain areas, as stated by Jahanshahi M (1995). [28]

We additionally, implemented some of the upper extremity activities to improve the overall functional capacity of the patient. The Upper extremity activities included guided bimanual task-oriented reaching (Fig. 1). This form of intervention was selected since it is well established that bimanual, synchronous movements represent inherently stable, preferred patterns of coordination as shown by Kelso et al in1986 and Robertson in 2001. [29,30] The activities featured rhythmic reaching for a ball (with shoulder flexion from 70° to 150°) and object placement on a table (with horizontal shoulder adduction 0° to 45° and abduction from 0° to 45° while at 80° of shoulder flexion), progressing from 2 sets of 10 repetitions to 4 sets of 15 repetitions of each activities.

Coordination exercise for lower extremity, like single limb loading, side stepping, tandem stepping and braiding on parallel bars with a mirror placed at the front for biofeedback.

Dynamic balance was improved by standing with double limb support and then progressing to single limb support on Biomechanical Ankle Platform System board (BAPS board), initially with large dome and then progressing to the use of small dome on the undersurface of the multiaxial platform. The patient was supervised by the physiotherapist to prevent the fall. [31,32]

Gait training was performed manually on parallel bars for 3 months. This intervention focused on proper weight shifting, uniform step length, maintaining the line of progression, increasing step length, control of gait speed, and proper trunk and pelvis alignment during gait (10 to 12 sessions of ambulation for 10mts).

Auditory and visual feedback was provided in the form of therapist's command and mirror image respectively. (Fig.2)

Figure:1. Bimanual task oriented activity for upper extremity coordination

Figure 2: Gait training on parallel bars with visual and auditory feedback from mirror and therapist respectively.

Once static and dynamic balance on parallel bars became less assistive, progression was made to train the patient on foam/cushioned floor for increasing proprioceptive perturbation.

The patient was encouraged to maintain a stable gait as far as possible while ambulating indoors. Stretching of lower extremity (2 to 3 sets for 30 seconds) muscles, mainly for the bilateral hip extensors and ankle plantar flexors was employed to maintain optimal muscle length and augment ankle dorsiflexion during the stance phase of gait. [33,34]

Exercises for the hip included:

- bridging while using a gym ball for LE stabilization,
- wall squats while stabilizing a gym ball against a wall, and
- single limb hip flexion while positioned in supine

Exercises for trunk muscle groups included seated hip flexion and extension with a neutral lumbar spine, progressing from 2 sets of 10 repetitions to 3 sets of 10 to 12 repetitions each.

These interventions were repeated 3 times a week for 3 months. The patient was asked to perform the same interventions each day at home for 1 hour during the 3 months.

Keeping in mind the negligence in exercise often seen from the part of the patient, it was necessary to ask the patient to perform the exercises at home, to maintain adherence to the rehabilitation protocol. Such patients are worth for fatigue and thus home exercise program were carried out for the gap duration. [34,35]

We used two scales to assess the severity of ataxic symptoms pre and post intervention. Although the lack of objective data in the study is noted, some components in the POMA-I used objective data such as the timed walk test, time required to bend over and rise from chair.

The patient demonstrated reduction in International Cooperative for Ataxia Rating Scale (ICARS) score by 9 points post intervention. There is evidence that patients older than 14years progress into the disease with 1.8±0.27 points per year.

As the patient in this case has shown a reduction in 9 points with intervention of 3 months duration, this improvement is worth to be called as considerable. Initially patient was able to walk without support but with considerable staggering and difficulties in half turn. At the end of 3 months intensive coordination training, walking capability improved.

Body sway with feet together was initially severe, which reduced to moderate sway after intervention, tested with eyes open and closed. [36]

With repeated practice of bimanual task oriented training, there was improvement in kinetic functions of upper limb. Equilibrium coordination test score improved for both sides in terms of decomposition, dysmetria and intention tremor of the fingers. Although no intervention was planned for speech or Oculomotor disorders, eye movements during task oriented upper extremity training might have resulted in improvement of ocular pursuit.

