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Neurorehabilitation for an individual with bilateral thalamic stroke and pre-existing visual impairment presenting with impaired use of sensory cues: A case report

BACKGROUND

Impaired balance is one of the primary causes of functional limitations in patients with stroke. Deficits in balance may be related to impaired muscle strength, range of motion, abnormal tone, abnormal postural reactions and sensory deficits. These impairments can result in increased risk for falls, impaired performance of activities of daily living, and impaired movement (Hun Jang and Lee, 2015). Related to sensory deficits specifically, balance disorder in stroke may be caused by decreased central integration of sensory cues including somatosensory, visual and vestibular input.

In a healthy human, somatosensory information is used to maintain static standing. Somatosensory information includes proprioceptive input from the joints and muscles. With increased movement or external perturbation, the visual system or vestibular system will be used to maintain balance. If information from one sensory system is inaccurate or impaired, information from another system will be integrated to maintain balance. Depending on the location of the stroke, the somatosensory system can be severely impaired, resulting in the need for increased use of visual and vestibular cues (Hun Jang and Lee, 2015).

Determining how to modify and progress neurorehabilitation is important to promote independence and quality of life in individuals with multiple impairments of their sensory systems post-stroke. Typical intervention for decreased proprioceptive awareness and sensation includes compensatory visual fixation, increasing use of visual and vestibular input, and augmenting training with external visual and auditory cues (Harrison et al., 2019). However, there is limited research addressing intervention for patients who have deficits in use of both somatosensory and visual input who cannot increase use of visual cues for balance or integrate visual external cues. This specific case will address the treatment interventions used to improve

function in a patient who is legally blind and experienced bilateral thalamic stroke resulting in impaired proprioceptive and kinesthetic awareness, with resultant impairment in functional mobility and balance.

CASE DESCRIPTION

This is a case describing a 23-year-old male who presented to outpatient neurorehabilitation to address multiple impairments following an infarction that affected both the left and right thalamus. This stroke resulted following surgical resection of a recurring optic nerve meningioma. Prior to the surgical intervention, the patient presented with a complete visual impairment; he was born with complete absence of vision in his left eye at birth, and demonstrated progressive and eventual complete loss of vision in his right eye at age 12. He also presented with a history of multiple intracranial fusiform aneurysms and a surgical history of optic nerve meningioma resection. The reason for decline and eventual loss of vision in his right eye is unknown. In addition to the bilateral thalamic stroke, the patient developed hydrocephalus following surgery resulting in placement of a ventral parietal shunt (VP shunt).

Once medically stable, the patient received acute rehabilitation followed by two months of inpatient rehabilitation. Prior to initiating his outpatient rehabilitation plan of care, the patient demonstrated multiple shunt malfunctions, resulting in severe dizziness and nausea with movement. Five months post-surgery, he transferred to outpatient rehabilitation.

Examination

At his initial outpatient rehabilitation evaluation, the patient presented in a manual tilt-in-space wheelchair accompanied by his mother. He demonstrated absent joint position sense and kinesthesia of bilateral upper and lower extremities, absent light touch, pain and temperature sensation, and absent touch localization of bilateral upper and lower extremities. In addition, he

demonstrated flexor spasticity of his upper and lower extremities, and 4/5 manual muscle strength of bilateral lower extremities. He demonstrated significantly limited range of motion in his bilateral shoulders, and limited strength against gravity in his bilateral upper extremities. A formal manual muscle assessment of his upper extremities was not completed at initial evaluation. He also presented with thalamic pain syndrome with significant report of pain in his left upper extremity.

During his initial evaluation, he presented with dizziness and nausea and was unable to tolerate a transfer out of his wheelchair. Reportedly, he required total assistance for all bed mobility, seated balance, and transfers. He was nonambulatory. Per report, he also required maximum assistance for feeding and dressing and total assistance for toileting. In supported sitting in his wheelchair he demonstrated a resting cervical posture of left 30 degrees side bending and right 45 degrees rotation. He was cognitively intact.

The patient was unable to maintain static seated balance without wheelchair support, and therefore earned a zero out of 56 on the Function in Sitting Test (FIST). Due to poor activity tolerance and inability to tolerate a transfer out of his wheelchair during his initial visit, no other standardized outcome measures were performed. The patient's personal rehabilitation goals were to achieve prior level of independence; this included living independently and negotiating the community with the use of a probing cane or service dog. The patient also was the owner of a non-profit company. His goal was to return to managing the company and travelling as needed.

Evaluation, Diagnosis, Prognosis

At initial evaluation, the patient's most limiting impairments included absent superficial and deep sensory awareness of his bilateral upper and lower extremities, absent vision, significant dizziness, and significant complaint of pain. It was determined that these impairments, impacting

central integration of sensory input, were contributing factors to impaired seated balance, lack of tolerance of movement, and current requirement of total assistance for all mobility.

