A Hybrid Home Rehabilitation Program for Moyamoya Disease to Facilitate Return to Work and Functional Independence: A Case Report from a Developing Country during the COVID-19 Pandemic

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ABSTRACT

A 35-year-old female, right-handed, non-hypertensive, and non-diabetic Filipino presented with decreased verbal output and weakness of the right upper and lower extremities during the peak of the COVID-19 pandemic. Cerebral angiography showed bilateral steno-occlusive disease, which was consistent with Moyamoya disease. She underwent inpatient rehabilitation consisting of physical, occupational, and speech therapy. Rehabilitation post-hospital discharge was continued using a hybrid rehabilitation approach with a mixed in-person home rehabilitation and remote telerehabilitation. The hybrid approach helped ensure continuity of rehabilitation care, minimize travel and exposure to the hospital or community amid the COVID-19 risks, and reduce costs, without entirely losing the benefits that could only be obtained from hands-on therapeutic evaluation and treatment. After six months of hybrid rehabilitation, the patient was able to return to work and regain functional independence.

Keywords: case report. telerehabilitation, hybrid rehabilitation, Moyamoya, COVID-19

INTRODUCTION

Moyamoya disease is a rare congenital or acquired cerebrovascular disorder characterized by progressive stenosis of the distal internal carotid arteries and the Circle of Willis.[1,2] Due to the resultant ischemia, there is massive compensatory angiogenesis appearing as "cloud-like" or "smoke-like" mesh of tiny blood vessels on cerebral angiography.[1] It affects females twice as much as males with prevalence ranging from 1.61 to 16.1 per 100,000 from the years 1995 to 2011 in East Asian Countries.[2] Aside from environmental factors, such as head and neck radiation or infection, genetics may play a role, albeit still suggestive, in the development of Moyamoya disease.[3] Clinically, the disease may present with varying neurologic deficits.[2] Rehabilitation oriented towards neurologic and functional deficits has been previously documented.[4] There is limited data, however, on the specific rehabilitation interventions and corresponding functional outcomes of patients with Moyamoya disease. This case report highlights the feasibility and value of a hybrid rehabilitation program, consisting of in-person and remote therapy sessions, for a patient with Moyamoya disease amid limited resources in a developing country and COVID-19 risks.

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CASE PRESENTATION

A 35-year-old female, right-handed, non-hypertensive, and non-diabetic Filipino presented with decreased verbal output and weakness of the right upper and lower extremities. She is a previous smoker and an occasional alcoholic beverage drinker. She works as an Information Technology (IT) professional for a global consulting company. She lives with her family in a province 98 kilometers away from our institution. She was initially received in the emergency room of a different hospital with a Glasgow Coma Scale of 15 and slurring of speech, and was assessed to have Transient Ischemic Attack. During admission, she developed weakness of the right upper and lower extremities and right-sided facial asymmetry. Cranial magnetic resonance imaging (MRI) on the 7th day post-ictus revealed an acute infarct on the left lenticulo-capsular region, corona radiata and centrum semiovale. Subsequent MRI on the 10th day post-ictus showed bilateral infarct on the aforementioned areas. No neurosurgical management was required. On the 13th day post-ictus, she was transferred to our institution, wherein medical management was continued. On the 16th day post-ictus, she underwent cerebral angiography confirming the diagnosis of Moyamoya disease.

The patient was referred to the Physical Medicine and Rehabilitation service on the 14th day post-ictus. On examination, she presented with dysthymic mood and flat affect. She had expressive aphasia with intact comprehension, mumbled incomprehensible words, but answered questions appropriately through writing. She was alert, oriented to person, place, and time, had intact recent and remote memory and calculation skills. Cranial nerve examination was pertinent for the facial nerve, which presented as minimal right-sided central facial palsy. She was neither able to open her mouth nor protrude the tongue voluntarily but had minimal spontaneous oromotor movements, reflexive swallows, and fair laryngeal elevation upon stimulation. She had minimal drift of the right upper extremity. No gross deficit in coordination was noted, but the patient had slower movements on the right extremities with fair performance of fine motor skills on the right hand. She was assisted in all activities of daily living.