Although for Performance oriented mobility assessment (POMA-I), a minimum of 5 points improvement is considered clinically relevant, the patient showed an increase in 4 points that needs to be noted. Performance oriented mobility assessment (POMA I) showed improvement in terms of tandem stance, which was not possible initially, but after 3 months of training the patient was able to perform the same with some degree of swaying and finger touch support.

Some improvement in gait was seen as the patient was able to attempt to overcome obstacles while walking, which was not possible initially. Timed walk test to cover 15 foot walkway with normal pace remained same pre and post intervention, but reduced when asked to walk with a pace as fast as possible within limits of safety, this can be a result of practiced walking in the first part of the timed walk test with normal pace.

DISCUSSION

In this study, we focused on the effect of coordination in a patient with Degenerative Spinocerebellar ataxia, probably the most difficult group of ataxias to treat, due to their progressive nature and involvement of entire cerebellum.

The results of the study showed reduction in ataxic symptoms of the patient using coordination training, measured by ICARS.

The specificity of improvements in motor performance of lower extremities is not due to strengthening of hip and leg muscles, but is influenced by increased capacities for dynamic regulations of balance. This observation is supported by a study conducted by W. llg et al (2009) providing the evidence that patients are able to improve multi joint coordination and dynamic balance by intensive and continuous physiotherapy training despite ongoing neurodegeneration. [37]

Adam's close-loop model states that, for improving motor performance, continued repetitive practice of activities, guided if required, is needed to facilitate learning and to minimize error. Improvements in motor performance can be attributed to implicit and explicit learning of skills. Intrinsic feedback is the information provided by the sensory system as a result of movement and is consistent with Gentile's notion of implicit learning. Implicit learning is not under conscious control, but the therapist can facilitate it by structuring the task and environment to support effective movement patterns. Extrinsic feedback supplements intrinsic feedback and forms the basis for explicit learning, [32] which is learning that results from clearly stated directions or instructions. [33] Verbal instructions and visual feedback in the form of mirror images are the methods used for promoting explicit learning.

Standing on BAPS board had improved outcome for balance. Studies by various authors have revealed its usefulness in improving balances and ankle proprioception. This study involved increasing the size of the hemisphere progressively on the underside of the multi-axial platform. A larger hemisphere increases the maximal angles at which the board can be tilted, which in turn can influence the quantity of lower leg muscle activation. [31,34,35]

Bilateral Upper extremity training given initially as guided movements may have caused functional carryover seen during independent bilateral or unilateral movement during task oriented reaching activities. This can be supported by Whitall et al (2000), who stated that bilateral upper extremity training may lead to unilateral improvement in performance in patients with chronic neurological dysfunction. [38]

Findings in a study by W. llg et al (2009) indicate that, improvements gained by coordination training could not be preserved for the demanding task of reactive postural control, until it is reinforced by home practice sessions. Thus a continuous intensive exercise program has prolonged beneficial effects. [37]

Lang and Bastian (2002), who suggested that cerebellar dysfunction precludes the acquisition of movement automaticity and that diverted attention can adversely affect task performance despite formalized practice. Thus, focused attention by visual and auditory cues on the part of patient may have contributed to improved upper extremity coordination. [39,40]

Buchner et al 1997 stated strength has been associated with improvement, although no weakness was found on manual muscle testing in any of the four limbs, maintenance of strength and mobility with therapeutic exercises is thought to improve the outcome. [41]

Gait training on parallel bars has modest improvements in the functional outcomes, as the parallel bars provided support and decreased the fear of fall providing stable background to practice gait.

It has been noted by Bastian AJ. (1997), that slower movements are associated with decreased ataxic symptoms during reaching tasks and that weighted movements reduce gait instability in patients with ataxia by Armutlu K.et al (2001). [40,42]

Therefore strategies for decreasing movement speed through mechanical constraints resulting from holding on to the parallel bars and visual feedback from the mirror helping the patient to slow down the speed of movement may outweigh the negative consequences of increased inertia.

CONCLUSION

Intensive coordination training for upper extremity, incorporating reaching activities with balance and gait training in parallel bars, supplemented with mirror and verbal feedback, improved symptoms associated with Spinocerebellar ataxia and function as measured by International Cooperative for Ataxia Rating Scale (ICARS) and Performance oriented mobility assessment (POMA-I).

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