A referral to his neurologist was recommended to manage his shunt settings and to determine appropriate pain management. Due to limited tolerance of therapy, therapists recommended a frequency of maximum two times per week, with the intention to increase frequency should the patient demonstrate improved tolerance of services. Based on the initial evaluation, it was determined that the most appropriate initial interventions should include maximizing tolerance to upright sitting and perceptual awareness of midline by maximizing augmented feedback using predominantly auditory cues to compensate for loss of proprioceptive and absent visual feedback (Harrison et al., 2019). In addition, core strengthening activities and increased weightbearing through upper and lower extremities was expected to maximize sensory input, so as to improve kinesthetic awareness (Weinberg et al., 1988).

Intervention

The patient began outpatient physical therapy five months post-surgery. During the first three weeks of rehabilitation, the patient reported significant dizziness and nausea in supported sitting in his wheelchair. During some initial treatment sessions, the patient was unable to tolerate any motion, resulting in shortened or cancelled sessions. However, intervention was still attempted. After one month of physical therapy, the patient's VP shunt was adjusted by the physician, resulting in improved tolerance to mobility. At this time, the patient began tolerating skilled physical and occupational intervention three days per week for sixty minutes each discipline.

Months One to Three

Initial interventions were determined based on current literature related to stroke rehabilitation, however were modified to include extensive augmented auditory feedback secondary to the

patient's visual impairment (Harrison et al., 2019). Initial rehabilitation focused on improving tolerance to unsupported short sitting and improving postural awareness and perception of midline. Short sitting tasks included maintaining short sitting for as long as tolerated and with progressively decreased upper extremity support. Initially, the patient was provided constant external feedback via verbal cues for trunk positioning due to lack of visual feedback for correction; however, these verbal cues were faded with increased stability. To improve postural awareness in conjunction with verbal cues, proprioceptive input was provided through the use of a weighted vest (Choi and Kang, 2017). In one session, the patient improved his independent seated balance time from two minutes to six minutes with the addition of the weighted vest. Weights were distributed equally throughout the vest so as to provide proprioceptive input and awareness of center of mass in the seated position. Subsequently, this method was used again in the standing position to improve standing balance and postural awareness for completion of standing tasks.

Likewise, the patient performed various abdominal strengthening tasks and weightbearing activities to increase proprioceptive awareness; these tasks included weight bearing through upper extremities and lower extremities in supine, side lying and seated positions for most of the session; the patient performed ten repetitions of each task minimum each session. Manual cues were provided including joint approximation at both lower and upper extremities.

As the patient tolerated increased movement, repetitive functional task training was initiated (Guan et al., 2017). For a minimum of twenty minutes each session, he was instructed in bed mobility and transfer training. He performed repeated rolling supine to side lying and as tolerated, supine to prone. Assistance and verbal cues were faded with repetition. He was also

instructed in repeated stand pivot transfers with assistance. Due to absent vision, he required significant external feedback via verbal cues for sequencing of the task.

Lastly, electrical stimulation was used to improve postural awareness and to increase awareness of and use of somatosensory input (Sharififar, Shuster, & Bishop, 2018). Transelectrical stimulation (TENS) for augmented sensory feedback during functional tasks was applied at his erector spinae and abdominal musculature to improve postural control; in addition, TENS was applied to his bilateral anterior tibialis to increase proprioceptive awareness at his distal extremities during upright standing balance activities for 15 minutes during a session. Similarly, he used the Functional Electrical stimulation cycle at the end of every session with added electrical stimulation to his bilateral quadriceps, hamstrings and anterior tibialis. Placement of electrodes was determined based on ease of access, use of large muscle groups and for increased sensory awareness at these locations (Sharififar, Shuster, & Bishop, 2018). To maximize aerobic exercise to promote increased exercise tolerance and sensorimotor function, the patient cycled for three to five miles at the end of every session (Gordon et al., 2004).

Months Three to Eight

With increased time, the patient began to report improved light touch sensation in bilateral lower extremities as well as his right upper extremity. He also demonstrated improved proprioceptive awareness of these extremities. He began tolerating upright standing for increased time with right upper extremity support. To maximize weightbearing through his lower extremities, he performed static standing in a standing frame for a three-week protocol, increasing standing tolerance to 30 minutes.

In order to provide additional proprioceptive input to progress his postural stability and positioning in relation to the environment, a hemi walker progressing to a straight cane was used

in his right upper extremity. Verbal cues were provided to improve his awareness of his trunk positioning based on perception of where his trunk was in relation to the cane. With repeated performance of this task and fading verbal cues, he progressed to static standing without assistance. Standing tolerance was increased from two minutes to 10 minutes, with added dynamic anticipatory activities as tolerated to promote increased postural control.