DIAGNOSTICS

Cranial MRI and angiography revealed acute infarcts in the right semioval center, corona radiata, and basal ganglia with temporal evolution of the left corona radiata, semioval center, and basal ganglia infarcts, and an attenuated M1 segment of both middle cerebral arteries and C7 segment of the left internal carotid artery. Cerebral angiography revealed a bilateral steno-occlusive disease of the terminal internal carotid, proximal middle cerebral and proximal anterior cerebral arteries, with presence of fine, irregular vessels and prominent collateral supply from the posterior communicating artery and leptomeningeal vessels, which are consistent with Moyamoya disease.

APPROACHES TO MANAGEMENT

Once the patient became medically stable, inpatient physical therapy was started to achieve the short-term goal of safe and early mobilization. The long-term goals for this patient, including safe oral feeding, functional independence, and return to work, could be achieved post-discharge through in-person outpatient therapy. However, given the COVID-19 risks and the distance barrier, telerehabilitation was offered particularly to continue occupational and speech therapy at home. Eventually the lockdown eased, and in-person home rehabilitation was done in between continued telerehabilitation sessions.

COURSE OF REHABILITATION MANAGEMENT

The patient underwent inpatient physical, occupational, and speech therapy. The physical therapy program included the following interventions: limb mobility and strength training, standing balance drills, ambulation training, and endurance exercise. Occupational therapy focused on retraining for activities of daily living, and speech and language pathology service facilitated speech and swallowing exercises. Due to persistently poor oral intake, she underwent percutaneous endoscopic gastrostomy (PEG). On discharge, she could ambulate for a short distance with standby assist and tolerated pureed food consistency during swallowing training. She was advised to undergo telerehabilitation at home until the COVID-19 protocols allowed in-person home therapy. Nonetheless, she opted to continue with telerehabilitation sessions, supplemented by in-person home therapy as she and her family perceived telerehabilitation to be more convenient, less costly, more time-flexible, and less COVID-19 risky. The cost for telerehabilitation and in-person sessions were the same at Php 1,000.00 per session. However, the indirect costs of telerehabilitation can be much lesser by reducing transportation and meal expenses, and minimizing work absences or loss of productivity.[5]

Figure 1 shows the rehabilitation goals for the patient. Tables 1 and 2 summarize the different therapeutic interventions that were employed via the hybrid home rehabilitation approach. Strict aspiration precautions during swallowing training and monitoring for progression of neurologic deficits were observed.

Occupational therapy was done in-person at home twice a week and via telerehabilitation once a week. Dysphagia management was done in coordination with the speech and language pathologist. Swallowing training started with pureed food and gradually progressed to regular food with incorporation of compensatory and restorative maneuvers. These maneuvers included the following:

1. Oral Peripheral Mechanism (OPM) exercises – These aim to improve strength, control, and coordination of the oral muscles used for swallowing. These may include active mouth opening exercises, tongue protrusion, and mobility exercises.
2. Masako maneuver – Sticking the tongue out between the teeth followed by swallowing aims to improve the strength of the pharyngeal constrictors and improve tongue base movement.[6]
3. Chin tuck against resistance – This exercise is done by applying resistance below the chin while the patient attempts to push the chin downwards. This aims to strengthen the suprahyoid muscles.[6]
4. Effortful swallowing – This is a maneuver intended to increase pressure on a bolus by swallowing and pushing it with sufficient force to facilitate clearance.

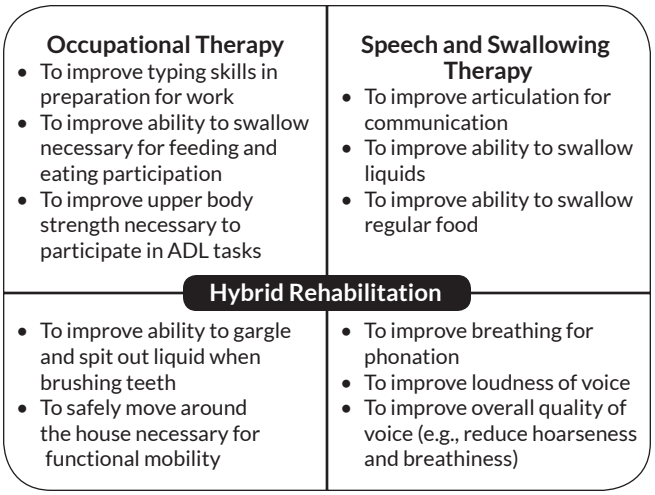

Figure 1. Rehabilitation goals for the patient.