Gait training was initiated once the patient tolerated upright standing with supervision. Initially, the patient performed gait training on the treadmill with use of the LiteGait harness without body weight support. He initially ambulated with right upper extremity support and moderate assistance of two therapists with assistance for lower extremity progression and trunk stabilization. With increased comfort and stability, speed and time were increased and assistance was decreased. He was provided with ongoing verbal cues for positioning on the treadmill secondary to impaired vision.

He then progressed to ambulation over ground on a firm surface with minimal assistance. While he demonstrated improved postural control in standing, his head posture remained unchanged. Due to inaccurate input from his head posture, he required constant verbal cue for guidance to minimize path deviation related to impaired perceptual orientation. As tolerated, his distance and speed was progressed over ground. Because he demonstrated improved sensation and mobility of his right upper extremity, gait intervention progressed to ambulation over ground with use of his probing cane. He was instructed to use the cane for auditory cue input as he did prior to surgery, but to also use the cane as a light touch cue. He was provided intermittent assistance as needed, but was cued to determine trunk and lower extremity positioning based on auditory and kinesthetic input from the probing cane (Boonsinsukh, Panichareon, and Phansuwan-Pujito, 2009).

Fluidotherapy

Fluidotherapy is a modality often used to improve quality of life in patients with complex regional pain syndrome. It is a dry modality that superficially transfers heat convectively and is used to treat patients with pain, muscle spasm, and decreased range of motion. Likewise, it has been used to provide desensitization in distal limbs. The Cellex, or synthetic cellulose particles, are circulated by a warm air current, which creates a fluid-like form. While it provides a similar sensation to a heated whirlpool, patients can tolerate higher temperatures with the fluidotherapy compared to typical modalities (Kelly et al., 2005).

Due to constant report of debilitating pain in his left upper extremity, a trial of fluidotherapy was introduced. The patient's left hand and forearm were inserted into the machine for varying ten to twenty minutes per treatment session. Following treatment, the occupational therapists performed passive range of motion to his left hand, wrist, and forearm to maximize effects of the modality. This intervention provided the patient with immediate pain relief. Ultimately, the patient chose to purchase a fluidotherapy machine to be used in his home environment, which he began using daily.

Insert table 1 about here

OUTCOMES

Because the patient demonstrated poor tolerance of assessment during the initial evaluation, some functional measures and outcome measures were not assessed until later months in his rehabilitation. His FIST score at initial evaluation was zero; this score improved to 32 out of 56 at three months. At reassessment at four months, his FIST score improved to 45 out of 56. At six months, the patient was initially assessed on the Berg Balance Scale, scoring 8 out of 56. See Table 2.

After eight months of outpatient neurorehabilitation, the patient transitioned to an intensive neurorehabilitation day program. At discharge from outpatient rehabilitation, he was able to perform all bed mobility with supervision, perform sit to stand and stand pivot transfers with stand by assistance, and required varying stand by assistance and minimal assistance for ambulation with probing cane. Likewise, he required minimal assistance for ascending and descending stairs using the right handrail. In addition, at discharge the patient's Berg Balance Score improved from 8/56 to 31/56. He demonstrated intact bilateral lower extremity light touch and touch localization, intact position sense at his bilateral ankles, and impaired light touch sensation at his right upper extremity.

Insert Table 2 about here

DISCUSSION

Impaired postural awareness and trunk control following stroke is common and typically results in significant functional impairment. Although there is some research regarding physical therapy intervention for patients with impaired proprioception following stroke, there is limited research addressing rehabilitation for patients with impaired proprioception and absent visual input. This case **report** demonstrates the implications of neurorehabilitation in treating patients with multiple sensory impairments.

First, fluidotherapy was a contributing factor to this patient's rehabilitation success. Initially, the patient reported significant pain in his left upper extremity, which limited his range of motion and weightbearing tolerance; this ultimately was a significant limiting factor to his participation in rehabilitation. Addition of fluidotherapy to his plan of care significantly decreased his pain and ultimately improved his participation. Kelly et al. (2005) found the use of fluidotherapy elevated superficial skin temperature. The treatment, which combines the effects of tactile

stimulation and heat, has been used to provide analgesic effects at the applied site and distal to the application. Likewise, Ozcan et al. (2019) found the use of fluidotherapy as an intervention for individuals with complex regional pain syndrome post stroke to be effective in reducing pain and edema and maximizing upper extremity function. This patient and his caregiver purchased a fluidotherapy for use in the home. With daily use, he demonstrated minimal to absent upper extremity pain, allowing for increased range of motion tolerance and increased tolerance for therapeutic intervention. Addressing barriers that limit participation in therapy is the first step in being able to provide optimal care for patients with multisensory impairments after stroke. This patient presented with impaired proprioception and absent vision. Once his pain was managed, it is assumed that his progress in rehabilitation was due to the upregulation of remaining sensory/proprioceptive cues, increased weighting of and reliance on vestibular cues, as well as integration of external auditory cues to provide augmented feedback regarding patient performance to optimize motor learning.

Vidoni and Boyd (2009) found that “the magnitude of learning related change was directly related to the preservation of proprioception.” Thus, this patient case describes interventions that improved proprioception, likely through techniques that helped to facilitate motor learning based on the preservation of proprioception he experienced after his stroke.

Following an extensive literature review, Schabrun and Hiller (2009) determined that electrical stimulation has moderate support for use in sensory training following stroke. Although optimal parameters have yet to be determined, electrical stimulation for short durations has been found to improve hand dexterity following stroke. In a study by Chan, Ng, and Ng (2015), transelectrical stimulation was used in combination with task related trunk training (TTRT). Components of the TTRT included pelvic bridging, sitting up, trunk range of motion and reaching. TENs was

applied simultaneously to the latissimus dorsi and external abdominus obliquus. Performing this intervention thirty minutes for five days per week for six weeks resulted in improvements in trunk control during seated balance.

When TENS was applied to our patient with the same parameters, the patient demonstrated improved upright posture. Prior to the addition of the electrical stimulation, this patient required minimal assistance to maintain static standing with feet together. With the addition of electrical stimulation, he immediately progressed to standing with feet together with supervision with minimal postural sway. He continued to demonstrate improved postural stability with forward and backward walking with the addition of electrical stimulation to his core musculature. Thus, the use of TENS for this patient theoretically contributed to increase in sensory feedback and improved postural stability.

The vestibular system detects the motion of the head in space by encoding self-motion information. Some of the vestibular nuclei neurons are designated as vestibular only; rather than projecting to oculomotor structures, these-specific neurons project to the spinal cord and mediate the vestibular spinal reflexes. They are interconnected with the nodulus of the cerebellum, which then project to the thalamus and cortex. Therefore, with intact vestibular neurons, the patient is theoretically able to use vestibular cues in the absence of visual cues to maintain postural equilibrium and spatial orientation through higher order vestibular processing (Cullen, 2012).

While a substantial amount of research has been completed regarding visuo-vestibular interactions, little is known regarding the training of vestibular cues in visually impaired individuals. However, it has been determined that the vestibular system is the predominant contributor to self-motion perception in individuals lacking visual input (Moser, Grabherr,

Hartmann, and Mast, 2015) and compensation includes increased vestibular sensitivity or upregulation and integration of signals that stimulate the remaining sensory organs.

Proprioceptive inputs reach the vestibular nuclei through dorsal root axons and through second order neurons. Likewise, cerebellar and cortical areas receiving somatosensory inputs project to the vestibular nuclei. These tracts make it possible to sense body motion. It is within the rostral fastigial nucleus of the vestibular cerebellum that proprioceptive and vestibular cues are integrated, resulting in accurate control of posture, balance, and self-motion perception (Cullen, 2012).

In addition to the upregulation and maximization of use of proprioceptive and vestibular cues, it is suspected that augmented external feedback using auditory cues provide neuroplasticity to the vestibulocerebellar circuitry to improve motor learning and subsequently overall patient function and independence. Without visual feedback, the therapist must provide augmented auditory and sensory input in place of visual cues. This input included the extensive use of verbal and manual cues and TENS. Likewise, the use of FES, extensive extremity weightbearing, and the use of a weighted vest provided proprioceptive feedback in conjunction with repetitive functional training. The combination of these cues resulted in maximized functional mobility.

It is also likely that augmented auditory cues promoted motor learning through cerebellar pathways. The cerebellum helps to maintain balance and posture by adapting sensory input from vestibular, auditory, visual, and proprioceptive receptors. It is integral in motor learning through an error feedback model and facilitates procedural learning (Shumway-Cook and Woolacott, 2017). Because this patient had long-standing absent visual input, information from auditory, vestibular, and proprioceptive afferents provided needed input for error-based motor learning

through improved functioning of vestibular and vestibulocerebellar pathways to promote optimal rehabilitation outcomes in this patient.

The patient reported in this case report was young, highly motivated, and cognitively intact.

Although every patient's motivation, cognition and communication will vary, these principles can be applied to varying degrees for patients with multisensory integration and postural deficits following injury to the brain. Likewise, the successfulness of this intervention further demonstrates that the ability to recover postural orientation during functional mobility is not directly related to the preservation of vision; neurorehabilitation to improve postural control is beneficial for those with varying physical presentation and severity of visual deficits. This case report illustrates the unique therapy considerations that need to be addressed by the therapist to optimize outcomes in therapy. The data from this case aims to illustrate the effectiveness of both upregulating input from remaining sensory systems and using external auditory cues for improving balance function and increasing independence in a severely impaired patient after a bilateral thalamic stroke.

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