Table 1. In-person and Virtual Rehabilitation Management

Occupational Therapy	
In-person interventions	***Interventions done virtually***
• Upper extremity ROM and strength training using OT equipment • Fine motor exercises • Eye-hand, arm-hand coordination drills • Dexterity exercises • Actual typing and accuracy drills training	• Use of household items for upper extremity ROM and strength training, fine motor exercises, eye-hand, arm-hand coordination, and dexterity drills • Web-based software that trains typing skills • Patient education on work ergonomics
• OPM and facial exercises • Indirect and direct swallowing exercises • Patient and caregiver education on use of compensatory strategies for safe swallow	• OPM and facial exercises • Indirect swallowing exercises
• Upper body strength and ROM exercises • Fine motor skill and coordination training • Simulation activities for bathing • Occupation-based training of ADL skills	• Upper body strength and ROM exercises using household items • Simulation activities for ADL • Task modification and adaptive techniques
• OPM and facial exercises • Occupation-based training of skill • Upper and lower body strength and ROM exercises • Patient and caregiver transfer training	• OPM and facial exercises • Environmental assessment and modification

ROM: range of motion; OT: occupational therapy; OPM: oral peripheral mechanism exercises; ADL: activities of daily living

Table 2. In-person and Virtual Rehabilitation Management

Speech and Swallowing Therapy	
In-person interventions	***Interventions done virtually***
	• Articulation drills, including over-articulation during videocalls
• Thermotactile stimulation • Masako maneuver and chin tuck against resistance exercises • OPM exercises • Effortful swallows • Progression of food consistencies	• OPM exercises
	• Diaphragmatic breathing exercises • Voice projection techniques • Biofeedback and verbal feedback
• Resonant voice therapy • Confidential voice therapy • Hum and nasal consonants • Laryngeal adduction exercise • Easy-onset phonation drills	• Resonant voice therapy • Confidential voice therapy • Hum and nasal consonants • Laryngeal adduction exercise • Easy-onset phonation drills

OPM: oral peripheral mechanism exercises

Training for activities of daily living and task-specific activities were done to improve independence. By the 5th month post-ictus, telerehabilitation sessions were more frequent than in-person home therapy, and activities focused on further improving typing speed and accuracy administered via online websites, such as Typing.com (https://www.typing.com/), Typing Academy website (https://www.typing.academy/typing-tutor/lessons), and Read & Spell website (https://www.readandspell.com/us/stroke-recovery). The patient graduated from occupational therapy when she was able to return to work by the 6th month post-ictus.

Speech and swallowing therapy sessions were continued at home twice a week for in-person sessions and once a week remotely via telerehabilitation. Voice training was included due to persistent hoarseness. Voice training exercises included the following:

1. Voice projection techniques – These techniques aim to increase the loudness of voice in a relaxed, easy manner without straining.[7]
2. Biofeedback and verbal feedback – This technique implores the use of imagery training in addition to physical sensation to achieve a desired vocal tract posture.[8]
3. Resonant voice therapy – The goal of these exercises is to increase voice loudness without effort and to use appropriate speaking pitch with relative ease on phonation through feeling and hearing.[8,9]
4. Confidential voice therapy – It is coined as such since the voice quality achieved in this therapy is the voice typically used to discuss confidential matters. The goal of this therapy is to reduce vocal fold contact, thereby decreasing the force of vocal fold collision and reducing hyper-functional behavior and muscle tensions.[9]
5. Hum and nasal consonants – Humming exercises target ease of phonation and appropriate speaking speech by emphasizing the sensation of resonance in the oronasal region with minimal effort and tensions at the laryngeal and neck regions.
6. Laryngeal adduction exercise – These are exercises for strengthening the laryngeal musculature to improve voice production.[10]
7. Easy-onset phonation drills – The goal of these drills is to reduce hyper-functional phonatory behaviors. The patient may be instructed to allow release of airflow before gently initiating sound.[9]

A fiberoptic endoscopic evaluation of swallowing (FEES) was done around six months post-ictus and showed no penetration nor aspiration with thin and thick fluids, pureed food, and food with mixed consistencies. The PEG tube was removed thereafter, and the patient safely returned to complete oral feeding.

Functional independence, assessed via the Functional Independence Measure (FIM), significantly improved from requiring maximal assistance (35/126) on discharge to minimal assistance (123/126) at 4 months thereafter. The patient's cognitive status was assessed via the Mini-Mental State Examination (MMSE) with noted improvement from having mild cognitive impairment during admission to no impairments after 3 weeks from discharge. The patient's typing speed, analyzed via online assessment tools, improved from 28.06 words-per-minute (wpm) with an accuracy of 80.37% by 4 months post-ictus, to 53.86 wpm with an accuracy of 90-98% by 5 months post-ictus. Speech was monitored remotely and objectively through the Praat™ voice analysis software which could aid in the tele-evaluation of a neurologic patient.[4] After around 4 months post-ictus, significant improvements were noted in terms of jitter, shimmer, and pitch.

DISCUSSION

A hybrid rehabilitation approach provides patients with additional options so that functional gains achieved from inpatient rehabilitation could be continued and improved at the comfort and safety of their homes. The patient and her family experienced the advantages of telerehabilitation in overcoming in-person rehabilitation barriers of distance, time, costs, and even COVID-19 risks. Nonetheless, having in-person occupational and speech therapy sessions allowed the patient to have a continued, supervised, safe oral feeding retraining program, which was found to be among the apprehensions of Filipino physiatrists about telerehabilitation.[11] Meanwhile, indirect swallowing exercises, oral peripheral mechanism techniques, facial self-massage, and other oromotor drills done through telerehabilitation could safely supplement direct swallowing trials done in-person.

The aim of telerehabilitation is to provide remote rehabilitation care using locally available and low-cost technologies.[12] Despite the global COVID-19 crisis, telerehabilitation may improve rehabilitation access to persons with disability.[13] A hybrid rehabilitation approach both addresses the clinical limitations of telerehabilitation and takes advantage of in-person training. This approach may foster patient participation and empowerment, varied and interactive therapy modes, and healthcare-related financial savings.

To our knowledge, no studies on hybrid telerehabilitation for Moyamoya disease have been published. This report has documented how telerehabilitation can be a useful approach to providing home-based rehabilitation among stroke patients during the COVID-19 pandemic. This provides a balance in continuing functional retraining without the risk of contracting COVID-19.[14,15] A randomized controlled trial on 124 stroke patients with upper extremity deficits showed no significant difference in arm motor function between home-based telerehabilitation and traditional in-clinic rehabilitation after 36 sessions.[16]

Through the home hybrid rehabilitation program, our patient eventually had improved functional independence and was able to orally eat safely and independently with neither

reported adverse event nor technical problems. The patient and family were cooperative and adherent to the prescribed hybrid rehabilitation program. Currently, the patient is back to work as an IT professional and successfully reintegrated into the society.

CONCLUSION

Home hybrid rehabilitation is feasible, safe, and effective for our patient with Moyamoya disease amid the COVID-19 pandemic. A strategic combination of in-person interspersed with virtual therapy sessions may improve compliance, interest, participation, and functional outcomes.

Ethical Statement

The patient's voluntary informed consent was obtained, and the CaRe guidelines were followed in the writing of the manuscript.

Statement of Authorship

ESN and CFDL contributed in the conceptualization of work, acquisition and analysis of data, drafting and revising of manuscript, and final approval of the version to be published. MFB and DAPI contributed in the acquisition and analysis of data, and drafting and revising of manuscript.

Author Disclosure

The authors had no conflict of interest to disclose.

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